



**University of
Leicester**

Decision-Making about Congenital Anomalies

**How do women and their partners make
the decision to continue or terminate a
pregnancy following suspicion or diagnosis
of a severe congenital anomaly?**

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HOW DO WOMEN AND THEIR PARTNERS MAKE THE DECISION TO CONTINUE OR TERMINATE A PREGNANCY FOLLOWING SUSPICION OR DIAGNOSIS OF A SEVERE CONGENITAL ANOMALY?

Background : Around 2-3% of pregnancies are affected by a congenital anomaly. However, anomalies account for around 30% of neonatal mortality in the UK. Whilst the incidence and rates of detection are similar across socioeconomic groups, rates of termination following diagnosis of a severe anomaly are lower in more deprived areas. The reasons for this are unclear and parental decision-making following suspicion or diagnosis of a congenital anomaly is largely unexplored. **Aims:** To explore the decision-making processes following diagnosis of a congenital anomaly and offer insight into how variations in termination rates arise. **Methods:** Data from interviews with parents-to-be and clinicians, and recorded consultations were collated. Analysis was undertaken using a constant comparative based approach. **Findings:** Following diagnosis of a severe congenital anomaly, parents-to-be face the devastating decision of whether to continue or terminate the affected pregnancy. Four typologies of decision-making were identified. These were entitled: Consequential, where parents sought to 'rationally' evaluate the best outcome for themselves and the unborn, Absolute, where fundamental beliefs pre-determined the decision taken, Delay/Avoid, where no active decision was made, and parents therefore continued with the pregnancy, and Assess/Reassess. This fourth typology subsequently sub-divided into two groups, Choice Removed, where indecisive parents were 'pushed' by clinicians into terminating the pregnancy, and Choice Disturbed, where the breakdown of the parent-clinician relationship resulted in parents disengaging with the clinical environment thus continuing with the affected pregnancy. Each type of decision-making resulted in differing tensions with clinicians who sought enactment of an 'ideal' decision-making process. **Conclusion:** This study provides a valuable insight into the lived experiences of parents. With this comes a greater awareness of the variations in the pathways and processes followed. The recommendations contribute to the understanding of those who determine policy and those who practice within the field of fetal medicine.

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DEDICATION

To my family. Thank you

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ABBREVIATIONS

ARC	Antenatal Results and Choices
BINOCAR	British Isles Network of Congenital Anomaly Registers
BPAS	British Pregnancy Advisory Service
CDH	Congenital Diaphragmatic Hernia
CNS	Central Nervous System
CVS	Chorionic Villus Sampling
DGH	District General Hospital
DS	Down's Syndrome
FASP	Fetal Anomaly Screening Programme
FISH	Fluorescent in-situ hybridisation
HERG	Health Experiences Research Group
NCAS	National Clinical Assessment Service
NHS	National Health Service
ONS	Office of National Statistics
OSOP	One Sheet of Paper
PPI	Public and Patient Involvement
RCOG	Royal College of Obstetrics and Gynaecology
SANDS	Stillbirth and Neonatal Death Charity
SES	Socioeconomic Status
UK NSC	United Kingdom National Screening Centre

1 BACKGROUND

A congenital anomaly is any defect present at a baby's delivery but likely to originate before birth, and includes any form of structural, chromosomal, genetic and biochemical defect and malformation (BINOCAR Working Group 2013). Whilst primary interventions, such as pre-conception folic acid, have played a significant role in reducing the in-utero prevalence of many anomalies (Botto, Correa 2003, Czeizel 2009), around 2 to 3% of pregnancies in high income countries will result in a fetus with a congenital anomaly (Askelsdottir, Conroy *et al.* 2008). The need for further preventative measures remains, with the current situation, in terms of secondary intervention, relying on antenatal detection and diagnosis, which has resulted in couples being offered the option to terminate an affected pregnancy. In parts of the United Kingdom (UK) this equates to around 70% of affected pregnancies (Budd, Draper *et al.* 2015), with 2,732 such terminations performed in 2013 in England and Wales (Department of Health 2014). Despite this, congenital anomalies remain a major cause of neonatal and infant mortality, accounting for around 30% of these deaths across the UK (Oakley, Maconochie *et al.* 2009).

Further examination of these deaths demonstrates a higher risk of mortality for neonates and infants affected by congenital anomalies in more deprived areas (Neasham, Dolk *et al.* 2001, Oakley, Maconochie *et al.* 2009, Smith, Manktelow *et al.* 2010, Olesen, Thrane *et al.* 2009). This variation exists despite an equal distribution of in-utero prevalence of severe congenital anomalies across all levels of deprivation (having adjusted for differences in maternal age) (Smith, Budd *et al.* 2011), and with comparable rates of detection of anomalies (Rowe, Garcia 2003). Much of this variation may therefore be explained by the difference in rates of termination for congenital anomalies, with fewer terminations performed for congenital anomalies in more deprived areas (Smith, Budd *et al.* 2011). However, the reasons for this are unclear, and parental decision-making following suspicion

or diagnosis of a congenital anomaly remains a largely unexplored area (Bijma, van der Heide *et al.* 2008, Pryde, Drugan *et al.* 1993, Shaffer, Caughey *et al.* 2006).

My thesis explores the experiences of parents as they navigate the decision-making process following suspicion or diagnosis of a severe congenital anomaly. The aim is to provide insight into how decisions are made, and subsequently gain an understanding of how the variation in termination rates arises. In turn, it will explore the possibility of identifying potentially modifiable factors, to ensure that future health policy and practice in this area best support individual parents and minimise socioeconomic inequalities in neonatal mortality.

1.1 THESIS OUTLINE

My thesis is divided into eight chapters. The first chapter provides the foundations for the thesis by explaining key aspects of a parent's journey and placing this in the context of the laws, policies and procedures that surround it. This is developed in the second chapter, where the literature around parental decision-making following suspicion or diagnosis of an anomaly is examined and the research question and aims of the study are set out. The third chapter provides a description of the methodology employed to address the research question. The tone of the thesis changes in the fourth chapter, where issues relating to the ethics of addressing this topic are explored, and the influence of my persona as a healthcare professional and researcher is considered. The sensitivity of the topic has remained in the forefront of my mind throughout the process, and has impacted significantly on the decisions made throughout. Therefore, a full chapter has been dedicated to recording my reflections on this. Chapters 5, 6 and 7 are a temporal representation of the findings from this research. In Chapter 5, the multi-layered contextual framework in which the decision-making process is enacted is analysed and presented. A number of themes pertaining to the decision-making process of the parents are described in Chapter 6. Examination of the spectrum of these responses enabled the decision-making processes employed by parents to be categorised. Comparisons across the categories highlighted instances of either similarity or difference, from which a conceptual model was constructed. This

model was subsequently reapplied to the data in order to highlight tensions arising from the parental enactment of the process and the idealisation of particular attributes of decision-making by clinicians. Chapter 7 provides an insight into the final stage of the decision-making process of parents who terminated their pregnancy, as they attempted to come to terms with and make sense of the decision they made. Chapter 8 concludes the thesis by providing a summary of the findings and their importance in determining future care provided for parents following suspicion or diagnosis of a severe congenital anomaly. This is subsequently developed into a number of recommendations for future practice, which represent improvements that can be made at each level of the multi-layered contextual framework.

Before embarking on this journey, the remainder of this chapter is designed to create a common understanding of terms. The history and laws governing congenital anomalies and termination are highlighted alongside the process itself. Subsequently the issue of stigma and its association with termination is examined. Due to the sensitive and emotive nature of the subject matter, great care has been taken to use language that reflects the reality as constructed by the participants. The use of language is a particularly powerful tool; therefore, the final section of this chapter provides an explanation for the use or avoidance of specific terms or phrases within this thesis.

1.2 SCREENING FOR CONGENITAL ANOMALIES IN THE UK

Thalidomide entered the German market in 1957 as a tranquilliser and sedative. An Australian obstetrician noticed that the drug alleviated morning sickness and it was soon marketed in 46 countries as a safe and effective over-the-counter drug for pregnant women. By 1961 a correlation between thalidomide and birth defects in babies was noticed, and soon after the drug was banned in most countries (Fintel, Samaras *et al.* 2009). The ghost of thalidomide resounds in many changes that were implemented internationally in relation to drug licensing and the surveillance of anomalies.

In 1964 the Office of National Statistics set up a National Congenital Anomaly System (NCAS) that collated the data on infants born with congenital anomalies. While reporting was voluntary, and it was well known for under reporting the true number of children diagnosed in England and Wales (Boyd, Armstrong *et al.* 2005), it did allow for surveillance of 'patterns' in reported incidents. In 1985 the first of a number of regional registers was established; however, without central funding coverage has remained patchy. This is currently being addressed through the creation of a national register led by Public Health England.

Information collated by the registers in England and Wales has reflected the development of the national screening programmes. In the late 1980's the association between reduced levels of alpha-fetoprotein and the risk of Trisomy 21 (also known as Down's Syndrome) was noted, and screening with these biochemical markers, in conjunction with an ultrasound, was rapidly implemented. Screening was not universal, however, and it was not until 2001 that the national Down's Syndrome Screening Programme was implemented by the UK National Screening Committee (UK NSC) (National Screening Committee 2013).

Antenatal screening has continued to develop with the introduction of the Fetal Anomaly Ultrasound Programme that amalgamated with the Down's Syndrome Screening Programme in 2007 to become the Fetal Anomaly Screening Programme (FASP) (UK National Screening Committee 2009). The standards for the screening programme were set in 2010, with the focus of the FASP being the antenatal identification of serious abnormalities that have a predicted detection rate of over 50% and are either a) incompatible with life or associated with significant morbidity or b) amenable to antenatal treatment, or may require immediate postnatal support (NHS Fetal Anomaly Screening Programme 2010). The anomalies included in the combined screening programme are presented in Table 1-1 overleaf.

Table 1-1 Description of congenital anomalies targeted through screening

Anomaly	Definition	Incidence	Outcome	Mode of Identification	Target identification rate
Anencephaly	Absence of the skull and brain – neural tube defect.	1/10,000 births	Death within a few days after birth	Booking scan or Anomaly scan	98%
Spina Bifida	Incomplete closure of the backbone and membranes around the spinal cord. Severity dependent on position.	1-2/1000	Variable from asymptomatic to severe	Soft markers on booking scan	90%
Severe Cardiac	Collective term applied to a number of cardiac anomalies. No consensus on what constitutes severe.	35/10,000 births	Variable, untreated may result in death	Anomaly scan	50%
Bilateral Renal Agenesis	Absence of kidneys, often genetic. Usually present with a deficiency of amniotic fluid.	1/10,000 births	Survive around 4 hours	Booking scan or Anomaly scan	84%
Lethal Skeletal Dysplasia Multiple types	Abnormal bone growth. As a result of abnormal rib growth, the chest and lungs do not fully develop.	0.95/10,000 births	Often stillborn or death soon after delivery	Anomaly scan	60%
Congenital Diaphragmatic Hernia (CDH)	Failure of the diaphragm to fuse properly during fetal development, allowing the abdominal organs to migrate up into the chest cavity.	4/1000 births	Mortality of 40-62%	Anomaly scan	60%
Exomphalos	Liver and bowel protrude through abdomen.	4/10,000	10-90% survival	Anomaly scan	80%

	Associated anomalies in up to 80% of cases.	births			
Gastroschisis	Defect in the anterior abdominal wall through which the abdominal contents freely protrude.	5/10,000 births	Survival rate over 90%	Anomaly scan	98%
Cleft lip/palate	Partial or complete clefting of the upper lip, with or without clefting of the alveolar ridge or the hard palate.	10/10,000 births	16% - structural abnormalities 7% - syndrome	Anomaly scan	Cannot reliably identify a cleft palate on U/S.
Trisomy 18	Also known as Edward's syndrome, it is a serious genetic condition.	3/10,000 births	50% do not survive past the first week	Soft markers on booking scan Invasive testing for diagnosis	95%
Trisomy 13	Also known as Patau's syndrome, it is a serious genetic condition.	2 in 10,000 births	80% will die in the first year	Soft markers on booking scan Invasive testing for diagnosis	95%
Anomaly	Definition	Incidence	Test	Detection Rate	Rate of false positives
Trisomy 21	Also known as Down's syndrome, it is a genetic condition.	1 in 600 to 800 births	Combined test	85% detection	5% false positive
			Quadruple test	81% detection	5% false positive
			Integrated test	95% detection	5% false positive
			CVS or amnio	100% certainty	

Sourced from Public Health England (Public Health England 2013)

1.3 ANTENATAL SCREENING FOR THE FASP ANOMALIES

An important distinction to make is the difference between the terms *screening* and *diagnosis*. While to some these terms may seem self-evident, there is a substantial body of evidence to suggest that within the context of antenatal screening women frequently do not understand the concept of screening and risk, and how this differs from diagnosis (Asplin, Wessel *et al.* 2012, Al-Jader, Goodchild *et al.* 1990, Baillie, Mason *et al.* 1997, Baillie, Hewison 1999, Freda, DeVore *et al.* 1998, Garcia, Bricker *et al.* 2002, Grewal, Moss *et al.* 1997).

The official definition of screening is “*a process of identifying apparently healthy people who may be at increased risk of a disease or condition. They can be offered information, further tests and appropriate treatment to reduce their risk and/or any complications arising from the disease or condition*” (UK National Screening Committee 2013). Diagnosis, on the other hand, stems from the Greek meaning ‘to know’ or ‘to recognise’ (Stevenson, 2013). These definitions highlight the essence of the difference between the two terms in relation to uncertainty. Screening directs women from a position of complete uncertainty to a position of quantified uncertainty (Aune, Möller 2012). Diagnosis provides some certainty. However, in some cases prognosis remains uncertain despite a diagnosis. In particular, the prognosis of structural anomalies such as cardiac, congenital diaphragmatic hernias (CDH) or central nervous system (CNS¹) anomalies retains an element of uncertainty until after birth. The impact of a CDH on lung development, for instance, is not detectable antenatally, with the possible outcomes being either a surgically-correctable lesion or death. CNS and cardiac anomalies, on the other hand, are more complex in terms of outcome, where survival can be associated with varying degrees of morbidity (Centres for Disease Control and Prevention

¹ The central nervous system (CNS) is the part of the nervous system consisting of the brain and spinal cord. Anomalies affecting the CNS include anencephaly, spina bifida, Ventriculomegaly (see Table 1.1).

2014). These uncertainties arise from the fluidity of diagnosis and subsequent prognosis. Diagnosis is reliant on ultrasound technology and subsequent human interpretation of the findings. The huge variability in diagnosis of cardiac anomalies between different centres (ranging from 16.7% to 94%) is highly suggestive of the significant impact of human interpretation (Moore 2013). Added to this is the uncertainty of prognosis as antenatally unidentifiable co-morbidities can significantly change a prognosis. For example, a postnatal diagnosis of pulmonary hypertension can change a prognosis from treatable to lethal (ELSO 2005).

In an attempt to capture this uncertainty, I developed the table below to group the anomalies in terms of severity and uncertainty of diagnosis and prognosis.

Table 1-2 FASP anomalies categorised by severity and uncertainty

	Possibility of a definite diagnosis	Uncertain diagnosis
Well defined, probable lethal prognosis	Anencephaly Trisomy 13 (Patau's) Trisomy 18 (Edward's) Bilateral Renal agenesis	
Uncertain prognosis	Trisomy 21 (Down's Syndrome)	Congenital diaphragmatic hernia Severe cardiac Open Spina Bifida Lethal skeletal dysplasia Exomphalos
Correctable		Cleft lip/palate Gastroschisis

The light blue sections include the anomalies that have been studied as part of this research. Determining what constitutes a severe anomaly is complex, as it involves making judgments about the prognosis. Where this is uncertain, those judgments are difficult to make. In this instance, I have classified all the anomalies in blue as severe, although they involve varying levels of uncertainty that are likely to impact on the decision to continue or terminate an affected pregnancy (Statham 2002, Jeon, Chen *et al.* 2012). As demonstrated in Table 1-2, the anomalies included form two groups. The first is associated with an element of certainty in diagnosis and subsequent prognosis and includes the lethal chromosomal anomalies, which can be identified by invasive testing, along with two structural anomalies, anencephaly and bilateral renal agenesis. All four of the anomalies are lethal, and there are no treatment options. The second group is associated with a greater degree of uncertainty, both in relation to diagnosis and prognosis. These anomalies are reliant on human interpretation of ultrasound scanning for diagnosis. In addition, treatment options are available, although these also come with some risks. Each anomaly also occurs across a spectrum; for example, one spina bifida diagnosis cannot necessarily be equated to another. Therefore, the point at which any of these anomalies become severe is very difficult to define, and likely to be subjective. This further adds to the complexity to the decision, where a risk of the baby being born severely disabled and surviving to childhood exists, as the anomalies may not be lethal in the short term. This adds a different dimension to the decision. These complexities are explored further in section 1.3.1.1.

1.3.1 *THE SCREENING PATHWAY*

Pregnancy is divided into three trimesters. The first covers weeks 1-13, the second weeks 14-26 and the third weeks 27-40. Screening is undertaken in the first two trimesters only.

Although screening is broadly standardised throughout England, variations in the tests offered exist; these vary according to the gestational age at presentation and, occasionally, local policy. Some women will be offered a 'booking' or 'dating' scan at around eight weeks' gestation. This scan is intended to estimate gestation and to check the viability of the pregnancy. In practice this is more often combined with

the later 'nuchal fold scan' performed at 11-14 weeks. In addition to providing an estimate of the due date, the scan looks for a nasal bone and measures the nuchal fold (the clear space in the tissue at the back of the fetus' neck). Additional space, or absence of a nasal bone, is a potential indicator of a range of anomalies including chromosomal, or some major structural anomalies. Depending on the gestation and therefore development of the fetus during screening, some anomalies such as anencephaly may be visible at this point. Around the same time as the scan, a blood test measuring three biochemical and protein markers in the mother's blood is taken. Combined, the blood test and scan results comprise the main screening test offered in the first trimester, the aptly named 'Combined Test' (or 'Triple Test'). This provides the woman with a 'risk value', computed by incorporating the blood test and scan measurements alongside demographic details of the mother.

Some women with a raised risk identified through the Combined Test may decide to access a further diagnostic test such as Chorionic Villus Sampling (CVS). Unlike the screening tests, these carry a small risk of miscarriage. The test involves removing a small piece of the placenta and examining the chromosomes; this is recommended between 11 and 14 weeks' gestation.

1.3.1.1 SECOND TRIMESTER SCREENING

In instances where the woman has presented too late for the Combined Test, or changed their minds about screening, a Quadruple Test is offered between 15 and 20 weeks. This blood test measures four markers in the maternal blood sample and provides women with a risk value. High risk women then have the option of proceeding to an amniocentesis. This involves removing a small amount of amniotic fluid that contains cells from the fetus. These cells are subsequently analysed for chromosomal anomalies. Amniocentesis can be performed from 15 weeks' gestation onward but, as with CVS, it carries a small risk of miscarriage. The second major screening process in the second trimester is the anomaly scan.

This is undertaken between 18 and 21 weeks' gestation. The detailed scan is intended to look for key structural anomalies and additional 'soft markers'². These markers are not anomalies themselves but may warrant further investigation. They include things like echogenic bowel, dilated atrium, additional fluid or a small fetus (Van den Hof, Wilson 2005).

Structural anomalies, such as cardiac, CDH or CNS anomalies can be diagnosed by an ultrasound scan. Earlier scans may not detect these anomalies as the organs may not yet be sufficiently developed. The association between selected structural and chromosomal anomalies is well documented (Acevedo-Gallegos, Garcia *et al.* 2013). Subsequently, identification of certain structural anomalies will result in the offer of invasive diagnostic testing.

Where a severe anomaly is identified, parents face further decision-making in terms of whether to continue or terminate the affected pregnancy, with termination offered as a 'treatment' option. For those who continue with the pregnancy, additional support and monitoring are offered. Information gained antenatally enables suitable plans to be put in place to manage mother and baby post-delivery. Discussion of issues such as location and method of delivery, as well as how much medical support to provide to the baby post-delivery, such as ventilation or potential surgery, are invariably discussed with parents at this point. For those parents who decide to terminate the pregnancy, a number of issues become pertinent. These include the medical process involved in terminating a pregnancy, including feticide, alongside issues of consent and the law. An overview of these will be provided in the next section.

² Soft markers are minor ultrasound abnormalities. They are considered variants of normal, which do not constitute a structural defect but may be associated with chromosomal or non-chromosomal abnormalities.

1.4 TERMINATION LAW AND THE LEGALITIES OF CONSENT

The legal framework is constructed within the framework of societal beliefs and values. Laws are written and amended in order to create a sense of order and to protect individuals. By doing so, constraints are automatically placed on others, and a balance is sought. Currently within the fetal medicine setting in England it is the role of clinicians to interpret the law and balance the needs of the mother and baby, which are inextricably linked. Pregnancy is a liminal border state that is neither one nor two people (Harris 2008). As the law stands, termination is a criminal act but is permissible within certain legal parameters. As illustrated in Table 1-3 overleaf, since the Ellenborough Act of 1803 the status of the fetus has been protected by law. Changes in technology and medical knowledge are reflected in the way the law has changed, particularly in relation to the gestational limit for termination. It was not until 1990 that the law was amended to differentiate between women who terminate for a congenital anomaly (Section E terminations) and those who terminate for other reasons (Section C terminations).

Prior to performing a termination, clinicians have a legal, ethical and professional duty to obtain informed consent from their patient (Department of Health 2003). This duty is derived from the principle of autonomy (Beauchamp, Childress 1989) as well as criminal and civil law, where treating someone without permission could be considered assault or battery (Maclean 2009). The issue of consent is more complex in contexts such as that in this study, due to the unique maternal-fetal relationship, which involves two patients, with access to one through the other (American Congress of Obstetricians and Gynecologists 2005). Consent for any procedure involving the baby must be sought from the mother.

Table 1-3 Key milestones in termination law from 1803 to date

Date	Law	Summary
1803	Ellenborough Act	Brought in the death penalty for termination after 'quickening' (when movement is felt at 16-20 weeks).
1861	Offences Against the Person Act	Made it a crime for a woman to 'procure a miscarriage', or for another person to help her do so.
1929	Infant Life Preservation Act	Created a new crime of killing a viable fetus (at that time fixed at 28 weeks) in all cases except when the woman's life was at risk.
1931	The Bourne Decision	Dr Bourne challenged the law in order to clarify what constituted legal practice in relation to termination. He was acquitted, thus setting a case-law precedent. Continues to govern termination in Northern Ireland.
1939	The Birkett Committee	Set up by the government in 1936 to clarify the law, they recommended that doctors could perform an abortion to save a woman's life. World War II interrupted implementation of findings.
1967	The Abortion Act	Legalised termination under certain conditions up to 28 weeks. Conditions included: risk to physical or mental health of mother or existing children, <i>substantial</i> risk that child would be born with severe physical or mental abnormality that would result in <i>serious</i> handicap.
1990	The Human Fertilisation and Embryology Bill	Introduced specific time-limits on termination. 24 weeks for terminations performed where the risk to the mother or her family of continuing with the pregnancy, is greater than terminating (Section C). Limit removed where there is a substantial risk that the child would be born with severe abnormalities (Section E), or to save the life or to prevent grave permanent injury to the physical or mental health of the pregnant woman.

Adapted from information provided by BPAS (BPAS 2013)

1.5 THE TERMINATION PROCESS

There are two methods of terminating a pregnancy: surgical or medical (RCOG 2010). Surgical termination can be performed up to 15 weeks' gestation by vacuum aspiration or between 15 and 24 weeks by dilatation and evacuation (BPAS 2010). Both forms of surgical termination are usually performed under general anaesthesia. Medical terminations are not limited by gestational age, although risks of termination increase substantially with increasing gestation. To induce medical abortion, the drug mifepristone is given followed by misoprostol 36-48 hours later (BPAS 2010). The medication induces labour, and the woman subsequently delivers the baby. A recent study found that women undergoing second trimester terminations, for any reason, reported surgical methods as less painful and more acceptable than medical (Kelly, Suddes *et al.* 2010). Furthermore, in the same study, more than half of those undergoing medical terminations reported the experience as having been worse than expected (Kelly, Suddes *et al.* 2010).

The issue of choice of termination method has been raised repeatedly by the British Pregnancy Advisory Service (BPAS³) and Antenatal Results and Choices (ARC⁴), particularly in relation to pregnancies associated with a severe congenital anomaly (Fisher 2013b). This stems from the marked variation between modes of termination performed under Section C or Section E, with Section C terminations significantly more likely to be surgical (RCOG 2010). One explanation for this variation relates to the inability to perform a post-mortem following a surgical termination, and hence obtain a definitive diagnosis (RCOG 2010). However, it has also been argued that the move of termination services to the independent sector

³ BPAS is a national organisation that supports women in making reproductive choices. This includes providing affordable services to prevent unwanted pregnancies with contraception or end them by termination.

⁴ ARC is a national charity that provides counselling and support to parents and professionals in relation to antenatal screening and its consequences.

has resulted in fewer National Health Service (NHS) clinicians skilled in dilatation and evacuation post 13 weeks' gestation (Fisher 2013b). Recent studies have noted that there is an urgent need to introduce new training strategies if women are to be offered the method most suited to them (Kelly, Suddes *et al.* 2010). Irrespective of the rationale, the result is an inequality in choice.

An added decision, for a small group of women, is whether or not to have a feticide as part of the termination process. In a medical context, the term feticide refers to the act of inducing fetal demise (Graham 2008). In terms of guidance, the Royal College of Obstetrics and Gynaecology (RCOG) strongly recommend the routine offer of feticide before later terminations (after 21+6 weeks). Live birth becomes increasingly common after 22 weeks' gestation and this guidance is centred on the avoidance of resuscitation that runs counter to the objective of terminating a pregnancy (Lohr 2012). Several methods are possible including intra-cardiac⁵ injection of potassium chloride, which tends to be favoured in the UK (Graham 2008), intra-amniotic⁶ injection of digoxin, and transection of the umbilical cord (Diedrich, Drey 2010).

Feticide remains an illegal activity, with exceptions made in certain circumstances. The word feticide is intended to represent a neutral phenomenon, in this instance, a medical procedure, but this may not be the case in other social discourses where it is used in arguments for redefining categories of fetal harm as ethically or legally unreasonable (Graham 2008). In section 1.7 the negative connotations associated with the terminology will be shown to be prominent in discussions with parents (Statham, Solomou *et al.* 2000). Little is known about how parents and clinicians feel about this procedure, (Graham 2008, Lohr 2012). One of the few studies published, which included, as secondary analysis, parental attitudes to feticide,

⁵ Intra-cardiac injection refers to the injection of a substance under ultrasound guidance, through the abdomen of the woman into the heart of the fetus.

⁶ This involves injection of digoxin through the abdomen of the woman into the amniotic fluid surrounding the fetus. The digoxin is absorbed by the fetus, causing death.

reported that 92% of participants expressed a strong preference for feticide (Jackson, Teplin *et al.* 2001). Although knowing the exact moment their baby died may be very distressing for parents, it may also provide some comfort for those who wish to know when their baby stopped suffering (Statham, Solomou *et al.* 2000).

1.6 STIGMA AND TERMINATION

Goffman described stigma as “*an attribute that is deeply discrediting,*” that reduces the possessor “*from a whole and usual person to a tainted, discounted one*” (Goffman 1963 pg.3). This definition has been applied and built on across many disciplines, nonetheless two components of stigmatisation consistently appear; first the perception of negative characteristics and second the global devaluation of the possessor (Norris, Bessett *et al.* 2011). Termination or ‘abortion stigma’ has been defined as “*a negative attribute ascribed to women who seek to terminate a pregnancy that marks them, internally or externally, as inferior to ideals of womanhood*” (Kumar, Hessini *et al.* 2009 pg.628). The manifestation and perpetuation of abortion stigma is highlighted in five steps: labelling of differences as deviant; linking those labelled with undesirable characteristics; separating them and us; instilling a sense of shame in those labelled; and finally discrimination or rejection of those concerned (Shellenberg, Moore *et al.* 2011).

Unlike many other attributes open to stigmatisation, including colour, race and gender, stigma associated with termination is considered a concealable stigma as it is unlikely to be known to others unless disclosed (Quinn, Chaudior 2009). Although the risk of stigmatisation usually pertains to the women who have had a termination, it may also apply to others, including healthcare providers and partners of women who have had a termination. As with women who have had terminations, these other groups are not fully in control over whether their status is revealed by and to others (Norris, Bessett *et al.* 2011). Consequently, those stigmatised by termination have to cope not only with the stigma once revealed, but also with managing whether or not the stigma will be disclosed (Quinn, Chaudior 2009).

1.7 TERMINOLOGY

The final section in this chapter provides a brief review of the terminology employed throughout this thesis. The use of language in highly emotive subjects such as this requires careful consideration. The decision to use one term over another is not incidental.

The word 'termination' has been carefully selected over that of 'abortion', as abortion frequently carries negative connotations. Where I use the term 'abortion', it is either intended to reflect the terminology used by a participant, or that identified within the literature. One of the more poignant choices has been the use of the word 'baby' rather than 'fetus'. This reflected the terminology employed by the parents and, along with the word 'parents', has been used throughout to convey the sentiments of the women and their partners. The word 'choice' regarding termination has been replaced by 'decision'. This holds particular poignancies, as for all the parents concerned there was no choice, as choice suggests the voluntary option of one action over another. Although a rational decision may exist, this may not necessarily translate into a rational choice. Throughout the thesis, the term 'healthcare professional' is used to include all professionals working within the healthcare setting. In order to differentiate, for the purpose of this thesis the term 'clinician' has been used to refer to specialist doctors and surgeons who care for women and their babies affected by a severe congenital anomaly. This includes fetal medicine clinicians, geneticists, neonatologists and sub-specialties with a special interest in fetal medicine, including cardiology and neurology or neuro-surgery. Midwives are referred to separately as 'midwives' and are not included in the 'clinician' label.

Specific terminology associated with termination law is also applied to the baby. As highlighted in Table 1-4, the status of the baby is dependent on the gestation at delivery and whether or not the baby shows any signs of life at birth. In turn, the given status precludes or enables the mother to access financial support and time off from work. A feticide may therefore have an impact on the definition applied to the baby, and thus the support available to the mother.

Table 1-4 Terminology applied to fetal death, and support available

	Late Miscarriage	Stillbirth	Neonatal Death
Definition	Baby is born, showing no sign of life, between 14 and 24 weeks of completed pregnancy.	Born after the 24th week of pregnancy, showing no sign of life.	Baby is born at any gestational age and showing signs of life and then subsequently dies within 28 days.
Maternity leave and pay	No entitlement to maternity leave or pay. Sick leave – reliant on GP to certify.	Entitled to full maternity benefits providing sufficient national insurance contributions have been made. Free prescriptions and dental care.	Entitled to full maternity benefits providing sufficient national insurance contributions have been made. Free prescriptions and dental care.
Child benefits	No child benefit or tax credits.	No child benefit or tax credits.	Child benefit up to 8 weeks after the baby's death.
Registering the birth and death	Cannot be registered. No birth, death or stillbirth certificate is released.	No legal requirement to register. A Medical Certificate of Stillbirth is provided that enables the baby to be registered if the parents wish.	Birth and death certificates are issued. There is a legal requirement to register the baby.
Legal requirement for a burial or cremation	No	Yes - hospital will arrange to do so free of charge.	Yes - hospital will arrange to do so free of charge.

Adapted from NHS Choices (NHS Choices 2015)

Finally, the difference between the terms ‘fetal’ and ‘congenital’ has been given consideration. Whilst the two terms are often used inter-changeably, there is a subtle difference in that a fetal anomaly relates to an anomaly during fetal life, while a congenital anomaly is an anomaly present at birth. Although an assumption could be that any anomaly present at birth must have been an anomaly prior to birth, and *vice versa*, this is not necessarily the case. For example, a Ductus Arteriosus (the small duct between the pulmonary artery and the descending aorta) is a normal finding in the fetus, but if it fails to close after birth it may require surgery to correct it. This congenital cardiac anomaly is known as a Patent Ductus Arteriosus (PDA). Conversely, some structural anomalies involving under-development of organs may grow after birth. Thus what may have been a fetal anomaly does not develop into a congenital anomaly. Although this difference will not impact on the anomalies included within this thesis, as they will be both fetal and subsequently congenital anomalies, the term ‘congenital’ has been applied throughout the thesis in relation to the anomaly.

1.8 SUMMARY

The variations observed in neonatal and infant mortality in babies with a severe congenital anomaly have been partly attributed to the decision to continue or terminate the affected pregnancy. With increasing consideration given to policy aimed at reducing inequalities, such as financial incentives to stop smoking during pregnancy (Tappin, Bauld *et al.* 2015), and mandatory fortification of bread or flour with folic acid (Crider, Bailey *et al.* 2011), it is important to understand how these differences in outcome occur. The aim of this study is to explore the decision-making process of women and their partners following diagnosis or suspicion of a severe congenital anomaly. An important element of this will be exploring the possible influences of socioeconomic status on this process. This is not an attempt to frame any particular decision as ‘flawed’ but rather to ensure that any variation is the result of free choice, rather than a systemic failure in service provision for a particular group.

2 REVIEW OF THE LITERATURE

This chapter provides an analysis of the relevant literature, and how it has contributed to the development and refinement of my overarching research question:

“How do women and their partners make the decision to continue or terminate a pregnancy following suspicion or diagnosis of a severe congenital anomaly?”

A sub-question exploring the influence of wider contextual factors, in particular socioeconomic status, on the decision-making process is also considered.

The literature search was divided into two parts: the first was designed to identify empirical literature relating to decision-making following diagnosis or suspicion of a severe congenital anomaly; the second reviews theoretical literature particularly pertaining to decision-making. I have applied differing search strategies to these two parts, which will be discussed separately alongside the literature identified and the rationale for this decision.

In order to capture the relevant literature, I needed a broad, structured approach, and therefore applied an amended Population-Intervention-Comparison-Outcome (PICO) method (Richardson, Wilson *et al.* 1995). In this manner, I dissected and restructured the question into its component parts to ensure that a search was undertaken for all the key aspects. The benefit of this approach in relation to defining clinical research questions has been documented (Sayers 2008, Sayers 2007) and adapting these principles to my context provided a framework in which to start the literature review. The component parts of the question were: congenital anomaly (population/problem); decision-making (intervention); socioeconomic status (comparison); antenatal testing (outcome 1); and termination (outcome 2). Alternative wording for each component was applied; for example, the search for ‘congenital anomaly’ included ‘fetal anomaly’, ‘foetal anomaly’, ‘fetal anomalies’, ‘fetal abnormalities’ etc. (See Appendix A for a full

breakdown). The search was run through the Scopus and Pubmed databases. Individually these searches identified over a million papers, covering a broad range of subjects from economics, consumerism and organisational decision-making to in-utero surgery. Combining the searches, for instance 'decision-making *and* congenital anomalies' or 'decision-making *and* termination', narrowed the literature and enabled less relevant articles to be excluded. For example, a large number of articles were identified when searching for 'congenital anomalies' alone. Many of these addressed clinical problems such as treatment options or surgical outcomes, and as such had limited relevance to the area of interest. Once the combined searches had been run and duplicates removed, fewer than 3000 articles remained. Finding the balance between being too broad and too specific was difficult, and I tended to over-search, particularly when little literature specific to the question was identified. All the combined results were hand-searched and articles of interest selected after screening the title; 711 titles remained of varying interest. Not all of these were subsequently drawn upon to direct the research, but have been peripherally influential in the way I approached the study. Although the initial search was undertaken at the beginning of the PhD, the search was saved and updates automatically forwarded to me on a monthly basis. Searches based on authors and citations of primary articles were also set up. In order to make some sense of the literature, I categorised it according to how it addressed a number of questions:

- *How do parents decide whether to continue or terminate a pregnancy affected by a severe congenital anomaly?*
- *What factors influence their decision?*
- *What do we know about parents' experiences of the decision-making process?*
- *What are the experiences and views of healthcare professionals about caring for parents during the decision-making process?*

This chapter examines each of these questions in turn, while providing a discussion of the literature available and identifying gaps in knowledge that subsequently provided the framework in which I developed my study. The chapter

ends with a statement of my research question alongside a description of my thought processes leading to its development.

2.1 HOW DO PARENTS DECIDE?

The first question I posed was: ‘How do parents decide?’ Searches identified no literature pertaining specifically to the decision-making process following diagnosis or suspicion of a severe congenital anomaly, although three papers highlighted the need to investigate this area further (Bijma, van der Heide *et al.* 2008, Schechtman, Gray *et al.* 2002, Shaffer, Caughey *et al.* 2006). At first glance, the titles of these three papers appear to directly address the research question posed; for example Bijma’s paper entitled: “*Decision-making after ultrasound diagnosis of a fetal anomaly*”. However, rather than exploring decision-making *per se*, this paper examined the influence of ultrasound on decision-making in the antenatal period; highlighting the lack of preparation of parents should an anomaly be identified (Bijma, van der Heide *et al.* 2008). The other two papers stemmed from the United States (US) and sought to identify or evaluate the influence of particular variables in order to explain or predict parental decision-making (Shaffer, Caughey *et al.* 2006, Schechtman, Gray *et al.* 2002). There are a number of similar such studies, the findings of which will be discussed in section 2.2. However, the titles of these were particularly pertinent as they indicated that the primary objective was an examination of decision-making following diagnosis of a congenital anomaly. Whilst both authors identified a number of variables associated with the decision to continue or terminate an affected pregnancy, the articles provided little insight into how that decision was made. The paper by Schechtman *et al* was based on a large scale quantitative study providing inferential statistics on termination decisions. They graded anomalies on an ordinal scale based on ‘severity’ where ‘one’ was no impact on quality of life through to ‘four’ that was lethal. They concluded that the more severe the anomaly, the more likely it was the parents would terminate. CNS anomalies were also more likely to be terminated than non-CNS anomalies. In addition, they showed that maternal age and education impacted on the decision, where younger and less well-educated women were more likely to continue an affected pregnancy

than their counterparts. Similarly, in a study of 833 fetuses with a chromosomal anomaly, Shaffer *et al* concluded that severity of the anomaly, maternal age and ethnicity contributed to parental decision-making following diagnosis of a chromosomal anomaly. A subgroup of sex chromosomal anomalies (non-lethal) was compared to a subgroup of autosomal anomalies (including Down's, Patau's and Edward's Syndromes). Whilst Patau's and Edward's are indisputably severe, as demonstrated in Table 1-2, Down's is less clear-cut. In addition, it is associated with a higher termination rate than other anomalies (Budd, Draper *et al.* 2015). The issues relating to Down's will be explored more fully later in this section; nonetheless, incorporating Down's within the 'severe' group may ignore intrinsic differences and subsequently impact on the conclusions made, namely that severity of the anomaly is associated with likelihood of termination. As I argue in section 2.2, despite the contribution of quantitative literature to the understanding of parental decision-making, in isolation, application of these studies to the question posed may be of limited benefit. In addition to these articles, two areas of interest were identified. These have been entitled 'Hypothetical Decision-Making' and 'Antenatal Screening and Decision-Making'.

2.1.1 *HYPOTHETICAL DECISION-MAKING*

UK-based surveys exploring views on termination following an antenatal diagnosis of an anomaly suggest that most people indicate they would not abort an affected pregnancy (Singer 1993, Green, Snowdon *et al.* 1993, Evers-kiebooms, Denayer *et al.* 1993). With the caveat that the surveys were undertaken over 20 years ago, and attitudes may have changed due to public knowledge and awareness of anomalies and the subsequent development of a national screening programme, current termination rates indicate that, following an antenatal diagnosis, a high proportion of parents do decide to end the affected pregnancy (Budd, Draper *et al.* 2015).

There are a number of studies that provide some explanation for this variation. For instance, Sawyer (Sawyer, Cerritelli *et al.* 2006) recorded the attitudes of parents to antenatal testing and termination of pregnancy in a hypothetical pregnancy. All the families had a young child with a genetic disorder and demonstrated high levels of understanding of the condition. These were then compared with actual

reproductive behaviours five years later. The findings highlighted that the hypothetical responses often did not reflect the decisions made in subsequent 'real-life' scenarios. Intra-parent comparisons showed that those 'real-life' decisions moved in both directions; i.e. some of those that said they would terminate, continued and *vice versa*. Due to the pre-existing high levels of understanding, this shift could not be explained by increased knowledge, with the authors concluding that attitudes towards antenatal diagnosis and termination of pregnancy differ, and are dynamic over time.

A further study, undertaken between 1998 and 2000, sought to investigate the differences in attitudes between women from the former East and West Germany (Erikson 2003, Erikson 2007, Erikson 2001). Although the Berlin Wall had come down eight years previously, reconciling the differences between the former East and West sectors took much longer, with an initial shared maternity system not agreed until 1993. Whereas East German laws on termination were liberal, those in the West were much more conservative, and framed around the beliefs of the Catholic Church. Benefits and expectations of antenatal screening similarly differed (Erikson 2001). This meant that during their formative years, participants would have been subject to different influences in terms of termination laws, policies and attitudes to reproduction. In terms of data collection, questionnaires followed by one-to-one interviews explored responses to hypothetical decisions on termination. The data generated were compared to actual termination rates and a 'disconnection' between the two highlighted. Whilst the majority of women from the former East Germany stated, hypothetically, that they would terminate an affected pregnancy, those from the former West were more likely to state that they would continue. However, comparison of actual termination numbers suggested that when faced with a 'real-life' decision, the differences between the two groups disappeared, with the rate of terminations in the former West Germany much higher than would have been suggested by the responses to the hypothetical scenarios. Meanwhile the hypothetical and 'real' decisions in the former East Germany were closer in range. The differences between the hypothetical responses of the two groups were suggested by the author to be a reflection of the diverse political environments in which their views on termination had formed.

Whereas women from the former East Germany tended to consider termination a right, those from the West viewed termination as a “*privilege managed by the state*” (Erikson 2003, pg.1991). Although not a primary discourse within the paper, these findings highlight the power of the external social context on responses to hypothetical scenarios. Despite shared macro level structures, such as a combined health service, women’s perspectives of their choices were seen through very different lenses. These perspectives had a greater or lesser impact depending on whether the scenario was hypothetical, or ‘real-life’.

2.1.2 ANTENATAL SCREENING, DOWN’S SYNDROME AND DECISION-MAKING

There is a growing body of literature exploring the decision-making processes enacted by parents when determining whether to accept or reject antenatal screening. These papers include studies from the Netherlands (Garcia, Timmermans *et al.* 2008), Iceland (Gottfredsdóttir, Björnsdóttir *et al.* 2009), Israel (Remennick 2006, Lewando-Hundt, Shoham-Vardi *et al.* 2001), Australia (Liamputtong, Halliday *et al.* 2003), US (Markens, Browner *et al.* 1999), Norway (Aune, Möller 2012), Taiwan (Chiang, Chao *et al.* 2006), France (Khoshnood, De Vigan *et al.* 2004) and the UK (Prathapan, Adams *et al.* 2012, Williams, Sandall *et al.* 2005).

Variations in screening and termination policies exist both within and between the countries from which the papers were collated (Cook, Erdman *et al.* 2014), with these differences reflected in their findings (Reid, Sinclair *et al.* 2009, Henry 2003). For example, an Israeli study examining low screening uptake rates within the Bedouin Tribe highlighted the influence of the payment system. In this example, a levy was paid to cover all care within a six-month period. By presenting late for antenatal care, subsequent payments for the baby’s immunisations would also be covered under the same levy (Lewando-Hundt, Shoham-Vardi *et al.* 2001). The decision-making process is shown to be heavily influenced by contextual factors including social, cultural and economic constraints (Henry 2003). The issue of context is particularly pertinent when addressing the question of how parents decide. International comparisons of antenatal screening uptake highlight wide variations across Europe (Boyd, DeVigan *et al.* 2008). Whilst much of this variation

can be explained, in part, by differences in screening policy, the variations persist even between countries with a similar approach. For instance, the uptake of Down's screening is 27% in the Netherlands versus 61% in the UK and 90% in Denmark (Crombag, Vellinga *et al.* 2014). These variations are perhaps indicative of macro differences concerning the way in which screening is offered (either as an extra option or as part of routine care), and how this offer is perceived within the social context (Crombag, Vellinga *et al.* 2014), as well as attitudes towards anomalies. Interpretation of findings should, therefore, take into consideration the context in which they were produced. Laws governing terminations vary significantly across the globe, from blanket bans in many countries in South America, to an unlegislated and subsequently medically determined termination policy in Canada (Cook, Erdman *et al.* 2014). Consequently, decision-making following diagnosis of a congenital anomaly will be heavily contextualised, potentially more so than for screening.

Irrespective of the broader context in which decision-making about antenatal screening occurred, the complexity of the decision-making process was widely recognised and a number of common influences identified by Reid *et al* in a meta-synthesis (Reid, Sinclair *et al.* 2009). Out of 389 papers identified, Reid *et al* selected nine for examination, with the remainder excluded as they predominantly reflected clinicians' views, or were a quantitative analysis of variables associated with decision-making (a pattern reflective of the searches undertaken for my study). Emergent themes were classified into five core concepts: 'destination unknown' reflecting the influence of parental views on termination and views on caring for a disabled child; 'to choose or not to choose', perceptions of the maternal-fetal relationship; 'risk is rarely pure and never simple', an interpretation of risk and the impact of the way in which screening was offered; 'treading on dreams' relating to the impact of technological developments on expectations of responsible motherhood and the quest for a 'perfect' baby; and 'betwixt and between', the liminal state associated with screening. This final concept included conflicting states such as the need to know or not to know, the right to know set against the right not to know, and the risk of knowing versus the risk of not knowing the diagnosis (Reid, Sinclair *et al.* 2009). Therefore, acceptance

or rejection of screening would reflect the weight and influence each of these concepts was perceived to carry.

The evidence collated included a number of papers specifically exploring the experiences of women who chose to reject screening (Liamputtong, Halliday *et al.* 2003, Markens, Browner *et al.* 1999). There is often a tendency to assume that this decision represents an attempt to resist the medicalisation of pregnancy or opposition to termination (Rapp 1998, Green, Snowden *et al.* 1993). Although these issues were high on the priority list of women who chose not to undergo screening, they were not the sole concerns raised. In particular, the issues of risk and understanding risk were commonly cited as reasons for the rejection of screening (Lewando-Hundt, Shoham-Vardi *et al.* 2001, Liamputtong, Halliday *et al.* 2003). For this group, a dichotomy arose between the medical need to minimise risk in all its manifest forms, and the women's perception of risk as an integral part of life (Lewando-Hundt, Shoham-Vardi *et al.* 2001).

This provided an alternative insight to previous research on women's acceptance of antenatal screening that suggests women often do not perceive a choice when accessing antenatal screening (Liamputtong, Halliday *et al.* 2003). This can be observed in the higher rates of uptake of screening associated with the offer of tests as part of a routine antenatal visit (Dormandy 1999, Pilnick, Fraser *et al.* 2004), where the very offer of a test may be perceived as a recommendation (Press, Browner 1997), and is compounded by evidence that women may be poorly informed, accept screening as routine, and have insufficient information to make an informed decision (Aite, Zaccara *et al.* 2011, Williams, Alderson *et al.* 2002b, Press, Browner 1997, Al-Jader, Parry-Langdon *et al.* 2000, Heyman, Hundt *et al.* 2006).

Initial suspicion or diagnosis of a congenital anomaly is not only traumatic in itself, but may be made more so by the conflict arising from the gap between parental expectations and reality. In addition to the poor parental understanding of screening discussed above, a number of 'mythical expectations' relating to pregnancy exist. These include: 'the baby will be fine if the pregnancy reaches the

second trimester'; 'an abnormal fetus will miscarry'; 'I wouldn't terminate anyway'; and 'Down's is the worst it can get' (McCoyd 2007). Faced with the reality of a diagnosis, the need to overcome these 'mythical expectations', in order to make a decision, adds to parental distress.

When attempting to address the question, 'How do parents decide,' much of the available literature relates to antenatal screening and the decision to accept or reject this, rather than the subsequent decision to continue or terminate an affected pregnancy. While both decisions are made within the antenatal setting, important differences exist. Although, in theory, an informed decision on screening would include consideration of the decision to continue or terminate an affected pregnancy, any decision regarding termination made at this point would essentially be hypothetical (Williams 2005), and therefore likely to differ from a decision based on a 'real-life' scenario (Erikson 2003, Wertz 1992, Press, Browner 1997, Sawyer, Cerritelli *et al.* 2006). Furthermore, attitudes to screening are not necessarily a good indicator of attitudes towards termination (Markens, Browner *et al.* 2010, Hewison, Green *et al.* 2007). Whilst the available evidence provides an insight into the broader issues of non-directiveness, informed choice and risk, application of the evidence to post-diagnosis decisions may not be without difficulty.

Whilst undertaking the literature search, I noticed that much of the evidence relating to decision-making and congenital anomalies pertained specifically to screening for Down's Syndrome (Bryant, Hewison *et al.* 2005, Bryant, Green *et al.* 2011, Carroll, Al-Janabi *et al.* 2013, Chiang, Chao *et al.* 2006, Crombag, Bensing *et al.* 2013, Crombag, Vellinga *et al.* 2014, Georgsson Öhman, Grunewald *et al.* 2009, Heyman, Hundt *et al.* 2006, Khoshnood, De Vigan *et al.* 2004, Reid, Sinclair *et al.* 2009, Pilnick 2008, Prathapan, Adams *et al.* 2012, Thomas 2014, Vassy, Rosman *et al.* 2014). This is likely to reflect screening technology and subsequent policy, where the discovery of first trimester ultrasound and serum markers has provided the medical expertise to enable screening programmes to develop (Wald, Kennard *et al.* 1998). Furthermore, the higher incidence of Down's Syndrome compared to very rare anomalies both supports the use of screening, whilst creating the

opportunity for study. In the UK the Down's Syndrome screening programme was one of the first national antenatal screening programmes, thus inviting evaluation (Morris 2011, Dormandy, Hankins *et al.* 2006). Perhaps as a result of the extensive press coverage, many parents continue to equate all antenatal screening with Down's Syndrome (Heyman, Hundt *et al.* 2006). I therefore decided to look more closely at Down's and compare it to other FASP anomalies in order to assess transferability of the literature available.

Unlike other major anomalies, UK evidence shows that rates of detection of Down's Syndrome vary with levels of socioeconomic deprivation (Budd, Draper *et al.* 2015). Knowledge and understanding of Down's Syndrome appear to be lower amongst minority ethnic groups and in areas of high deprivation (Chilaka, Konje *et al.* 2001). This is reflected in variations in uptake and access to screening for Down's Syndrome (Alderdice, McNeill *et al.* 2008). However, this variation is not apparent for the remaining FASP anomalies. Termination rates are also significantly higher in the UK for Down's Syndrome than those for other severe FASP anomalies (86% versus 70%), although both groups have similarly wide variation in termination rates between areas of deprivation (Budd, Draper *et al.* 2015). It is unclear, therefore, whether these observed differences are as a result of differing parental attitudes and understanding of the anomalies, or systematic differences in the way screening is offered and subsequent counselling performed. Nonetheless, care should perhaps be taken when transferring findings of studies undertaken in one setting to another.

2.1.3 SUMMARY

When addressing the question of how parents decide, a review of the literature highlighted a number of areas to explore. First, hypothetical decision-making may not necessarily reflect actual decisions. As demonstrated in the studies by Sawyer and Erikson, attitudes are dynamic over time and are reflective of the context in which they are made. Hypothetical responses, therefore, may reflect the expectations rather than the actions of the participant. Second, antenatal screening is essentially decision-making in a hypothetical scenario. Decisions made in relation to antenatal screening and those following diagnosis of an anomaly,

therefore, should be viewed separately, and assumptions that attitudes to one will reflect attitudes to the other, approached with caution. Third, despite the growing international literature base available, transferability of findings from one context or setting to another requires consideration. Where laws, policies and attitudes governing reproductive issues vary significantly, their influence is likely to be reflected in the findings of the respective studies. Fourth, the variations between socioeconomic groups in the offer and uptake of Down's Syndrome screening, as well as the subsequent detection, highlighted a pattern distinct from that observed in the literature pertaining to the FASP anomalies. In addition, the high termination rates compared to other FASP anomalies drew attention to potential intrinsic differences between them, which may make transferring these findings difficult, particularly as no comparative literature was identified, thus making it difficult to identify the cause of these observed differences.

2.2 WHAT FACTORS INFLUENCE DECISIONS?

A number of papers examine the variables associated with women's decisions following positive congenital diagnosis. They have predominantly focused on linking variables, such as gestational age at diagnosis, severity, type of anomaly, religion and socioeconomic status, to women's decisions to continue or terminate pregnancies. Although predominantly quantitative, there were a few mixed-methods and qualitative studies. Methods included systematic review (Jeon, Chen *et al.* 2012), multinational database cohorts (Marteau, Nippert *et al.* 2002), retrospective cohort studies (Shaffer, Caughey *et al.* 2006), focus groups (Ahmed, Atkin *et al.* 2006), surveys (Hewison, Green *et al.* 2007) or questionnaires (Korenromp, Page-Christiaens *et al.* 2007). The studies were undertaken in a number of countries including the UK, US, Israel, Netherlands, France, Uruguay, South Korea and Switzerland. The majority of the studies examined specific anomalies or groups of anomalies, in particular chromosome anomalies (with a sub-group examining sex chromosome anomalies), and cardiac anomalies. Analysis also varied, with some papers presenting descriptive statistics, whilst others supplied inferential statistics.

A number of variables were identified as 'predictors' of decisions. Severity of the anomaly was frequently highlighted as an influential factor (Chenni, Lacroze *et al.* 2012, Feijen-de Jong, Jansen *et al.* 2011, Schechtman, Gray *et al.* 2002) as well as the type of anomaly, for instance the influence of a chromosomal anomaly was determined to be strongly significant and increased the likelihood of termination fourteen-fold (Zyblewski, Hill *et al.* 2009). In part, this may be explained by the association of uncertainty of fetal prognosis as a factor favouring continuation of pregnancy (Pryde, Isada *et al.* 1992). Conversely, the certainty gained from a diagnosis of a chromosomal anomaly may reflect the decision to terminate. Early gestation and high socioeconomic status were also highlighted as associated with the decision to terminate a pregnancy (Ahmed, Atkin *et al.* 2006, Balkan, Kalkanli *et al.* 2010, Chenni, Lacroze *et al.* 2012, Feijen-de Jong, Jansen *et al.* 2011, Kramer, Jarve *et al.* 1998, Pryde, Drugan *et al.* 1993, Schechtman, Gray *et al.* 2002).

Although the influence of each variable was examined separately within the literature, a number of interdependencies were apparent. For example, gestational age at diagnosis and type of anomaly are likely to heavily influence each other. As the fetal organs develop at varying rates, different diagnostic techniques are applied at different stages of the pregnancy to identify these anomalies. Consequently, some anomalies can be identified earlier than others and this makes it difficult to isolate the influence of a specific anomaly (with its own intrinsic risk) and the acceptability of termination after a particular time. This is particularly true for cardiac anomalies, which, along with skeletal anomalies, are most likely to be missed on initial scans (Boyd, Tonks *et al.* 2011, Raupach, Zimmermann 2004).

There were a number of conflicting findings highlighted. Several papers recorded maternal age as being an influencing factor; yet no consensus exists on the direction of influence, with some papers concluding that younger women are more likely to terminate (and conversely older women continue) (Balkan, Kalkanli *et al.* 2010, Quadrelli, Quadrelli *et al.* 2007, Shaffer, Caughey *et al.* 2006), and another suggesting younger women are more likely to continue (and conversely older women terminate) (Hamamy, Dahoun 2004). In addition, others concluded that there was no relationship between maternal age and the decision to continue or

terminate an affected pregnancy (Hawkins *et al.* 2012, Hamamy, Dahoun 2004, Chenni, Lacroze *et al.* 2012). The context in which the studies were undertaken perhaps again provides some explanation. For example, one study from Uruguay (Quadrelli, Quadrelli *et al.* 2007) presented conflicting findings to a study undertaken in Switzerland (Hamamy, Dahoun 2004). Whilst Uruguay is a country where access to termination remains illegal (Gadow, Petracchi *et al.* 2006), Switzerland provides opportunities for women to make a decision within a controlled legal framework. The variability in outcome is perhaps reflective of the context rather than a conflicting finding *per se*.

An association between level of deprivation and the decision to continue or terminate an affected pregnancy has also been demonstrated, where high levels of deprivation are associated with the decision to continue a pregnancy (Smith, Budd *et al.* 2011). This remained the case after adjustment for maternal age and when considering chromosomal and non-chromosomal anomalies separately. This study used data from the largest congenital anomaly register in England, which uses multiple sources of case ascertainment at different points along the care pathway, thus ensuring a high rate of data capture. As a large-scale study providing adjustment for other variables, including maternal age, ethnicity and type of anomaly, this study provides convincing evidence on the impact of deprivation on the decision to continue or terminate an affected pregnancy and subsequently on neonatal mortality rates.

The influence of ethnicity, possibly reflected in different belief systems and religion, has also been highlighted (Ahmed, Green *et al.* 2006, Ahmed, Ahmed *et al.* 2012, Gitsels-van der Wal, Manniën *et al.* 2014, Rapp 2000). In addition, there is evidence to suggest that minority ethnic groups access antenatal services later in pregnancy (Rowe, Garcia 2003, Rowe, Garcia *et al.* 2004, Raleigh, Hussey *et al.* 2010, Alderliesten, Vrijkotte *et al.* 2007), and this subsequently impacts on the choices available and time to make them. However, the influence of this variable may also reflect, to an extent, the impact of deprivation where minority ethnic groups are at higher risk of deprivation (Bécares, Stafford *et al.* 2011).

The impact of the counsellor on the decision has also been raised, particularly within the literature relating to sex chromosomal anomalies, where counselling by an obstetrician rather than a geneticist is associated with a higher termination rate (Kim, Park *et al.* 2002, Holmes-Siedle, Ryyänen *et al.* 1987). Although this may be a reflection of certain parents self-selecting by seeking more detailed information from the geneticist (Statham 2002), it may also reflect the influence of counselling styles, where obstetricians perceive themselves as more directive than geneticists (Marteau, Drake *et al.* 1994). The extent to which this has changed over subsequent years is unclear, where counselling appears to have taken on a more multidisciplinary approach and therefore the influence of a single clinician may have less impact on the decision.

2.2.1 SUMMARY

Review of this literature has provided some clues to factors that may influence parental decision-making following a diagnosis of a congenital anomaly. However, the lack of consistency in the findings and the importance of context when interpreting these suggest that although these studies may provide some insight into factors that are influential, examination of variables alone cannot capture the complexity of the process. Whilst the variables identified provide clues as to which groups are 'at risk' of inequalities, conclusions cannot be drawn as to the rationale for the differences observed. In addition, there remains no evidence as to whether or not these differences are as a result of a systematic failure to provide equality in access and care, which subsequently impacts on decisions made, or whether the difference is a reflection of parental choice. Capturing the influence of these variables within my study was essential and consideration was therefore given to the creation of a sampling frame in which to recruit the participants.

2.3 WHAT DO WE KNOW ABOUT PARENTS' EXPERIENCES OF THE DECISION-MAKING PROCESS?

The lack of literature relating to parents' experiences following diagnosis of a congenital anomaly is well-documented: "*in contrast to the great volume of research on women's decisions about prenatal testing, there is a dearth of research*

on women's decisions following the diagnosis of a fetal abnormality" (Marteau, Dormandy 2001, pg.188). This paper was written nearly 15 years ago and, despite some developments since then, the evidence pertaining to the experiences of parents following diagnosis or suspicion of a severe congenital anomaly remains limited.

The studies that are available are a mixture of qualitative and quantitative literature. Whilst the qualitative papers tend to examine the narratives of parents following the diagnosis of a congenital anomaly and decision to continue or terminate the affected pregnancy, the quantitative literature is predominantly concerned with the measurement of grief reactions and examination of the psychological impact. Much of the qualitative literature stems from two academic research groups and a national charity. These studies were UK-based, and therefore the laws and policies guiding parental decision-making will reflect those of my study. In addition, there are a number of international articles that explore a variety of aspects of the women's experiences. For ease of presentation, an overview of the main contributors is initially provided, before an analysis of the literature as a whole.

The first research group is the Health Experiences Research Group (HERG), a group of social scientists based at Oxford University's Department of Primary Care Health Sciences. Their main interest is in researching patients' experiences of health and illness. They have created a unique database of personal and patient experiences through in-depth qualitative research into over 70 different illnesses and health conditions, which are published on two websites – www.healthtalk.org and www.youthhealthtalk.org. This is the largest such repository of interviews, and serves to support people with a variety of medical conditions by sharing the experiences of others. In terms of parental experience of antenatal screening and subsequent decision-making following diagnosis of a severe congenital anomaly, there are over 57 narratives recorded, made up of interviews with individual mothers, fathers and couples. These include parents whose babies have been diagnosed with a range of congenital anomalies including severe cardiac and lethal chromosomal anomalies, some of whom continued and others who terminated the

affected pregnancy. They are all recorded at some point between one and eight years following the end of the pregnancy and are presented in accessible summaries of findings, each of which reflects an aspect of the care pathway. These include: making decisions about screening, experiences of screening, further tests, and subsequent decisions to terminate or continue a pregnancy. Areas such as feelings and emotions after birth or termination and living with a child with chronic health problems are also covered.

A number of publications have arisen from secondary analysis of these interviews and relate to various elements of the parental experience during the decision-making process (Entwistle, France *et al.* 2011, France, Wyke *et al.* 2011, France, Locock *et al.* 2012, France, Hunt *et al.* 2013, Hunt, France *et al.* 2009, Locock, Crawford *et al.* 2005, Locock, Alexander 2006, Lowe, Powell *et al.* 2009). Particular issues addressed include: disclosure post termination (France, Hunt *et al.* 2013), decision-making in the immediate aftermath of termination (Hunt, France *et al.* 2009), impact of types of information (in particular personal or others' experiences of disability) on decision-making (Entwistle, France *et al.* 2011, France, Wyke *et al.* 2011), as well as providing one of the few insights into the perspective of the father (Locock, Alexander 2006).

A further set of papers arose from the Cambridge Maternal Study, an extensive piece of work led by Helen Statham (Statham, Solomou *et al.* 2001). Although the study has not been published in its entirety, a number of publications have been produced (Statham, Solomou *et al.* 1999, Statham, Solomou *et al.* 2001, Statham, Solomou *et al.* 2000, Statham 2002, Statham, Solomou *et al.* 2006). The aims of the Cambridge Maternal Study were to investigate the experiences and views of women and their partners when an anomaly was confirmed antenatally, and compare these to the experiences of women when the anomaly was not identified until after birth. This was a large-scale project involving the recruitment of 143 women who terminated and 80 women who continued, either through choice or where the anomaly was not antenatally detected (a 39.5% response rate). The sample was stratified in terms of terminated pregnancy, continued pregnancy and postnatal diagnosis. Women (and some partners) were followed up from 6 weeks

to 14 months post-diagnosis. Data were collected through a series of three questionnaires, an interview and a diary to record contact with healthcare professionals. Attrition rate was around 25%, with 75% of the participants completing all aspects of the study. There is little to critique in terms of the comprehensiveness of this study. However, access to the document in its entirety is difficult, as it exists as a single paper copy in the Cambridge University library, which can only be read onsite. The articles arising from the study have a strong psychology focus and much emphasis is placed on quantifying psychological measures of distress and grief. This provides a strong evidence base from which to understand the impact of the diagnosis of a congenital anomaly on women and their partners, yet is less helpful in terms of understanding their decision-making processes.

There are also a small number of articles arising from data collated by ARC (Fisher, Lafarge 2015, Fisher 2008, Fisher 2013b). A total of 361 women were recruited through the organisation. All had experienced a loss following diagnosis of a congenital anomaly and had subsequently sought support from the charity and opted to join their mailing list. Whilst the sample was heterogeneous in terms of indication for termination, method of termination, and gestation at diagnosis, there will be an inherent bias as only women who sought additional emotional support from the organisation are included (Fisher, Lafarge 2015). All the women were asked to complete an online questionnaire and offer comments. Subsequently a number of women were invited to participate in an interview. The online questionnaire was designed to explore the women's choices, particularly in terms of method of termination. Despite the caveats, the data generated provided some insights into the care parents received during the termination process.

In addition to the considerable contribution of the UK groups identified above, there are a number of publications arising from studies undertaken abroad that provide further insight into the experiences of parents. There are several authors who have focussed on how parents make sense of their experiences following a diagnosis of a severe anomaly (Sandelowski, Jones 1996b, Sandelowski, Jones 1996a, Sandelowski, Barroso 2005, Lalor, Begley *et al.* 2009, McCoyd 2007,

McCoyd 2003), whilst the findings from a number of quantitative studies focus on the grief reactions of women (Davies, Gledhill *et al.* 2005, Hunfeld, Wladimiroff *et al.* 1993, Iles 1989, Iles, Gath 1993, Kersting, Dorsch *et al.* 2005, Kersting, Kroker *et al.* 2009, Korenromp, Page-Christiaens *et al.* 2007, Korenromp, Page-Christiaens *et al.* 2009, Salvesen, Øyen *et al.* 1997, Wool 2011, Zolese, Blacker 1992). The importance of context will be raised again in respect to much of this last group of literature.

What do these studies, together, tell us about the experiences of parents? It is clear that the diagnosis of a congenital anomaly and the subsequent decision that parents face is a traumatic event with potentially significant and lasting emotional impact, whether parents decide to continue or terminate the pregnancy (Hunfeld, Wladimiroff *et al.* 1993, Statham, Solomou *et al.* 2001).

In terms of parental experience of the decision-making process itself, the literature available is limited, and predominantly highlights issues that parents take into consideration when making the decision. These include balancing the welfare of the baby, the impact on siblings, and their own ability to cope (da Costa, de Lourdes *et al.* 2005, Menahem, Grimwade 2003, Korenromp, Page-Christiaens *et al.* 2007, Rajaratnam, Marcus *et al.* 2010). In addition, parents express a need to create an 'imagined future', based on their understanding of others' experiences of disability (France, Wyke *et al.* 2011). In turn, this helps them to create a narrative from which they can inform their decision.

'Good care' during the termination process has been explored by the team at ARC (Fisher, Lafarge 2015) where five themes relating to parent's experiences of good care were identified. These included being cared for in a time frame and environment that felt right (not being pressured), receiving the right level of care (including aspects such as sufficient pain control), enabling choices (particularly in relation to choice of method), the role of healthcare professionals and organisations (namely providing information and support), and acknowledging each woman's particular unique circumstance. Following the termination, the added distress caused by the need to make unexpected decisions has also been

highlighted (Hunt, France *et al.* 2009). These decisions might include whether to have a funeral or a cremation, how to remember the baby or, more immediately, whether to hold the baby or not. These are discussions that midwives may find difficult to engage parents in, complicated further by the rareness of the events (Sandelowski, Barroso 2005) and their own feelings of shock and distress (Hughes, Turton *et al.* 2002).

As highlighted, the psychological impact on parents of a diagnosis of a congenital anomaly, and subsequent loss, is significant (Davies, Gledhill *et al.* 2005, Fisher, Lafarge 2015, Hunfeld, Wladimiroff *et al.* 1993, Iles 1989, Iles, Gath 1993, Korenromp, Page-Christiaens *et al.* 2007, Korenromp, Page-Christiaens *et al.* 2009, Lafarge, Mitchell *et al.* 2013, Lalor, Begley *et al.* 2009, McCoyd 2007, McCoyd 2003, Statham, Solomou *et al.* 1999, Statham, Solomou *et al.* 2001, Salvesen, Øyen *et al.* 1997, Wool 2011, Zolese, Blacker 1992, Williams, Munson *et al.* 2008). 'Chosen loss', where loss is experienced as a direct result of a personal decision, remains a minimally explored area (McCoyd 2007), with the findings of one small study exploring the views of women who continued with an affected pregnancy, suggesting that the grieving process may be 'easier' for those that continue as the regret associated with termination is avoided (Chitty, Barnes *et al.* 1996), and the stigma attached to termination is avoided (Norris, Bessett *et al.* 2011). However, there are a number of studies which provide a comparison and measurement of grief reactions following the decision to terminate an affected pregnancy, and pregnancy loss through stillbirth or late miscarriage (Iles, Gath 1993, Hunfeld, Wladimiroff *et al.* 1993, Salvesen, Øyen *et al.* 1997). Of these studies, only Salvesen *et al.* measured grief in the first few days following the event, highlighting that women who terminated their pregnancy had a lower measure of immediate grief reaction (Salvesen, Øyen *et al.* 1997), although after the first four weeks there was no difference between the two groups (Iles, Gath 1993, Hunfeld, Wladimiroff *et al.* 1993, Salvesen, Øyen *et al.* 1997). Clinical reports suggest that women who terminate often feel they are not entitled to their grief (Doka 1989). Yet the grief experienced in this context may be the most challenging due to the need to overcome the 'mythical expectations' in order to access adequate support (McCoyd 2007). Within the literature available a number of predictors of grief

complications were identified: high levels of doubt during decision-making, strong religious beliefs (if terminating), and advanced gestational age (Korenromp, Page-Christiaens *et al.* 2007, Statham, Solomou *et al.* 2000). Davies, however, concluded that older gestation at termination was only a risk factor in the first few months, after which the influence of gestation at loss disappeared (Davies, Gledhill *et al.* 2005). This sample had a high loss to follow up in the second trimester group, which may have impacted on the findings. Younger women, poor social support, stigma and multi-parity have been highlighted as risk factors for complicated grief reactions (Zolese, Blacker 1992, Statham, Solomou *et al.* 2000). Lack of partner support was identified by a number of authors as significant (Black 1989, Korenromp, Page-Christiaens *et al.* 2009, Statham, Solomou *et al.* 1999); in addition low socioeconomic status was recorded as a risk factor (Statham, Solomou *et al.* 2001). Conversely, active decision making appears to protect from high levels of regret and grief (Smith, Dixon *et al.* 2009) and it has been hypothesised that externalising responsibility for decision-making also protects from psychological risk (Sandelowski, Jones 1996a).

No identifiable literature explores the potential for regret at not having terminated. However, literature pertaining to parents of children with Down's Syndrome, suggests that a small percentage of parents regretted continuing with the affected pregnancy (Skotko, Levine *et al.* 2011). Exploring regret following the decision to continue an affected pregnancy is likely to be intensely complex due to the way in which regret for the decision can be interpreted as a rejection of the infant. The difficulties and hardships encountered by parents who continued with affected pregnancies is perhaps a better marker of 'regret' (Tavormina, Boll *et al.* 1981, Robinson, Jackson *et al.* 2001, Kazak, Marvin 1984, Kirk 1998).

Whilst the majority of the literature explores the experiences of women, very little extends to the perspectives of the father, despite his presence being one of a small number of factors that positively impacts on the emotional wellbeing of the women (and the men) after a termination (Statham, Solomou *et al.* 2001, Korenromp, Page-Christiaens *et al.* 2007). Pregnancy has been described as a transitional or liminal state, as much for men as women, with men often feeling

vulnerable, excluded and redundant (Draper 2003). Coping mechanisms identified in the narratives of couples after a termination have suggested that some men channel their anxieties into active information gathering (Locock, Alexander 2006). In addition, their actions may take a firm, directional line, with some couples reporting that the man appeared to feel a greater degree of certainty about the decision. However, this could result in tensions between the couple where the woman was left feeling pressured and rushed (Locock, Alexander 2006).

Research on disclosure, and information on whether or how to disclose in the context of termination for a congenital anomaly, is sparse. Interviews on the healthtalk website provide some insight and have been synthesised in an article based on secondary analysis (France, Hunt *et al.* 2013). None of those interviewed completely concealed; most disclosed selectively, telling close friends and family they had terminated while they told acquaintances they had miscarried. Guilt and fear of being judged were common reasons for avoiding full disclosure, although taking time off work and needing practical or emotional help were reasons for disclosing.

For women who continued with the affected pregnancy, specific difficulties were identified. In some cases the option to terminate was lost due to the lateness of diagnosis and parents have been described as experiencing a 'choice lost', where they regretted not learning of the anomaly sooner (Sandelowski, Jones 1996a). Furthermore, the postnatal narratives of a group of mothers whose pregnancies were affected by a lethal anomaly highlighted that antenatal diagnosis was beneficial (Matthews 1990). Both these studies were undertaken in the US, and although it provides an insightful analysis of how parents make sense of their decision, the findings may be less applicable in England, where, unlike the US, there is no gestational limit for a termination following diagnosis of a severe congenital anomaly or differential access to health services.

As with the tendency to assume that refusal of screening reflects an opposition to termination (Rapp 1998, Green, Snowdon *et al.* 1993), there is some suggestion that some women who continue an affected pregnancy do so because the option of

termination is something with which they are unable to engage (Sandelowski, Barroso 2005). Although this is not always the case, as highlighted by a case study from the UK (Locock, Crawford *et al.* 2005), a 'disowned choice' has been observed, where parents have suggested that for moral or religious reasons the choice was not theirs to make (Sandelowski, Jones 1996a). Importantly, some parents appear unable to make a decision, and thus continue the pregnancy essentially by default (Chaplin, Schweitzer *et al.* 2005). Again this was a US study, where anti-abortion rhetoric potentially has a greater influence on parents' decisions. In this study, a small group of women whose pregnancies were affected by spina bifida engaged in prolonged decision-making processes and, unable to decide, they essentially 'drifted' into continuing with the pregnancy. As highlighted in Table 1-2, spina bifida is associated with a high degree of uncertainty in terms of prognosis, and significant variability in terms of outcome. Much of this relates to the positioning of the spinal lesion. It is unclear from the study how severe the lesions were and therefore what the likely outcome of the pregnancy would be. As has been discussed in section 2.2, severity of the lesion is likely to influence the decision-making process and subsequent decision.

A further study exploring the experiences of women who continued an affected pregnancy describes the time after the anomaly was suspected as paradoxical, with positive and negative consequences to the knowledge experienced (Hedrick 2005). Whilst time is good, in that the passing of time presents the opportunity for more information and an end to uncertainty, it is also the enemy, as the end of the pregnancy draws closer, and the safety provided by the pregnant state ends. The intense grief experienced by parents when they first received the diagnosis was also reflected upon as a paradox. Whilst they mourned the loss of their 'perfect' baby, many parents employed coping strategies, which they subsequently reflected upon as character building or strengthening. The final paradox arose from the knowledge that the baby had an anomaly and although that instilled an intense loss, bonding with the unborn baby meant that he or she was still theirs. Although this study included a number of different anomalies, none of them were lethal. Whether these same themes would represent the sentiments of parents, whose pregnancies were affected by a severe congenital anomaly, is unclear.

Women who continue a pregnancy affected by a severe or lethal anomaly have also been reported to find social situations difficult to manage (Smith, Dietsch *et al.* 2013), in particular, dealing with being 'public property' when the fetus had a severe or lethal anomaly. Women were reported to develop coping strategies, such as avoiding disclosure, or providing avoidant or minimal answers, or avoiding social situations. Adding to these difficulties are reports that some parents found variability in the capacity of friends and families to provide emotional or practical support (Chaplin, Schweitzer *et al.* 2005). The issue of support was pertinent for all parents, irrespective of their decision.

2.3.1 SUMMARY

Decision-making following diagnosis of a severe congenital anomaly remains a largely unexplored area, and the experiences of parents relatively neglected. Resources such as the healthtalk website provide an excellent opportunity for parents and clinicians alike to gain some insight into how parents experience the decision-making process. Secondary analysis of the interviews has shed light on a number of aspects of the care pathway; however, to date nothing has been published addressing the question of how women and their partners make the decision to continue or terminate a pregnancy following diagnosis or suspicion of a severe congenital anomaly. Whether or not secondary analysis of these data could be used to address the research question formed part of my decision-making in relation to methods proposed.

What has been more widely explored is the psychological impact of loss in the context of a congenital anomaly. In practice, this assists practitioners in identifying women who are at high risk of complicated grief reactions, but is perhaps less useful in addressing the question of how parents experience grief. Whilst McCoyd *et al* go some way to addressing this; the perspectives are based on a US context. Whether the same perspectives would be applicable within the UK setting is unclear. As previously discussed, the abortion debate in the US is far more prominent than in the UK and the impact of the legal restrictions on the participants should perhaps be given some consideration.

Overall, when addressing the question of parental experiences of the decision-making process, small areas of literature exist that go some way towards providing an understanding of the varied experiences of parents. Nonetheless contextual issues and biases exist within the literature available, resulting in ongoing uncertainties and gaps in our understanding.

2.4 WHAT ARE THE EXPERIENCES AND VIEWS OF HEALTHCARE PROFESSIONALS?

The experiences and views of healthcare professionals within the context of decision-making following suspicion or diagnosis of an anomaly is again an under-researched area. Much of the literature available pertains to antenatal screening and highlights a variety of ethical issues including non-directiveness in consultations (Aite, Zaccara *et al.* 2011, Williams, Alderson *et al.* 2002b, Press, Browner 1997, Al-Jader, Parry-Langdon *et al.* 2000, Heyman, Hundt *et al.* 2006), which closely ties in with the concept of shared decision-making (Elwyn, Edwards *et al.* 2000, Charles, Gafni *et al.* 1999, Edwards, Davies *et al.* 2009, Légaré, Ratté *et al.* 2008), giving balanced information on anomalies (Anderson 1999, Farsides, Williams *et al.* 2004, Williams, Alderson *et al.* 2002a) and the practical and ethical issues of screening (Ekelin, Crang-Svalenius 2004, Hey, Hurst 2003, Heyman, Hundt *et al.* 2006, Williams, Alderson *et al.* 2002a). In addition, there is some evidence that examines the interactions between clinicians and patients within the antenatal setting (Pilnick, Zayts 2014, Pilnick 2008, Pilnick, Zayts 2012, Pilnick, Fraser *et al.* 2004).

Non-directiveness, as a concept, has evolved from a narrow definition of what should not be done, to a broad definition that promotes active counselling skills in support of patient autonomy and informed decision-making (Weil 2003). Although initially a response to the abuses of human genetics in the early 20th century (Clarke 1997), it also reflects changes of power within the doctor-patient relationship, where a move away from a paternalistic view of medicine is being seen (O'Neill 2002). A key debate in the literature revolves around the feasibility

of a non-directive approach, and whether adherence to its principles offers patients 'good care' (Mol 2008, Schwennesen, Koch 2012).

Shared decision-making extends the concept of non-directiveness to include the broader principles of patient control, autonomy and presents as a challenge to clinicians' authority (Charles, Gafni *et al.* 1999). Although shared decision-making is only one of a number of patient/clinician decision-making models discussed within the literature (Levine *et al.* 1992), this model is derived from the same principles ensconced in the concepts of 'informed choice' and 'informed consent' (Sutherland *et al.* 1989) and is increasingly advocated as the 'ideal' decision-making model within a medical encounter (Légaré, Ratté *et al.* 2008). Nonetheless, the literature is well versed in the difficulties associated with the implementation of shared-decision making (Elwyn, Edwards *et al.* 2000, Elwyn, Edwards *et al.* 2003, Edwards, Davies *et al.* 2009). In particular, the difficulties in advocating a single route to decision-making, where patient and clinician preferences can vary and change with time (Charles, Gafni *et al.* 1999). Underlying issues resonate clearly with those reported by clinicians in the literature pertaining to non-directive counselling.

A number of the publications pertaining to non-directiveness in particular, are based on a large-scale project entitled 'Cross Currents in Genetics' funded through the Wellcome Trust Biomedical Ethics programme (Heyman, Hundt *et al.* 2006, Farsides, Williams *et al.* 2004, Alderson, Williams *et al.* 2004, Williams, Alderson *et al.* 2002a, Williams, Alderson *et al.* 2002b, Williams, Alderson *et al.* 2002c, Williams, Alderson *et al.* 2002d). The project involved 70 semi-structured interviews and 11 two-hour seminars with a variety of staff working in a teaching hospital and a district general hospital (DGH). The staff recruited worked in a variety of specialities including obstetrics, gynaecology, paediatrics and neonatology and consisted of groups of doctors, nurses and midwives of varying grades and seniority. Despite the genetics slant of the title of the over-arching project, this was generally un-reflected in the papers arising from it, whose focus was on antenatal screening, and, apart from the article by Heyman *et al.*, not specific to chromosomal anomalies. The scale and methodology of this study gives

an unparalleled insight into the perspectives of clinicians and the difficulties encountered when providing antenatal counselling to parents. Combining a sociological and ethical approach to the investigation created opportunities for healthcare professionals to highlight the paradoxes that exist within the field of antenatal screening. In addition, the authors commented that the rubric of an 'ethics seminar' attracted a wide range of practitioners who may not otherwise have participated (Alderson, Williams *et al.* 2004).

The daily moral dilemmas involved when considering the rights and wrongs of termination; the nature of the maternal-fetal relationship and establishing the moral status of the fetus; and the questioning of at what point *severe* might become incompatible with a *reasonable* quality of life create an emotive environment. For professionals, managing these dilemmas alongside the "*interface between their professional and private moral values*" (Farsides, Williams *et al.* 2004, pg.505) is complex. In addition, the pressures to commit to a 'non-directive' counselling approach left little room for support and debate. The context provided by this work is fundamentally important for my project as it provides an in-depth understanding of the dilemmas encountered by clinicians when counselling pregnant women and their partners. A further study (part of the Cambridge Maternal Study) involving interviews with 15 clinicians was undertaken by Statham *et al* when investigating ethical dilemmas in antenatal care (Statham, Solomou *et al.* 2006, Statham, Solomou *et al.* 2003). This focused primarily on attitudes to termination legislation and the difficulties encountered in ensuring they worked within the current termination legislation. A shift in attitudes as to what constituted a severe anomaly was noted at the time, where the definition threshold had been raised by clinicians and the rationale for this unclear. Subsequently, recommendations were made that this should be revisited in order to monitor changes. This research was carried out around a decade ago, and no subsequent studies have been identified.

Some insight into the interaction between parents and clinicians in the antenatal setting is gained from studies by Alison Pilnick and colleagues (Pilnick, Zayts 2014, Pilnick 2008, Pilnick, Zayts 2012, Pilnick, Fraser *et al.* 2004). Video recordings of

antenatal consultations were used to capture the interactional processes through which choice, in terms of antenatal screening and subsequent decision-making, was negotiated, then established or contested. Analysis was undertaken using conversation analysis, a method that seeks to investigate how participants understand and respond to one another. Unlike discourse analysis, the central focus of conversation analysis is how sequences of actions, rather than rhetorical organisation, occur (Silverman 2006). Particular interest is therefore taken in how participants turn-take, repeat, interrupt etc. within naturally-occurring talk. This subsequently provides an insight into how the doctor/patient relationship is enacted in practice. Previous work has demonstrated how doctors' perceptions of patients' social characteristics result in varying stances towards lifestyle issues (essentially stereotyping) (Strong 2001, Silverman 1987, Lutfey, McKinlay 2009). Similar interactions were demonstrated by Pilnick, where use of conversation analysis to explore the interactions between clinicians and parents highlighted how socioeconomic factors were interpreted by clinicians and subsequently influenced decision-making (Pilnick, Zayts 2012). Although no overarching differences were noted in the way that the interactions were structured between women from different socioeconomic groups, when socioeconomic characteristics were made visible within the consultation the extent to which clinicians allowed these to impact on decision-making differed (Pilnick, Zayts 2012). Capturing these dynamics between clinicians and parents within my study was therefore likely to provide a different perspective on the decision-making process. This was kept in mind when designing the study.

2.4.1 SUMMARY

Although there is a limited body of evidence examining parents' decision-making within the antenatal setting, as was highlighted in Reid *et al's* meta-synthesis, the views and experiences of clinicians pertaining to decision-making in this setting are proportionally well represented (Reid, Sinclair *et al.* 2009). The issues pertaining to non-directive counselling and informed, shared decision-making feature prominently in the literature, with the tensions and difficulties encountered by clinicians well documented (Aite, Zaccara *et al.* 2011, Williams, Alderson *et al.* 2002b, Press, Browner 1997, Al-Jader, Parry-Langdon *et al.* 2000,

Heyman, Hundt *et al.* 2006). The 'hardening' in clinicians' attitudes in relation to what constitutes a severe anomaly has also been highlighted, and despite recommendations that changes in attitudes should be monitored, no further evidence examining this aspect has been uncovered (Statham, Solomou *et al.* 2006, Statham, Solomou *et al.* 2003).

The complexity of the doctor-patient relationship and the impact of the interaction between the two parties have also been identified as being instrumental in the decision-making process (Pilnick, Zayts 2012). In terms of addressing the overarching question posed, this highlighted the need to give consideration to exploring the topic from multiple perspectives, and potentially capturing elements of the interaction between clinicians and parents.

2.5 DECISION-MAKING THEORY

Having explored the empirical literature, I subsequently expanded my review to examine the theoretical aspects of decision-making. There is an extensive literature pertaining to decision-making which focuses predominantly on the way individuals, groups and organisations arrive at judgments and decisions, particularly in situations involving risk and uncertainty. Theories and findings from decision-making research have been examined and applied extensively in areas such as finance and economics, marketing, health (from the perspective of patients and clinicians), politics and law, and in management and organisational and institutional development. As a result there are multiple perspectives on the subject, a large volume of literature, and a range of conflicting findings and recommendations. A search on the Scopus database, using the terms 'choice' and 'decision-making', but restricted to humans and English language and published in the last 20 years, produced over 490,000 results. As discussed in section 2, combined word searches were undertaken within which decision-making and the antenatal period were combined; however, this precluded access to the wider decision-making literature base, and indeed failed to identify any literature specific to the decision-making process in the context of diagnosis of a severe congenital anomaly. The method proposed in section 2, although successful in

identifying empirical literature, provided an incomplete perspective on theoretical decision-making and therefore an alternative method of literature identification was sought for this aspect.

Through internet searches, I identified a number of senior UK university academics with a special interest in decision-making who had published extensively about decision-making theories. Having approached them directly for some guidance, they highlighted a number of areas I had not explored, including the work of Klein on naturalistic decision-making (Klein 1993). Although potentially a less systematic approach, there is an empirical evidence base to support the effectiveness of using 'experts' as a resource for identifying literature (Papaioannou, Sutton *et al.* 2010, McManus, Wilson *et al.* 1998). There was an overall consensus that conventional searches are limited in their effectiveness when identifying key theories within this field. This is partly due to the volume and diverse specialties included, but also the idiosyncratic ways in which much of the theory is presented. For example, Kahneman's debates with Amos Tversky outlined in 'Thinking Fast and Slow' (2011), reflect the richness of the theory and the diversity of thinking about concepts such as 'understanding', 'validity' and 'intuition' (Kahneman, Tversky 2011). In addition, many of the original papers presenting theories were first published in the 1960's and thus may have been missed through traditional search methods, although it is likely some might have emerged through citation or reference searching.

Articles identified were hand-searched for further references. This provided a starting point from which I could develop an understanding of the theoretical underpinnings of decision-making. The issue of suitable methods for searching for appropriate literature relating to decision-making is not unique to my experience. A scoping study by York University used similar methods to search for an appropriate theory to apply to a study on decision-making within the context of disability (Beresford, Sloper 2008). A number of the academics identified by this group correlated with those I had approached prior to identifying the paper. This provided some endorsement to my approach.

2.5.1 *RATIONALISING THE LITERATURE*

The heterogeneity and the vastness of the decision-making literature made it essential to identify literature that could be applied to this study. First, I undertook a broad overview of the available literature. The questions posed within the literature varied considerably. A way of classifying the different conceptual and methodological approaches applied has been classified in terms of ‘utility’ (Beach, Lipshitz 1993), i.e. the subsequent application of the research. The three categories identified were:

- *Descriptive – why and how decisions are made the way they are*
- *Normative – how decisions should be made in some ideal sense*
- *Prescriptive – how decision-making can be made more effective*

In addition to the utility, there are distinct differences in the ways the theories were conceptualised. This becomes clearer with an understanding of the historic development of decision-making theory.

2.5.1.1 *HISTORICAL PERSPECTIVE*

Decision-making theories and models can be traced back centuries to the study of simple gambles. First attempts to describe and explain people’s decisions were documented as early as the 18th century, when Bernoulli, a Swiss mathematician, attempted to explain people’s aversion to risk (Kahneman, Tversky 2011). This concept was further developed in the 1800’s by Fechner, a German philosopher and physicist, who pioneered the study of ‘psychophysics’, an evolving study of the relationship between the subjective quantity in the observer’s mind to the objective quantity in the material world. This subsequently evolved into the science of psychology (Benjamin Jr 2007). The study of decision-making theory has subsequently become an area of academic study in its own right (Klein 1993).

One of the greatest divides in the development of decision theory occurred in the early 1900s with the development of experimental psychology (Hammond 1993). At this point, the teachings of two leading psychologists, Wundt and Brunswik, divided. Whilst Wundt argued for the controlling of variables in order to study the phenomenon of interest, the Brunswikan perspective supported the argument that

the uncertainty and irregularities of the environment were the phenomenon of interest (Bergmann 1952). Whereas Wundt sought a clean, clinical environment in which to direct studies (i.e. the laboratory), Brunswik supported study within the natural environment (Araujo, Davids *et al.* 2007). This highlighted differences not only in the way in which problems were studied, but also in the way they were conceptualised. The distinctions arising from these differing traditions have been labelled as: empiricism, relativism, and naturalism (Cohen 1993b).

Empiricism relates to models based on empiricist or positive assumptions, where decisions that fail to meet the predetermined outcome of the model are deemed 'irrational'. In other words, if the decision outcome does not fit that within the model, the decision is invalid or lacking in reliability. This body of literature is often correlated with the 'normative' category identified above, where the search for an ideal decision-making process relies on the assumption that there is a 'correct' decision.

The relativists attempt to explain actions when, for example, there are failings in the decision-making process or in the logic applied by the decision-maker. One of the best examples is that of Kahneman and Tversky's paper on heuristics and biases where biases are seen as deviations and used to explain variation from the model (Tversky, Kahneman 1974). They promote the concept that there are a number of different biases, all of which arise from the way we process thoughts. Within the context of risk and decision-making, their Prospect Theory highlighted biases that are created by 'framing'. For instance, one experiment explored how phrasing affected participants' choice of medical treatment (Tversky, Kahneman 1981). By framing a treatment positively (33% of recipients would live), or negatively (66% of recipients would die), participants' responses differed significantly. Whilst 72% opted for the option when positively framed, only 22% accepted the option when framed negatively. The so-called framing bias is subsequently used to explain the deviation from the 'rational' decision.

Finally, the naturalists seek to investigate decision-making from a descriptive perspective, with the understanding that there is no pre-ordained and objective

rationality involved. Much of the work within this perspective has developed within what is considered to be the natural environment i.e. as opposed to a controlled laboratory setting. Examples of this work include exploring firefighters' decision-making during fires how do they 'know' when a building is not safe to enter? Similar aesthetic knowledge and decision-making has been identified in experienced healthcare professionals, where they 'know' that there is something wrong, where the 'knowing' is a form of internal understanding, which may not initially be supported by empirical data, but has been gained through experience (Carper 1978).

2.5.1.2 RATIONALISING FURTHER

There is a gradual move away from the conventional empiricist research doctrine in relation to decision-making (Hammond 1993), which can be seen in the similarities developing between the models stemming from different approaches. For example, empiricists talk about a dual system approach of analytical and intuitive thinking while those from the naturalistic camp refer to a continuum between analytic and intuitive thinking. When considering my research question, I have four over-arching concerns that relate to the empiricist and relativist paradigms:

- *Whether the assumption of rationality is appropriate i.e. is there a right or wrong decision to terminate or continue an affected pregnancy?*
- *Where rationality does not occur, the need to identify a reason to explain this (bias) i.e. where parents make different decisions, is it appropriate to assume that one is right and the other wrong, and thus try to explain why they are different?*
- *The importance placed on the decision outcome, rather than the cognitive process undertaken to achieve the outcome i.e. is it more important to understand how parents make their decision or to focus on the decision that they made?*
- *The ability to study the phenomenon separately from the context i.e. is it feasible to explore the decision-making*

processes of women and their partners within an artificial setting?

The assumption of empiricist and relativist decision-making theory is that the decision-maker is rational; an assumption that has little empirical support (Brehmer 1984, Christensen-Szalanski 1986). Furthermore, the perception of 'rational' in terms of the 'right' decision or judgment sits uncomfortably in relation to decision-making following diagnosis or suspicion of a severe congenital anomaly. Indeed the decision itself is purely an outcome of the decision-making process. Attempting to apply 'rationality' to this process assumes a fixed reality, something that I will go on to argue against (see section 3.3). Secondly, the relativist stance conveys the need for conformity where rationality does not occur. This is achieved through the identification of 'biases'. There is evidence to suggest that the existence of biases in the process may not merit correction. One study investigated the information that doctors were giving patients in order to make decisions (Christensen-Szalanski, Boyce *et al.* 1987). Doctors performing circumcision operations over-emphasised rare risks, but under-estimated common risks (the bias was in the manner in which the information was imparted). However, elimination of this bias did not change the number of infants who had circumcision but made parents feel less secure with the clinicians and more resentful. This suggests that correcting biased processes may not always change decision outcomes. Although correction of biases may be justified in certain circumstances, consideration of the impact will be required.

Both empiricist and relativist studies demarcate the decision outcome as the outcome measure of importance. Intrinsically the descriptive processes of the naturalistic school of thought favour instead the cognitive processes through which the decisions are made. Therefore, in the context of my study, the focus would remain on the decision-making process rather than the decision itself. Finally, the impact of studying the phenomenon within the 'natural' environment is also a factor in my choice of methodology. For example, a study by Konecni *et al* (1980) studied judges and parole officers in a laboratory and real-life settings (Konecni, Mulcahy *et al.* 1980). The criminal sentences they meted out were compared in the two settings and were demonstrated to be significantly different.

Brunswik himself stated: “*proper sampling of situations and problems may in the end be more important than proper sampling of the subjects, considering the fact that individuals are probably on the whole much more alike than are situations among one another*” (Brunswick 1956, pg.39). Applied to decisions following diagnosis or suspicion of a severe fetal anomaly, Erikson’s work making comparisons across the old German borders clearly highlights the impact and importance of context on the decision-making process.

2.5.2 SUMMARY

The methodology applied to searching for the theoretical decision-making literature differed from that applied to the empirical literature search. This was predominantly due to the need to balance the breadth of the literature with restricting the volume sufficiently to make analysis manageable. Categorisation on the basis of ‘utility’ has been suggested to be an effective mechanism to identify literature relevant to the area of interest (Beach, Lipshitz 1993). Further rationalisation based on the underlying epistemological basis of the theories was made (Cohen 1993b). Descriptive studies from the naturalist paradigm were of particular interest in that they a) were interested in how and why decisions were made the way they were, b) prioritised the study of decision-making processes over the decision itself, c) did not perceive there to be a right or wrong decision and d) identified the importance of context.

Whilst the use of hypothetical scenarios to explore decision-making processes of women provides an insight into social attitudes to termination, they are less reflective of the ‘real-life’ responses of women facing a decision that are influenced by the dynamics of time and complexities of wider contextual factors (Sawyer, Cerritelli *et al.* 2006, Erikson 2003). In addition, the perception of ‘rationality’ in relation to decisions in this context undermines the concept of choice. From a theoretical perspective, these examples illustrate the need to study this phenomenon from a naturalistic perspective.

2.6 DEVELOPMENT OF THE RESEARCH QUESTION

Despite an appreciation that a research question will change several times throughout the process of a PhD, identifying an appropriate starting point was far harder than I had anticipated. As Burck pointed out: *“Without a well-honed research question, framed so that it is possible to carry out, a qualitative researcher is in danger of losing their way and of becoming ensnared in the enormous quantity of detail of the research material”* (Burck 2005, pg. 240) The potential to draw parallels to other areas of healthcare resulted in a constant need to clarify and redefine the boundaries of the study. In addition, the numerous ‘gaps’ in the literature made it particularly difficult to maintain focus.

My starting point was a desire to explore the issues that may underlie the observed variation in neonatal death rates as a result of the decision to terminate or continue a pregnancy affected by a severe congenital anomaly pregnancy, i.e. that women from lower socioeconomic groups are less likely to terminate than those from higher socioeconomic groups (Smith, Budd *et al.* 2011). My literature review showed that there is little research underpinning care provision in this area overall. In particular, there is poor understanding of the decision-making process employed by women following diagnosis of a severe congenital anomaly with little literature available that could be applied to address the issues underlying the observed variation. The need to remedy this in order to ensure that care is tailored to meet parents’ needs is well documented: *“studies specifically designed to evaluate and understand women’s decision-making regarding the termination decision, and how their background, values, and life experience contribute to these decisions, are critical to furthering our knowledge of why women make particular choices and how we, as providers, can better inform and serve their needs.”* (Shaffer *et al.* 2006, pg. 671)

The main purpose of the literature review presented in this chapter has been to refine my research question and subsequently inform the study design. During this process I have considered various aspects of the parents’ journey from antenatal screening to delivery or termination. Overall, there are two important but separate

time points when women make decisions: the first is whether or not to participate in screening and the second is the decision to continue or terminate the pregnancy after diagnosis of an anomaly. For some women, these decisions may appear as a continuum, where making an informed choice about screening includes considering the option for termination. However, the literature suggests that this may not always be the case, with the role of screening often poorly understood, and the concept of risk and diagnosis confused (Williams *et al.* 2002a, Heyman *et al.* 2006). Indeed, the interaction between these two decision-making processes is multifaceted, and use of one as a proxy for the other, or assuming that one choice will be representative of the other, is perhaps an underestimation of the complexity of the process (Ahmed *et al.* 2006). The need to study the decision-making process specific to the decision to continue or terminate the affected pregnancy therefore became apparent. However, the impact of the initial screening decision on the subsequent decision to terminate or continue a pregnancy could not be ignored. Data collection methods needed to be designed to capture this.

The importance of 'context' on the decision-making process is evident, and reflects the influence of external factors such as the legal availability and perceived social acceptability of termination. The need to capture the influence of context within my study was paramount and therefore consideration was given to including data generated from sources other than the pregnant woman. In addition, the theoretical evidence on decision-making has highlighted the need to explore the decision-making processes of women and their partners from a naturalistic perspective. This supports the need to collect data within the clinical environment, in 'real-time', and to focus on the process rather than the outcome. As highlighted in the evidence derived from Pilnick's studies, the outcome of a consultation is reliant on the interaction between clinician and patient (Pilnick, Zayts 2014, Pilnick 2008, Pilnick, Zayts 2012, Pilnick *et al.* 2004). This provides further justification to avoid sole reliance on data derived from narratives, but rather to explore the interactions between clinicians and parents.

The differences between the literature on Down's Syndrome and the FASP anomalies, in terms of findings, suggested caution should be applied when transferring findings generated in studies examining one anomaly to studies exploring others. The inequalities highlighted within the provision of antenatal screening, uptake, and subsequent anomaly identification, depending on level of deprivation, raised the potential that the differences in mortality outcome start much earlier in the care pathway in the case of Down's Syndrome. I therefore felt that Down's Syndrome should be excluded from this study as unexplained variations between Down's and the FASP anomalies would be likely to complicate and confound findings.

Alongside the evidence associating socioeconomic status with the decision to continue or terminate a pregnancy affected by a severe congenital anomaly, review of the literature identified a significant number of papers highlighting other associated variables. These included maternal age, type of anomaly, uncertainty attached to diagnosis or prognosis, gestational age at diagnosis and ethnicity. Although consensus over the impact or direction of impact was often lacking in the literature, this may have been attributable to the variations in context in which the studies were undertaken. Apart from further supporting the need to consider contextual factors in the design of the study, identification of these variables enabled me to give consideration to the development of a sampling frame in which to recruit participants. The benefits of this lay in ensuring heterogeneity of the sample, which in turn ensures a wide ranging transferability of findings (Malterud 2001).

The literature presented in this chapter has proved invaluable in contextualising the work undertaken for my PhD as it has provided a starting point to understanding the experiences of women and their partners. Nonetheless, much of the emphasis is placed on variables associated with the decision or parental and clinician experience, but has not been extended to address the process through which parents make the decision to continue or terminate an affected pregnancy. A clear gap remains in our understanding of the decision-making processes enacted by women following a diagnosis of a severe anomaly. In addition the

influence of socioeconomic status on that decision remains unexplored. In order to gain some insight into the reasons for the socioeconomic inequalities in neonatal mortality seen in babies born with severe anomalies, this is an area that requires addressing. The research question posed is therefore:

How do women and their partners make the decision to continue or terminate a pregnancy following suspicion or diagnosis of a severe congenital anomaly?

The influence of wider contextual factors, and in particular socioeconomic status, on the decision-making process is of particular interest.

3 METHODS

Undertaking a study involving highly sensitive issues, such as this, has been at the forefront of my mind from the outset. There are not only concerns over the potential unearthing of highly emotional and personal information, but also the questionable right of a researcher to intrude into a family's grief. In order to overcome some of these issues, I identified a number of parents and clinicians who have advised me throughout the research process. Within the thesis they are referred to as the Patient and Public Involvement (PPI) group (see Appendix B). As Noland argues, it is essential to have an insight into how participants will respond to being recruited to a study, particularly when researching sensitive topics (Noland 2012). The engagement of parents, who had experienced decision-making in this context, at an early stage of the study design process, enabled me to filter ideas through their perspective. Active involvement in this way can lead to research of greater quality and relevance because of the unique contribution that users can make (Brett *et al.* 2014). The consultation work I undertook has been instrumental in influencing all aspects of the study, from relevance of the question to feasibility and acceptability of the design, to undertaking the research and disseminating the findings. The voices of the PPI group resound throughout this chapter as decisions and choices made in relation to the research methods for this study are explained.

The chapter is divided into six sections. First, lessons learned from the literature are re-iterated. Second, my research perspective is outlined and discussed, including delineation of the ontological and epistemological stance adopted, and rationale for adopting a qualitative methodology. This is extended in the third section, where my approach to data collection is presented including: determining who was the subject of interest, a discussion of the research methods considered and adopted, highlighting the practical issues faced, and a critique of the choices made, and a description of the sampling strategy including a reflection of the recruitment strategy employed. The fourth section comprises a detailed account of

the management of the data, including discussion of issues pertaining to privacy and confidentiality, and practical aspects such as transcribing. Fifth, my approach to data analysis is presented and critiqued. The sixth section gives consideration to the methodological quality and rigour of the study, and incorporates issues relating to the credibility, dependability and transferability of the findings.

3.1 LESSONS FROM THE LITERATURE

The available literature has been very useful in determining both the need for the study (as highlighted in the previous chapter) and the subsequent study design. Whilst there is clearly a gap in our understanding of parental decision-making following diagnosis of a severe congenital anomaly, I had ongoing concerns that the lack of literature was a reflection not necessarily of the difficulty of addressing the question, but of the appropriateness of doing so. Alongside the advice taken from the PPI group, I sought additional evidence from the literature specific to the appropriateness of recruiting parents around the time of diagnosis.

There is a small number of articles which explore issues arising from decision-making following diagnosis of a congenital anomaly: information requirements and counselling (Menahem, Grimwade 2003); timing of termination decisions (Gawron *et al.* 2013); and narratives of choice (Sandelowski, Jones 1996a). Despite their differences, they are of particular methodological interest due to the similarities in the timing of their recruitment, with all the women recruited within a few days of termination or birth. This set a precedent in relation to the feasibility and appropriateness of approaching women at a vulnerable time. The recruitment rate for Gawron's study was 30 of 34 women approached, with the participants invited to participate in a 60 minute interview on the first day of a two-day termination process. The authors suggested that this high recruitment rate, despite the timing of the interview, was a reflection of the women's desire to express their feelings (Gawron *et al.* 2013). Menahem's study involved completion of questionnaires post-termination or post-delivery rather than interviews. However, the author also commented that several of the women approached stated they were grateful for the opportunity to respond (Menahem, Grimwade

2003). Two women (out of 40) refused to participate, with one expressing anger at having been approached, and the other unsure why she had refused. Similarly high recruitment was identified in work by Sandelowski, with subsequent correspondence with the author suggesting that recruitment was unproblematic and gaining ethical approval uneventful (Sandelowski, Jones 1996a). These studies were all undertaken in either the United States or Australia. The impact of context is important in so far as the termination law, particularly in the US, is less fluid than in the UK. In addition, they were all carried out in private medical facilities. The high recruitment rates may be partially attributed to the demographics of the women attending private facilities, with literature suggesting that patients of high socioeconomic status are more likely to participate in research (Ford *et al.* 2008, Gross *et al.* 2005, Heinrichs *et al.* 2005). Therefore, although the findings may not be directly transferable, the feasibility and appropriateness of approaching parents at a vulnerable time has been tested.

With this reassurance, I have applied the lessons learned from the literature to designing a study to examine the decision-making process of parents following diagnosis or suspicion of a severe fetal anomaly. The lessons learned are summarised below:

- *Decision-making regarding whether or not to undergo antenatal screening may not reflect post-diagnosis decision-making.*
- *The context in which the decision-making process is enacted is important. This includes issues such as laws and policy governing termination, as well as social context in terms of perceived acceptability of termination.*
- *Hypothetical decision-making does not necessarily reflect 'real-life' decision-making.*
- *Down's Syndrome differs from the FASP anomalies in important ways.*

- *There are a number of variables associated with the decision to continue or terminate an affected pregnancy: capturing these within the sample requires consideration.*
- *Patient-clinician interactions are important, and therefore capture of ‘real-time’ decision-making will be beneficial.*

These lessons have been instrumental in guiding methodological decisions throughout my study.

3.2 RESEARCH PERSPECTIVES

As a relatively novice researcher, I have sometimes found elements of the terminology used in research literature confusing. Reassuringly, my experience may not be unique, with terms such as ‘epistemologies’, ‘theoretical perspectives’ and ‘methodologies’ often “*thrown together in grab-bag style as if they were all comparable terms*” (Crotty 1998, pg.3).

Debates about the nature of the social world and what can be known about it (ontology), the nature of knowledge and how it can be acquired (epistemology), and how we can study it (methodology) underpin the different approaches adopted by researchers (Ritchie *et al.* 2014). The rationale for choice of one approach over another is directed not only by the philosophical beliefs and understanding of the researcher, but also by the need for research methods and strategies to fit the context of the research (Ritchie *et al.* 2014, Mason 2002, Patton 2002, Seale 1999a, Seale 1999b).

3.2.1 ONTOLOGY

The debate surrounding the nature of reality has been shaped by two overarching ontological positions, namely realism and idealism, between which the existence, or lack, of an external reality (outside our independent beliefs and understandings) is contested (Ritchie *et al.* 2014). My research perspective aligns with the belief that an external reality exists, but is only known through the human mind and socially constructed meanings, a stance termed ‘subtle realism’ (Hammersley 1992, Blaikie 2007). This approach has been described as a “*sensible*

pragmatism that assumes reality is filtered through various lenses, but that it is none the less not infinitely malleable, and that it is, to an albeit limited extent, knowable" (Murphy, Dingwall 2003). This offers an attractive compromise between the perspective of what Lincoln and Guba call 'naïve realism', which supports the notion that reality can be observed directly and accurately (Lincoln, Guba 1985, pg. 290), and that of relativism or radical idealism, which argues that there is no shared social reality, only a series of individual constructions (Ritchie *et al.* 2014, Madill *et al.* 2000). For me, the perspective of the subtle realist is particularly compelling as it acknowledges an external reality, thereby supporting the notion that people's perceptions and experiences are influenced by forces outside their control, whilst acknowledging that people will differ in the way they make sense of their everyday reality (Madill *et al.* 2000). Within the context of this study, this perspective is consistent with the holistic view of health as a "*complex mix of social, economic, political and economic factors*" (Baum 1995, pg. 459) as well as giving validity to the varying perceptions of clinicians and parents. These ontological beliefs are reflected in my choice of methods, discussed later in this chapter.

3.2.2 EPISTEMOLOGY

Within the context of this study there are two key epistemological issues that dominate the way in which knowledge is acquired and the relationship between the researcher and the researched, and how this influences the interface between facts and values.

Knowledge can be acquired inductively (bottom up) where patterns are derived from observation of the world, or deductively (top down) where a hypothesis is tested (Shaffer 1989). In this study a broadly inductive approach has been adopted. However, this can result in an oversimplification of the interpretation of the data, with the suggestion that there is no such thing as pure induction or deduction as the data collected and interpreted have not been approached with a blank mind (Blaikie 2007). All researchers are individuals and how questions are conceived and asked, and the responses interpreted, are to a greater or lesser extent filtered through personal biographies and experiences. Hence, even researchers from a 'hard science' background can talk of personal knowledge

arrived at through personal engagement with data and how the relationship between the researcher and the researched is defined. Consequently, it is difficult to subscribe to the view that the researcher can be objective or neutral when the relationship between the researcher and social phenomenon is interactive (Snape, Spencer 2003). A more nuanced view of this relationship has been termed "*empathic neutrality*", a position that argues that research cannot be value free and without bias, assumptions and values (Ritchie *et al.* 2014, pg. 22). As a result, I have attempted to lay these biases, assumptions and values bare.

3.2.3 METHODOLOGY

The subtle realist's focus on representation rather than a search for the 'truth' supports a stance from which either quantitative or qualitative methodologies are arguably appropriate to address the given research question (Mays, Pope 2000). This in turn enables the researcher to take a more pragmatic approach in relation to choice of methodology (Murphy *et al.* 1998).

For the purpose of this study, a qualitative methodology has been employed in order to better understand the complex reality of decision-making. Descriptive, qualitative studies are appropriate when, as in this study, a topic has been subject to limited exploration. Further, the strength of qualitative studies lies in their ability to highlight a range of questions and generate insights far removed from testing normative hypotheses (Mason 2002). The ability of qualitative research to allow for handling subjective insights in depth while concentrating on participants' perspectives, understandings, and subjective views (Sulmasy, Sugarman 2001), and the focus on rich, holistic descriptions (Guba, Lincoln 2005, Murphy *et al.* 1998) and contextual understandings (Popay *et al.* 1998) was particularly compelling.

As discussed in Chapter 2, a significant volume of the literature identified on the issue of decision-making and congenital anomalies stems from the positivist tradition, where variables associated with a particular decision have been highlighted. The fact that there is variation in the findings of a number of these studies may be further indication that quantitative studies are unable to capture

the complexity of the decision-making process. Indeed, exploration of the theoretical underpinnings of decision-making literature highlights the need to investigate decision-making in this context through application of naturalistic methodologies. This further supports the need for a different approach to exploring this phenomenon. Hence a qualitative study has been determined to be the most appropriate to address the research question posed.

3.3 DATA COLLECTION

In order to address the research question posed, the ‘where’ (in respect to the selection of the recruitment site), ‘who’ (in terms of defining the participants), and ‘how’ (in relation to data collection methods) needed to be considered. This section provides a description of each of these three areas in turn. I therefore start by describing how I selected the recruitment sites, and then move on to defining the population of interest and sampling strategies applied. Next, the methods available are then explored, alongside their benefits and limitations in addressing the research question posed. My decision-making process is subsequently described in relation to the choice of methods. Throughout, a description of how my decisions worked in practice is provided, and my reflections upon these choices presented.

3.3.1 *CENTRE SELECTION*

The choice of hospital trusts in which to conduct the research was predominantly pragmatic. However, three inter-relating factors were considered, namely geographical location, sub-specialities offered, and volume of patients.

In relation to location, ease of access to the centre was important in terms of cost (time and financial). Furthermore, inclusion of centres that collated and contributed antenatal data into the regional congenital anomaly register was deemed desirable, with three trusts meeting this criterion. A number of anomaly registers exist across England and Wales. The local register is one of the largest, and captures the data across the region. Selection of this geographical area enabled access to baseline prevalence data pertaining to specific anomalies and

thus allowed for an element of planning in terms of recruitment. I had envisaged that this would also enable a retrospective analysis of the cases 'missed' and discussion on potential biases arising from this. However, at the time of writing, data from the recruitment period have not been validated, and changes currently affecting the registers have meant that accessing these data will be delayed.

The second factor considered was the provision of specialised services, with cardiac and neurological antenatal services falling into this category. These services are centrally rather than locally commissioned and offered at large tertiary referral centres only. Parents with a pregnancy affected by an anomaly requiring these specialities would be referred to the appropriate centre, rather than being cared for in their local hospital. Two of the trusts in the region covered by the register provide such services. The first provides a regional, specialised neurological service and the second a regional, specialised cardiac service. Parents are referred between the two for specialised opinions and subsequent treatment where appropriate. The third and final trust covered by the register was not commissioned as a specialist centre. Inclusion of both specialist centres allowed the option to follow parents throughout their journey, with consultation recordings collated from both trusts. The third trust had a different referral pathway, and commissioned their specialist services from a trust outside the boundaries of the local anomaly register. This made evaluating the numbers of anomalies difficult, and meant seeking approvals from a fourth trust in order to capture the data from multiple consultations. I therefore made the decision to exclude the third trust at the outset, with the option to return if recruitment numbers were low.

Concentrating recruitment in these larger centres was time efficient and enabled access to a diverse sampling pool. In addition, both trusts run their fetal medicine service across two centres as well as offering a number of satellite clinics in nearby locations. This meant that there was access to a wide range of populations.

In terms of the third factor, patient volume, the data collated over the past five years were examined and the number of anomalies recorded was used to estimate

likely identification rates in the various centres. In a year (averaged over the past five years), over 150 women would have been eligible for recruitment into the study from the two inter-referring tertiary referral centres, as their pregnancies had been affected by one of the severe congenital anomalies described in Table 1.1. Although sample size was not predetermined, it was felt that approaching these two trusts would provide a sufficiently large and diverse sample pool to achieve the aims of the study. The option to extend to the third trust was available, as previously stated, if recruitment proved difficult, but was not required. The centres selected for this study serve a large diverse population, as is illustrated in the sections that follow.

3.3.2 *DEFINING PARTICIPANTS*

The decision-making processes, following a diagnosis of a severe congenital anomaly, are not enacted in isolation, but as part of a manifold set of interactions embarked on by multiple players. As highlighted in the literature, the interactions between clinicians and parents can be influential in the decision-making process. Determining who should constitute a participant was complex, as the influence of ‘others’ would probably vary and could include extended family, friends, religious leaders or other advisors. I made a pragmatic decision, therefore, to include healthcare professionals and the parents as the participants, while remaining mindful of the potential influence of others. This is demonstrated in the presentation of the findings in Chapter 5. In order to explore the decision-making processes of the parents, the recruitment of women and their partners was crucial. Although it could perhaps be argued that data generated from clinicians were not central to the process, there were a number of reasons that I felt justified in including them. My primary intentions were fourfold: first, it served as a preparatory phase, where I could gain some insight into the practicalities of the process before recruiting parents; second, it enabled me to engage with the clinicians and develop some trust; third, from a practical perspective, it meant that the clinicians were actively aware of the research and provided an opportunity for them to engage in the consent process; fourth, parents did not make their decisions in isolation, but as part of an ongoing consultation process, therefore getting ‘both sides’ of the story was extremely useful.

3.3.3 *SAMPLING OF HEALTHCARE PROFESSIONALS*

The sample of healthcare professionals comprised consultants from fetal medicine, neonatology, surgery, cardiology, neurology and genetics as well as a small number of fetal medicine midwives. Due to the small number of clinicians working within these specialities, a detailed breakdown of the demographics of the sample will not be provided. A total of 18 healthcare professionals were interviewed, with men marginally outnumbering women by 10 to 8. This broadly reflects the demographics of the consultants of interest, although the midwifery workforce was all female. Healthcare professionals from all four centres were included. The ethnic origin and place of training of the sample was heterogeneous and included healthcare professionals from Asia, Africa, the UK and Europe, with the countries from which their specialisation was obtained also varying widely. The sample was strategically defined, with all the fetal medicine and specialised fetal consultants approached (eleven in total), alongside five of the consultants from the supporting services and two fetal medicine midwives. These individuals were identified by the fetal medicine consultants as primary care providers within the fetal medicine environment, as they attended multi-disciplinary meetings and joint counselling sessions. In addition, full-time fetal medicine midwives were invited to participate. This meant that part-time midwives were excluded. This was predominantly due to the difficulties arising from shift practices and requirements to cover other areas of the service. Of the 22 healthcare professionals approached, 21 agreed to be interviewed (one declined on the basis that they were to retire within a few weeks). Of those, one withdrew due to time constraints, and in two cases the interviews were interrupted due to clinical issues and were therefore not used. In total, 18 healthcare professionals were interviewed. The heterogeneity of the sample was important in providing contextualisation of the service provision, and is a reflection of the enormous diversity of the NHS workforce. Although the primary aim of these interviews was to construct an understanding of the contextual factors influencing the decision-making process, the interviews also provided an opportunity to meet the staff and reiterate the practicalities of this study. Interviewing midwives was more difficult in work time, due to working patterns. Whereas appointments for interviews were booked with clinicians,

midwives had less control over their working day and interviews were undertaken during lunch breaks. This resulted in a number of the interviews with midwives being abandoned when clinical issues arose and their lunch breaks were disturbed. Only completed interviews were included in the sample, as a number of midwives were concerned that statements they had made could be taken out of context. Generally this group was difficult to access due to practical issues. Although only two midwife interviews were included in the final analysis, these added to the diversity of the sample and provided a different perspective.

In addition, five clinicians consented to consultation recordings but were not interviewed. This group comprised the clinician approached who was not interviewed, a specialist surgeon, a senior consultant from the directorate who attended a multidisciplinary consultation, and two registrars (one was visiting from another centre and the second commenced their rotation in fetal medicine after clinician recruitment and clinician interviewing was complete).

Half of the clinicians invited to participate had been involved to varying degrees on advising in the project development. This was likely to have been reflected in the high recruitment rate.

3.3.4 *SAMPLING OF PARENTS*

The primary study sample consisted of prospective parents whose pregnancies were affected by a severe congenital anomaly. Antenatal and maternity services, where ultrasounds and routine checks are performed by midwives and sonographers, run alongside fetal medicine clinics in most NHS hospitals. When an anomaly is suspected, parents are transferred from the antenatal clinic to fetal medicine, where they should be seen within five days (Public Health England 2010). In order to gain an insight into parental decision-making, parents were recruited from a number of fetal medicine clinics.

Purposive or maximum variation sampling is used to produce relevant data for theory generation (Ritchie *et al.* 2014). Applied to this study, this meant that variables identified within the quantitative literature were included to ensure

diversity and thereby ensure, as far as possible, that a full range of views and experiences were accessed. As discussed in section 2.2, there is general consensus that chromosomal anomalies, with poor prognosis and high certainty, are more likely to be terminated than structural anomalies with less certain outcomes. In addition, low socioeconomic status, minority ethnic status and increased gestational age are indicators of the decision to continue with the affected pregnancy. Findings from these papers present a snapshot of the parents' decision, namely the outcome, rather than an insight into how the outcome is achieved. Nonetheless, application of the findings described above has provided support for the development of a stratified sampling frame that has included coverage of the categories highlighted below.

- *Selection by severity of anomaly (as defined in Table 1-2)*
- *Type of anomaly i.e. major structural or chromosomal*
- *Gestational age when anomaly first suspected*
- *Ethnicity*
- *Socioeconomic status / levels of deprivation*
- *Decision to terminate or to continue*

Despite evidence supporting the need for a stratified sample, two practical issues arose in relation to implementing this. The first related to how to ensure the sample reflected the views of women who terminated and those who continued, and the second pertained to the definition of 'severe', particularly with regards to cardiac anomalies.

The majority of pregnancies affected by a severe congenital anomaly are terminated, (Budd *et al.* 2015), and a significant risk was that the sample would be unrepresentative of those who continued their pregnancy. Although the decision to terminate or continue a pregnancy had not been made at the time of enrollment into the study, recruitment was fortunately unproblematic and the women enrolled provided me with a sample from both pathways. Indeed, this generally held true for the remaining variables, although where more than one suitable patient was identified on the same day but on different sites, I would have to

identify which parent would contribute most to the heterogeneity of the sample. This was only an issue on one occasion.

The difficulty in determining what constitutes a severe anomaly is a theme that runs throughout this thesis. For the purposes of this study, nine of the combined FASP anomalies were included:

- *Anencephaly*
- *Spina Bifida*
- *Severe Cardiac*
- *Bilateral renal agenesis*
- *Lethal skeletal dysplasia*
- *Congenital diaphragmatic hernia (CDH)*
- *Exomphalos*
- *Trisomy 18*
- *Trisomy 13*

These were described in Table 1-1. In addition, a search of the regional anomaly database was undertaken to identify any additional conditions that had been terminated under Section E, to ensure no anomalies were excluded. However, no further conditions were identified.

For this study, I excluded three of those screened for fetal anomalies: Down's Syndrome, cleft lip and palate, and gastroschisis. Neither cleft lip nor gastroschisis are life-limiting when not associated with a chromosomal disorder or syndrome. As such, they will have no bearing on the neonatal mortality rates if the pregnancy is continued. These anomalies therefore lie outside the scope of this study. I also specifically excluded Down's Syndrome. As discussed in section 2.1.2, there is a wealth of literature pertaining to Down's, much of which provides a conflicting pattern to that presented for other anomalies. This included variability in access, uptake and detection rates of Down's Syndrome with levels of deprivation, unlike other major anomalies (Alderdice *et al.* 2008). In addition, the termination rates are significantly higher than those for other severe FASP anomalies (86% versus 70%) (Budd *et al.* 2015). Despite the variability in disability in infants with Down's

Syndrome, the impact on mortality rates will be small. The rationale for the variations seen between Down's and the other FASP anomalies is unclear and not within the remit of this project.

In addition to these exclusions, a definition of 'severe' cardiac was sought. Defining this was both necessary and infinitely difficult. To date there is no clear professional consensus on what constitutes a severe cardiac anomaly, although registers collect details on 13 'severe' cardiac anomalies (Dolk *et al.* 2011). However, many of these are again not life-threatening, and therefore I needed to apply caution in universally including them all. After much consideration, I made the decision to review cardiac anomalies on a case-by-case basis, with regards to whether the anomaly met the criteria for a termination under Section E. In practice, the cardiac anomalies included within the study were either hypoplastic left hearts (where the baby is born with only a right-side functioning heart), or a combination of cardiac and other structural or chromosomal anomalies.

Alongside the criteria pertaining to the type of anomaly, a series of inclusion/exclusion criteria were made explicit. As summarised in Table 3-1 below, all women whose pregnancies were suspected of being affected by a severe congenital anomaly were eligible to be included. I was reliant on the clinicians to determine the point at which there was a 'reasonable' suspicion of an anomaly, as enrolling all 'high risk' women would not have been practical or ethical, as many would subsequently find that the pregnancy was normal. This approach risked missing a small number of patients, or recruiting them further down the care pathway if an anomaly was subsequently diagnosed. It also risked not recruiting women who refused further testing. In practice this appeared unproblematic. First, clinicians appeared to be good predictors of the clinical outcome when presented with a series of soft markers, and were thus able to determine which pregnancies would be severely affected, even if they could not confirm a diagnosis. All the women identified as 'high risk' subsequently went on to have a diagnosis of a severe anomaly. In addition, the structural anomalies were visible on ultrasound; therefore parents went from suspicion to diagnosis of the initial anomaly very quickly. Finally, a number of women who refused or delayed invasive testing were

recruited as clinicians had flagged the cases even though a definitive diagnosis was not made until just before the birth of the baby.

Table 3-1 Inclusion and exclusion criteria

Inclusion criteria	Exclusion criteria
All women whose pregnancies were suspected of being affected by a severe congenital anomaly screened for by the FASP screening programme...	- Women who did not wish to participate - Women for whom the option of a termination, for clinical reasons, was not offered - Women whom the clinicians did not feel were suitable for the study - Women who required a translator - Women who had a diagnosis of Trisomy 21, gastroschisis or cleft lip/palate

In a perfect world, women who do not speak English would have been included in the study. However, this study was a PhD project and had minimal funding attached. The additional costs necessary for translation could not be justified. Failure to include them is a limitation and will reduce the external validity and transferability of the findings. The group who were excluded would have included a number of ethnic minority women and future research is needed to focus specifically on their needs. Due to the current lack of evidence surrounding this issue, this project was largely a scoping and exploratory exercise. Redirecting resources into achieving this, whilst limiting the scope of the investigation, was felt to be unjustifiable.

3.3.4.1 *SAMPLE SIZE*

For qualitative studies, the sample is ideally judged complete when theoretical saturation has been reached. Theoretical saturation refers to the point at which new interviews generate no data that significantly modify existing analytic themes (Bryman 2001). In relation to the clinicians, the 'whole' population (within the centres selected) was interviewed. The group presented a relatively homogenous perspective of the topic investigated; therefore similar themes arose within all the interviews and the analytical themes were not significantly modified after the first ten interviews. Had recruitment extended to other centres nationally it is unclear whether this would still have been the case. Although perhaps not necessary in terms of data generation, interviews with all the clinicians were completed as part of my preparatory work. In relation to parental recruitment, an end point was required in terms of sample size. Specifying upfront the number of participants required within a qualitative study such as this is challenging, as it is unclear how large a sample will be required for data to be saturated (Bruce 2007, O'Reilly and Parker 2012). In addition, the uniqueness of each person or group, it can be argued that no data can be truly saturated (Wray et al 2007) and total saturation is probably never achieved (Corbin and Strauss 2008). These suggestions are largely at odds with the use of data saturation as a quality marker within qualitative research, and the difficulties in transparently accounting for sample size (Guest *et al.* 2006). At the outset of the study, I had anticipated that I would require between 10 and 40 couples to produce some meaningful findings. This range was partly derived from others' experiences of undertaking a PhD, my own experience, and pragmatically, the limitations imposed by time and finance. The need to quantify sample size arose from governance requirements to provide an estimate prior to approvals being released. This again is at odds with the concept of data saturation, where sample size is determined once analysis is underway and perhaps highlights a further contradiction between the process requirements and methods of qualitative research.

3.3.5 *RECRUITMENT STRATEGY*

Much time was employed planning the recruitment strategy. However, in practice the process became much more streamlined and simple. This was likely to have been as a result of developing good relationships with clinicians in the field.

When an anomaly was suspected, parents were invited to wait and be reviewed by a fetal medicine clinician. Generally parents were seen within a few hours, and where this was not possible they were invited to return the following day. Whilst arrangements were being made, parents were shown to a consultation room on the fetal medicine unit. At some point during this time a clinician or midwife approached the parents and asked if they would consider taking part in some research. All agreed to speak to me and, armed with cups of tea, I briefly explained my wish to record the consultation. Consent for this was taken at the time. However, a full explanation of the aims of the study was withheld until the parents were much further on in their journey. Approaching parents at such a vulnerable time was hard to start with, for me and the clinicians. Invading an immensely distressing and personal time felt uncomfortable. Nonetheless, I had been assured by the PPI group that the distraction (and tea) would be welcome and comforting during this time. This was reflected in the positive responses I received from the parents recruited. Clinicians had initially expressed reservations about approaching parents during this period. Considerations were given to alternative recruitment strategies that involved identifying the parents further down the pathway once a relationship had developed with the clinicians, but this meant there was the potential for a number of consultations to be 'missed'. In practice, once the first couple of patients had been recruited, the clinicians appeared to feel more confident in the approach, and subsequently 'full' data were obtained from the majority of the parents.

The need for a step-wise approach to consent had been identified during planning discussions, with many of the parents in the PPI group expressing concern that despite the welcome diversion, parents were unlikely to absorb the details of the study at this first contact. Examples of a step-wise approach to consent were

identified in the literature pertaining to 'at risk' groups such as the frail, elderly and mentally infirm (Rikkert *et al.* 1997, Bhutta 2004, Dunn, Jeste 2001). The sensitivity of the topic and the timing of the approach meant parallels between the situations identified and this study were pertinent.

The intended recruitment strategy for parents was planned to run in parallel between the four centres. Recruitment packs were put together and clinicians were asked to distribute them to suitable parents. It quickly became apparent that this process was unlikely to generate many recruits as, without my presence as a reminder, the project was quickly forgotten. The approach was adapted and I began to attend the fetal medicine clinic when scanning lists were being run and I decided to focus on one site rather than splitting time across multiple sites. This proved beneficial in two ways: first, recruitment picked up quickly; and second, observations on the working practices and problems encountered by staff alerted me to issues within the day-to-day workings of the centre. This provided additional background information that was used to support or question findings from data generated in interviews and consultations. The limitation of this approach was that researcher presence was only possible in one trust at a time. This meant that the majority of patients were recruited from the same trust. However, consultation recordings were more equally divided across the two trusts, as parents were seen in specialist clinics (neurology and cardiology).

3.3.6 *SAMPLE DESCRIPTION*

A total of 20 women and 18 men were recruited. Nineteen of the women had a partner at the time of diagnosis, although one couple separated soon afterwards. Of the 18 couples, 15 were interviewed together, and three women were interviewed alone, either through choice or due to practical constraints. Although the husbands of these three women were not interviewed, they participated in consultations from which data were generated. The number of consultations recorded for each mother or couple ranged between one and seven, with the most frequent being three consultations. This was reflected in the number of hours of recordings that ranged between 1.5 and 15 hours per mother or couple. Consultation recordings and parental interviews represented a large data set

consisting around 70 hours of recordings, with additional supporting data generated through the recording of interviews with clinicians thus taking the hours of recordings to well over 80. When seeking to demonstrate data saturation, the time expended on each participant, for example the length of interviews as opposed to simply the size of a sample has been argued to be a more valuable reflection of the quality of the research (O'Reilly, Parker 2012). In this instance, the number of participants recruited was perhaps less representative of the volume of data generated than the hours of recordings collated. Heeding the warnings of Corbin discussed above, I will not claim data saturation, rather state that following analysis of the data available, data from the final three participants provided no new themes, with the data derived from the interviews and consultations supporting the categories already established. This is supported by Francis *et al's* proposal for a '10+3' formula to establish data saturation (Francis *et al.* 2010). This formula requires a minimum of ten interviews to be conducted followed by a further three to evaluate if any new insights are produced. In this study, no new themes or categories emerged following analysis of the data generated from the 17th woman recruited.

3.3.6.1 *SAMPLE CHARACTERISTICS*

Striking a balance between protecting the anonymity of the parents and illustrating the diversity of the sample is hard to achieve. I have selected a number of variables that are presented in Table 3-2 and Table 3-3. First, the types of anomaly are illustrated, and subsequently the level of deprivation is recorded along with the decision to continue or terminate the pregnancy. The sum of the anomalies presented below is greater than the 20 pregnancies included as some pregnancies were affected by more than one anomaly. This demonstrates a distribution of anomalies, including structural and chromosomal, as well as those affecting different systems.

Table 3-2 Distribution of anomalies

Type of Anomaly	Distribution of Anomalies
Severe cardiac	5
Congenital Diaphragmatic Hernia	3
Trisomy 13, 18 and Triploidy	6
Spina bifida	5
Anencephaly	4
Exomphalos	1

In order to determine socioeconomic status of the parents, deprivation, based on postcode of residence was calculated for an initial check and subsequently validated using individual level information obtained from patient notes and the parents themselves.

The level of deprivation was calculated as described in full in Appendix E. Simply, it involved attributing a postcode to a super output area (SOA), then converting the SOA to a deprivation decile figure. The SOA are geographical areas and represent the smallest areas for which deprivation data are available. In this instance the Multiple Index of Deprivation has been used, as it incorporates seven measures of deprivation, including health, education, income, employment, housing, crime and living environment. The deprivation spread for this study is presented in Table 3-3 overleaf. This demonstrates a distribution of women, who terminated and continued, across all levels of deprivation.

Table 3-3 Pregnancy by quintiles of deprivation (1 most deprived)

Decile of Deprivation	Continued	Terminated
1 and 2	4	1
3 and 4	4	2
5 and 6	0	2
7 and 8	1	2
9 and 10	1	3

I subsequently ‘converted’ the deprivation levels into three groups by combining levels of deprivation and cross checking with the additional information collated once parents had been recruited. These groups represented low, medium and high socioeconomic status. Not only does grouping in this way make it more difficult to identify individuals but, I hope, improves the readability of this thesis. The information collated in order to facilitate the grouping included: educational attainment of both parents, occupation, job title and industry, and ethnicity. Additional information relating to housing (whether owner occupier or tenant) and income was generally gauged during discussions, but not requested explicitly due to the sensitivity of the information. These categories broadly reflect aspects of the information collated for the Multiple Index of Deprivation, but represent individual rather than area data. Classification of socioeconomic status is complex and due to its multifaceted nature, accurate measurement is problematic (Deonandan *et al.* 2000). In the seminal review of measurement of social class, Liberatos *et al* concluded that there is no best measure, however, recommended that researchers incorporate at least two indicators of social class in their measurement, with use of occupation is one of the most widely applied indicators (Liberatos *et al.* 1988). A UK based ranking of occupation has been applied to this study. This was derived from the British Registrar General's levels of social class (Benzeval *et al.* 1995). Application of occupation as the index of socioeconomic status is widely relied on in the UK, whilst in the US measures based upon education are more commonly used (Davey Smith *et al.* 1998). As ranking is

available for both measures, the combination of these two was perceived as a valid tool for categorising. With some evidence to suggest that use of area data as a proxy measure of socioeconomic status may be a better discriminator in the study of pregnancy outcomes than classification by occupational social class (Spencer *et al.* 1999) and indications that household socioeconomic status measured by the postal code methods, is more closely approximated by men's occupations than by women's (Deonandan *et al.* 2000), incorporation of population and couple based data should theoretically have provided an accurate measure of socioeconomic status.

Apart from one couple, deprivation levels and socioeconomic status correlated. I have included the couple in the corresponding socioeconomic group rather than deprivation level. The couple concerned lived in a new-build council-owned home on the edge of a well-established estate. It is likely that the proximity to the estate and recent completion of the property resulted in its inclusion in a potentially unrepresentative decile of deprivation. Although calculating deprivation levels in the method demonstrated appears to be largely representative, use of the method when applied on a small scale may highlight inconsistencies between calculated deprivation levels and socioeconomic status.

Table 3-4 Pregnancy by socioeconomic status

Socioeconomic status	Continued	Terminated
Low	9	3
Medium	0	2
High	1	5

Other variables taken into consideration included ethnicity and gestation at diagnosis. Five women from minority ethnic groups were recruited into the study, providing some additional insights into the decision-making process. In terms of gestation, this varied between 9 and 21 weeks for suspicion and diagnosis, thus ensuring a heterogeneous sample on this count. Although none of the parents

approached by the clinicians declined to participate, it is unclear whether others were not invited. The resulting sample correlated well with the sampling frame; however this happened, to a large extent, fortuitously.

In the literature, most women terminate an affected pregnancy, therefore I was a little surprised to have a sample where an equal number of women continued and terminated their pregnancies. Due to the numbers recruited, this could have been chance, however, some women who terminated may not have been approached to participate as they made a quick decision and I missed them. In addition, anecdotally I have been told that termination rates are higher in local DGH's compared to tertiary referral centres. This may represent the influence of the counsellor (Kim *et al.* 2002, Holmes-Siedle *et al.* 1987). Nonetheless, extending the study to different sites may have produced different results. Comparison of the sample to the anomaly register data in the future may provide further insight into this.

3.3.7 *METHODS CONSIDERED*

The complexity of determining an appropriate data collection method is heightened in the context of sensitive research such as this, due to the extensive ethical and moral issues that arise (Dickson-Swift *et al.* 2007). Therefore, a step-wise, methodical approach to determining the most appropriate methods for the study was employed. This ensured that additional measures were in place to check and recheck the suitability of the methods, not only in the ability to address the question posed, but also in relation to the impact of the methods on the participants. First, the literature identified and explored in Chapter 2 was examined and critiqued in relation to choice of method. This was extended to additional literature, such as that pertaining to bereavement and parental decision-making in the neonatal period, particularly in relation to premature births. The benefits and limitations of the different methods employed were considered. Second, members of the PPI group reviewed proposed methods for acceptability and feasibility. Finally, the supported methods were re-explored in terms of credibility and dependability.

3.3.7.1 VIGNETTES

The first method considered was vignettes. There are significant ethical issues surrounding recruitment of couples at a particularly sensitive and emotionally charged time. One approach used by a number of authors, in order to avoid these issues, has been the use of vignettes (Lawson 2006, Learman *et al.* 2005). In these kinds of studies, a population with a number of shared characteristics is recruited and asked to respond to a number of hypothetical scenarios. The benefits of this approach in capturing the voices of informants, particularly in the discussion of sensitive topics, whilst enabling them to retain a high level of control is well established (Schoenberg, Drew 2002, Barter, Renold 2000). However, there is some evidence to suggest that responses to anecdotal or hypothetical scenarios, in relation to terminating or continuing a pregnancy, do not necessarily correspond with what the same woman would do if faced with a real decision, (Erikson 2003, Sawyer *et al.* 2006), as the cues and factors that guide decisions in real-life may be missed (Palomaki *et al.* 1996). The clinician and parent arms of the PPI group raised further concerns over the findings that would be generated by vignettes. Clinicians cited scenarios where parents moved to another option once more information was obtained or a diagnosis made, while parents provided examples of how their own beliefs were questioned as the theoretical possibility became a personal reality. In addition, the use of vignettes restricts the exploration of the phenomenon of interest within the naturalistic setting, thus underestimating the importance of context, something highlighted within the literature as well as from a theoretical perspective as discussed in section 2.5.

3.3.7.2 FOCUS GROUPS

The data collected in focus group sessions typically consist of tape-recorded group discussions among four to ten participants who share their thoughts and experiences on a set of topics selected by the researcher (Morgan, Spanish 1984). Participants respond to, and build on, the views expressed by others in the group. It becomes a synergistic approach that produces a range of opinions, ideas and experiences that generate insightful information (Litosseliti 2003). The strength of this method, therefore, lies in the interaction of multiple perspectives and the

discussion arising therefrom (Ritchie *et al.* 2014). In addition, they present a more natural environment than interviews as the participants both influence and are influenced by others, as would occur in real-life (Kreuger 1994). Focus groups perhaps occupy a middle ground between observation and in-depth interviewing (Morgan 1997). However, they are not without problems. In particular the appropriateness of the use of focus groups to explore sensitive topics is commonly questioned (Smithson 2000). Within this study, the potential for stigma to be attached to termination, and the need to disclose in order to participate, was of particular pertinence.

The PPI group strongly expressed reservations about the use of focus groups in eliciting data from parents, where the personal nature of each decision was perceived as prohibitive of group discussion. Of particular concern was that the parents who terminated the affected pregnancy could have been required to disclose this to a group of parents who potentially disapproved of their decision. These concerns were reflected in my management of the PPI group, where the parents who terminated and those who continued, never met. One solution may have been to separate the groups, as I had done for the PPI parents. However, the low incidence of anomalies meant that I may have had to wait for long periods before having sufficient numbers of parents to generate a discussion. In particular, recruiting a group of parents who were continuing their pregnancy would have been difficult due to the time limits imposed by the pregnancy itself. Changing the timing of the focus group from antenatal to postnatal was not considered feasible as the parents could then be caring for a terminal child. I felt this would likely impact significantly on attrition rates.

The use of focus groups to examine decision-making processes from a naturalistic perspective is relatively uncommon (Klein 1997), something that appears to have changed little in subsequent years. The reason for this is unclear, although could be attributed to the difficulties perceived in capturing the essence of a decision-making process in a context complicated by immense time stresses and shifting or ill-defined goals (Orasanu, Connolly 1993). In the context of decision-making following diagnosis or suspicion of a congenital anomaly, these difficulties can be

observed in the limitations created by pregnancy gestation, and the uncertainty created by shifting and developing understanding of the prognosis and diagnosis. Thus essentially the focus group will provide a snap shot of opinion at a single point in time.

The literature exploring the clinician's perspective within the context of suspicion or diagnosis of a severe anomaly has been predominantly undertaken using focus groups, with subsequent interviews to enhance the data generated (Williams *et al.* 2002, Farsides *et al.* 2004, Alderson *et al.* 2004). This has proved particularly effective in enabling healthcare professionals, of different levels, to engage in discussions surrounding antenatal screening and counselling, and subsequently question their own practice and assumptions in a protected environment. An unexpected therapeutic effect of these focus groups was that clinicians discovered that others shared their dilemmas (Williams *et al.* 2002). Replicating this may have provided the opportunity to explore, in greater depth, clinicians' views on aspects such as interpretation of the termination law. However, recruitment of clinicians from four clinical sites, for a focus group, raised the major practical issue of getting the clinicians together.

3.3.7.3 DIARIES

It was anticipated that the decision-making process would take place in a variety of settings, including at home with friends and family, as well as during planned and unplanned discussions with professionals, and through support groups. From a research point of view, observation or recording of all these interactions was impractical. The extended time and confidentiality issues likely to be encountered were too great for a single researcher to address.

Consideration was therefore given to asking the women participating to write short diary entries or vocalise and record their feelings over a period of time. From a theoretical perspective, use of diaries is perhaps an effective way of capturing decision-making from a naturalistic perspective. Not only are the data recorded in 'real-time' but it engages with the context in which the decision is being made whilst capturing the essence of the process rather than the outcome *per se*.

This method is well accepted in the literature, with Bytheway et al. concluding that the use of a diary can “*help to distinguish what people **actually** do from what they **say** they do*” (Bytheway, Johnson 2002, pg.171). However, critics of this method argue that participants’ literary abilities and propensity for writing may be the primary determinant of the quality of data generated (Elliott 1997). This is likely to impact more significantly on women with a lower educational ability. Members of the parent advisory group also raised concerns over the time input required with diary entries.

3.3.7.4 INTERVIEWS

The strength of interviews, like focus groups, lies in their ability to capture the social worlds of the interviewees through the creation of narratives (Miller, Glassner 2011). Whilst research focussing on the meanings attached to individuals’ experiences, or the way that these experiences are communicated to others, is ideally studied through the use of narrative, (Elliott 2006) it does not transparently reflect experience: rather, it gives meaning to it (Ferber 2000), and provides a contextual understanding of how people make sense of their experiences, as well as an understanding of the social forces that shape them (Elliott 2006). Hurwitz describes the use of narrative as a tool for mediating between “*subjective and objective points of view and between the personal, institutional, and social dimension of health and illness*” (Hurwitz 2004, pg.2). They can thus reflect the impact of the event on the individual within their own context (Williams 2004), as well as the way in which people inform and shape their behaviour. At this point a resonance with the previous debate on ontological perspectives can be felt. The ‘subtle realist’ perspective is clearly reflected in the need to explore an individual’s experience (through narrative), but the inability of narrative to capture a shared, external reality also requires consideration.

Traditionally, medicine has focused predominantly on the quantitative empirical fact or nomothetic domain, and sought to prioritise ‘objectivity’. Knowledge labelled as subjective is frequently discredited (Elliot 2005). This, however, results in a singular perspective of knowledge, and the diversity of viewpoints is lost (Hurwitz 2004). Narratives create an essentially subjective account of an

experience or encounter as lived by an individual in their social context. The inclusion of narrative data in addressing the question posed for this study is essential. Nonetheless, reliance entirely on narrative could have implications for the data to answer the research question posed, as well as transferability and dependability of the findings. The importance of capturing a multidimensional perspective is not to be underestimated when attempting to elicit the 'truth', and is widely deemed to be good research practice (Ritchie *et al.* 2014). The dichotomy discussed above highlights the benefits of narratives, whilst recognising them for what they are, a distilled illumination of the discourse influenced by events and factors occurring down the line. Consequently, analysis of interviews should be viewed in this light and not interpreted as empirical fact, but as further illumination, probably embellished and reframed over time.

There is a growing challenge to the over-reliance of interviews as a method, where it is seen as a reflection of social and cultural trends, such as celebrity media interviews (Gubrium, Holstein 2011). Many of these concerns arise from the failure to appreciate the different epistemological standpoints from which the data are analysed (Yeo *et al.* 2013), something that has been explicitly discussed within this context.

From a theoretical perspective, the use of interviews to examine decision-making from a naturalistic paradigm is broadly reflective of the issues raised through the proposed use of focus groups, where sole application of this method of data collection may limit the capture of 'real-time' decision-making processes, instead providing a reconstructed perspective on the events.

Use of secondary analysis of existing interviews was also given consideration. As discussed in section 2.3, HERG have collated a large number of interviews with parents. Application of a research question to existing data is appealing in terms of saving time and effort of both researcher and participant (Ghauri 2005). Ethically, re-using data (with appropriate consent in place) represents good value for the participants (Grinyer 2009). However, the appropriateness of the data available also required consideration (Denscombe 2011). I therefore had to weigh up the

benefits of secondary analysis with the opportunity to create a dataset with the primary aim of addressing the research question posed. The consideration given to collating data from consultation recordings was a primary factor in not pursuing the potential to apply secondary analysis to existing data.

3.3.7.5 RECORDING CONSULTATIONS

A substantial body of evidence demonstrates the usefulness of recording consultations, especially as a means of generating data to allow for an analysis of communication processes in a range of clinical settings (Pryde *et al.* 1993, Boyd *et al.* 2011, Raupach, Zimmermann 2004, Bryman 2001). In addition, the work by Pilnick *et al.* identified in the literature review, highlighted the need to capture data created through the interaction between clinician and parent (Pilnick, Zayts 2014, Pilnick 2008, Pilnick, Zayts 2012). In addition, data generated through this method would assist in capturing the essence of the healthcare context within which the decision-making process was being enacted.

In relation to the acceptability of recording consultations, there is literature that suggests that participants become rapidly accustomed to the recording devices, and they have little influence on the behaviour of practitioners or patients (Bucher *et al.* 1956, Adams, Cox 2008). There was broad acceptance of the use of consultation recordings as data sources by parents and clinicians in the study advisory groups. Whilst the clinicians, in particular, expressed concerns over video recording, stating that they felt it was too invasive, there was a unanimous acceptance of voice recording for research purposes.

From a theoretical perspective, consultation recordings provide a distinct opportunity to capture the data in 'real-time'. From a naturalist's perspective this is ideal as it will reflect not only the uncertain, dynamic environment or context in which the decision is being made, but also seek to encapsulate the process rather than the decision itself.

3.3.7.6 ETHNOGRAPHY

The use of recordings of consultations ties in closely with ethnography, in that they both generate data from naturally-occurring encounters. Ethnography involves understanding the social worlds or cultures of particular groups. In particular it provides insight into shared beliefs, behaviours and values (Ritchie *et al.* 2014). In this instance, observation of parents and clinicians would allow the researcher to seize “*the unscripted, unrepeatable, and often unutterable stuff of existence beyond the grasp of interview-based inquests*” (Desmond 2007, pg.288). However, it has also been argued that recordings can equally provide these insights, whilst providing a more ‘accurate’ record, which is both more detailed and complete than that obtained through human observation (Grimshaw 1982, Hanson 1994, Gottdiener 1979), where recorded data may be re-played, enabling analysis to be delayed until the researcher has left the field (Gottdiener 1979, Albrecht 1985) and enables other researchers to repeat analysis (Grimshaw 1982, Hanson 1994, Gottdiener 1979). One particular reservation about using ethnography was that it was in a clinical situation with which I was broadly familiar. Setting aside taken-for-granted assumptions, in order to achieve ‘analytic distance’ from what I was observing was a significant potential limitation to any findings generated, and required careful consideration. Practical issues such as time constraints, where I would be required to be physically present, thus limiting the number of units from which I could recruit, was also taken into consideration. I also felt uncomfortable with the thought of observing consultations of parents where subsequent investigations confirmed that their baby did not have an anomaly. This discomfort arose from two considerations; first, the PPI group had highlighted the added anxiety that a crowded room created; and second, collection of data which were not applicable to the study felt ethically questionable.

As with consultation recordings, ethnography is an ideal tool for capturing the decision-making process from a naturalistic stance. The process (rather than outcome) is the primary objective of the data collection, which is gathered within the context in which the decision-making process is enacted.

3.3.8 METHODS USED

What is generally evident in the research literature is that debates over methodology in research rest on the suitability of the method for generating evidence in response to questions, rather than on the 'validity' of the method itself. In addition, exploring the research question from more than one perspective, often realised by means of applying different methodological approaches, can provide greater insight into the phenomenon of interest (Flick 2004). However, this comes with the warning that; *"putting the picture together is more problematic than such proponents of triangulation would imply. What goes on in one setting is not a simple corrective to what happens elsewhere: each must be understood in its own terms"* (Silverman 1985, pg.21).

The use of diaries, focus groups or vignettes to generate data in this context were fairly quickly discounted. In seeking to elicit the views of a wide range of women, diaries risked excluding some groups by virtue of their literacy ability. Use of voice recorders instead of written diaries was considered, but was precluded by cost. Focus groups of parents were vetoed by both clinicians and parents in the PPI group due to the perceived risks to participants. Despite the methodological benefits and proven track record of focus groups for the clinicians, I felt that the practical difficulties and subsequent limitations, had there been limited attendance, outweighed the potential benefits. Vignettes, although given consideration by the PPI groups, were excluded by me on the basis of the inherent methodological problems associated with their generation of purely hypothetical data. Generation of data through observation (either ethnography or recordings) and interviews were the most appropriate options, and reflected the requirements of the naturalistic theoretical stance taken.

The limitations of narratives as a method of generating data have been discussed. However, triangulating narratives with an observational method has been suggested to be an effective way of developing an informed understanding of patients' experience and, in this instance, accounts of the processes involved in their decision-making (Silverman 1998). I therefore had to select the

observational method which I felt was most appropriate to combine with the interviews. Little evidence from ethnographic studies was identified within the literature reviewed. This added to the difficulties to make a case for its application for this study. In addition, ethnography was poorly understood by both parents and clinicians, with responses to initial suggestions of adoption of this method highlighting surprise; *"You mean you just stand around and watch?"* (Parent PPI). Although these concerns could have been overcome through careful explanation, the intrusiveness of the researcher in a deeply personal and emotional moment was perceived by many of the PPI parents to be prohibitive. Many spoke of large numbers of clinical staff present at appointments, and the added fear that it engendered; *"If there are so many doctors, it must be really bad"* (Parent PPI). There was overall agreement amongst the parents that additional personnel within the consultations, whether research or clinical, should be avoided.

Broaching the issue of video or voice recorders provided mixed responses from the PPI group. Although all parents and clinicians initially responded negatively to the concept of video recordings, most of the parents reconsidered the idea and felt that it was viable. The clinicians, however, retained their view that it was too invasive. As these were the gatekeepers to recruitment, I made the decision to continue with voice recordings only, which had been unanimously accepted. The use of video recorders over voice recorders may have provided the opportunity to analyse non-verbal communications in a way that the voice only recordings could not. However, the potential added data had to be balanced with the ability to recruit.

I therefore utilised two main data collection methods: semi-structured interviews with parents and clinicians, and consultation recordings. Alongside I have made generalised field notes from meetings attended and time spent in the clinical setting. Data collection was divided into three phases. The first involved recruitment and interviewing of clinicians; second, recruitment of parents and the recording of consultations; and finally interviewing of parents. Despite the employment of similar methods for both groups of participants, each had their own unique difficulties. Overall, I felt that this decision was most likely to achieve

an effective balance between acceptability, practicability and appropriateness of the methods in addressing the proposed research question.

3.3.8.1 CLINICIAN INTERVIEWS - DESCRIPTION

The aim of the interviews was to elicit clinicians' views about their practice and provide contextual information. All the interviews were held in the clinicians' offices. An interview schedule (Appendix C) was developed but used sparingly, with the clinicians authoritative in their control of the interview. Interviews with clinicians lasted an average of 40 minutes, ranging from 30 minutes to one hour 10 minutes. They tended to be marginally structured, but conversational in tone.

3.3.8.2 CONSULTATION RECORDINGS - DESCRIPTION

There was significant variability in the number of consultations recorded for each parent. This ranged between 1 and 9, with of the most frequent being 3. Parents who continued tended to have more consultations, although some of these were short scan appointments rather than extended counselling sessions. The variation in length of the consultations reflected this, ranging from 20 minutes to 2.5 hours. Although predominantly fetal medicine counselling appointments, the consultations included a range of appointments, such as genetics counselling, meetings with neonatologists to devise a post birth plan, specialist reviews and multi-disciplinary team meetings (MDTs⁷), and scans.

3.3.8.3 PARENT INTERVIEWS - DESCRIPTION

The sensitivity of the subject area and the vulnerability of the parents at the time of the approach needed careful consideration. Although the subject and the methodology are justifiable, the potential for harm is apparent for the researcher and participants. The major risks associated with interviewing parents arose from the sensitivity of the subject matter and the potential for distress caused to

⁷ MDT - Weekly meeting involving clinicians, midwives and nurses from specialities involved in the care of the mother and baby. This could include neonatologists, geneticists, surgeons and fetal medicine specialists.

participants and researcher. Parents were provided with a list of organisations approved by the clinicians, to whom they could speak for support. Similarly, I had prearranged access to counselling services for myself, as well as a number of alternative support mechanisms. All the decisions made with regards to the practical aspects of the parental interviews were reviewed by the PPI group in terms of the potential harm to participants, and ways of mitigating risk to them.

Whilst clinician interviews and consultation recordings were well structured in terms of what I needed to take into consideration, the parental interviews were more complex and required extensive planning. A number of issues were considered prior to undertaking the interviews with the parents. These were: timing of the interview; location, including whether this should be done face-to-face or by telephone; whether or not to include partners; and, importantly, what to ask them.

The interviews took place around the time of the 6-8 week post-termination appointment (for those who terminated), or around four weeks pre-delivery (for those who continued) as these time points were identified as practical by clinicians and supported by the parent advisory group. Clinicians noted that women returning to the clinic for their post-termination counselling at 6-8 weeks typically wanted and were ready to talk. This appeared to be a sensible approach that perhaps met some needs of the parents as well. For those that continued, concerns were expressed by the PPI group that parents may find it difficult to find time for an interview if they were caring for a newborn with a terminal condition. From a practical perspective this appeared sensible, although the decision had to be weighed up with the methodological disadvantages of interviewing women at different times in their journeys, thus making comparisons difficult in terms of care during and after delivery.

A further decision related to whether or not the interviews would be undertaken face-to-face or by telephone. Although telephone interviews might allow respondents to feel detached from the interviewer, and thus relaxed and able to disclose sensitive information, the lack of visual cues might result in the loss of

contextual and non-verbal information that might in turn compromise rapport, probing, and interpretation of responses (Aquilino 1993). I therefore made the decision to undertake all interviews face-to-face. The option of telephone interviews would have become more technically challenging when interviewing the couple together but, more importantly, the emotiveness of the subject felt too difficult to approach over the phone. In practice, it became evident that the decision to undertake face-to-face interviews was the right one.

Women who had terminated the pregnancy were not keen to return to the hospital, whilst those who were continuing were often deferential about the location. I always offered the woman a choice as to whether she would prefer to be interviewed at home or at the hospital. Ensuring a safe and comfortable environment to facilitate the sharing of personal experiences is an essential part of the interview process as it can influence the dynamics of the interviewer-interviewee relationship (DiCicco-Bloom, Crabtree 2006). The interviews were generally undertaken in the woman's home with three exceptions, all of whom continued with their pregnancy: one participant had been hospitalised and the interview was undertaken in a private room on the ward; one couple chose to be interviewed at the hospital, as they had other children at home and felt the hospital would be more private; a third interview was undertaken in a consultation room at the father's request.

All the interviews were led by myself. The influence on the narratives constructed, when using a single (as opposed to multiple) interviewer has been discussed (Matteson, Lincoln 2009). The narratives constructed are not the work of the interviewee alone, and therefore alternative perspectives should be expected. However, in this instance, interviewing by a single interviewer was perceived as optimal, particularly in light of the need for close collaboration and trust between the researcher and clinicians. Financial constraints also made this a more viable option.

An interview prompt guide, based on a review of the relevant literature and discussions with clinicians, charities and parents, was used to help structure the

interviews. The interview guide has been based on that used by HERG in Oxford and starts with offering the participant an opportunity to tell their story. More specific prompts were then used to guide the interview, with the emphasis on allowing participants to construct their own accounts of the process and talk about the issues of greatest importance to them. The interview schedule (Appendix C) was revised and refined throughout the project in response to emerging themes and specific issues raised. In addition, listening to the consultations for each patient ahead of the interview allowed a certain degree of personalisation. The contextual details of the interviews were documented in a reflective diary to aid with analysis.

The decision on whether to include or exclude partner presence and participation required considerable thought. Although the decision about whether to continue or terminate a pregnancy legally remained with the woman, where a partner was present, it was likely to have been a joint decision. Therefore, gaining some insight into the perspectives of both parents was felt to be beneficial. My main decision involved whether to interview parents separately (thus including a set of interviews with partners) or together. Recent literature has suggested that joint interviewing, although well established in practice, is under-explored from a methodological perspective (Morgan *et al.* 2013, Morris 2001). This growing literature has begun to address the practical, ethical and methodological implications of interviewing two people simultaneously, and suggests that there are some advantages over individual interviews (Brannen 1988, Gerhardt 1991, Sakellariou *et al.* 2013). This includes the illumination of resources that are used within the decision-making process through the observed interaction of the couple (Sakellariou *et al.* 2013). In many ways this was reflective of the benefits of a focus group. The advantage of this in addressing the research question posed is clear. In addition it has been observed that very similar accounts were frequently generated by the two parties when interviewed separately (Morris 2001), thus suggesting that the narrative relayed by individuals in an interview is constructed within the relationship and not independently. From a practical perspective, interviewing couples separately would have involved identifying times when the other partner was out. This felt uncomfortable, particularly in relation to the

emotiveness of the topic, where partners are often perceived as the main source of support (Lafarge *et al.* 2013). The decision was made to offer the woman a choice. In total, five women were interviewed alone and 15 couples were interviewed jointly, with many reflecting on the cathartic nature of talking through the process together, often for the first time. Further details pertaining to the sampling of the couples can be found in section 3.3.

Although one interview only lasted 15 minutes, the others ranged between 45 minutes and 1.5 hours. The circumstances surrounding the short interview will not be discussed within this thesis, due to risk of identifying the parents.

3.3.8.4 PATIENT PROFORMA - DESCRIPTION

One additional method of generating data was added towards the end of the planning phase. Data relating to the anomaly (including gestational age and diagnosis), test results and demographic details were recorded from the patient notes. The triangulation of the consultation and interview data together with the information collated from the patient's notes provided a powerful method for understanding communication processes and the impact of these on patient decision-making.

In addition, information extracted about the parents' demographics enabled the comparison between area level and individual level socioeconomic status or deprivation to be undertaken (see Appendix D and E). Where the demographic information collated from the notes was incomplete, 'missing' data was subsequently requested from the parents during the interview.

3.4 MANAGING THE DATA

3.4.1 *PRIVACY AND CONFIDENTIALITY*

In terms of ethical procedures to guarantee privacy and confidentiality, every effort was made to ensure identifiable information was not included in this thesis or any other research output. Parents and relatives were given a random identifier, including a number which was applied to both partners in a couple. Prior to the number, the prefix mother or father was applied. After the number, the suffix 'terminated' or 'continued' was documented. Clinicians are referred to by a number throughout. These numbers were randomly applied after I had interviewed all the clinicians. The centre they work in is not related in any way to the numbers, and their gender is recorded as neutral.

All interviews and consultations were recorded on small encrypted and password-protected voice recorders. The recordings were downloaded onto my University computer as soon as possible after completion. The recordings were transcribed verbatim, and subsequently erased. No hard copies of the transcripts were retained. Consent forms were kept in a locked cupboard until they were scanned onto the University system and saved in a password-protected file.

3.4.2 *TRANSCRIBING*

Interview and consultation recordings were managed in similar ways. On completion of the recording, the data were downloaded onto a University computer (because this provided additional security). I transcribed verbatim all but three recordings myself, with the others sent out to a professional transcriber because of time constraints. Transcribing the data, although complex, time consuming (Britten 1995) and fraught with technical dilemmas (Fasick 2001, Wellard, McKenna 2001), has been cited as being central to the reliability and validity of qualitative data collection (MacLean *et al.* 2004, Seale, Silverman 1997). In addition, transcribing the data myself was beneficial as it enabled the correlation of verbal with non-verbal exchanges that had been noted during the interviews. As I had not been present during the consultations, the same argument

is not pertinent. Nonetheless, transcribing enabled a first-hand knowledge and understanding of the data to be gained. Ideas and thoughts that arose whilst transcribing were jotted down in the margins. These were returned to when the formal coding process commenced. As a result, the transcribing process became an integral part of the analysis process (Halcomb, Davidson 2006).

All recordings and transcripts were anonymised and labelled with a centre and study specific number. A further number was added to signify the order of the consultations. As per disposal policy, the unanonymised transcripts were destroyed once an anonymised version was created. A special encrypted and encoded file was set up on a shared drive, where the anonymised transcripts were made accessible to my supervisors.

3.5 DATA ANALYSIS

Qualitative data analysis can be described as: *“working with data, organising it, breaking it into manageable units, synthesizing it, searching for patterns, discovering what is important and what is to be learned, and deciding what you will tell others”* (Bogden, Biklen 1982, pg.145). The breadth of the definition highlights the numerous approaches to analysis of qualitative data.

3.5.1 METHODS CONSIDERED

There are a number of methods of analysis that could have been employed to manage the data generated in this study. The underlying premise of many of these are similar as they direct the researcher from describing to exploring and explaining underlying processes, patterns and structures (Rapley 2011). However, certain data sources offer themselves to certain methods of analysis, as well as restricting others. Consultation recordings perhaps lent themselves to exploration using conversation analysis, (Schegloff, Sacks 1973). The benefit of this would be to highlight the processes through which the clinicians and parents produced their own behaviour and understood and dealt with the behaviour of others (Heritage, Atkinson 1984), thus exploring the interaction of the two parties through their use

of language. This is something that I may consider undertaking in the future, as this would provide an alternative way of understanding the dynamics between the doctor and parents. Although detailed transcription of consultations has been undertaken verbatim, the recordings have been destroyed, as per study protocol. Therefore it will be difficult to undertake this to a 'gold-standard'.

Discourse analysis was also considered as an analytic tool for exploring the consultation recordings. This form of analysis provides a critical analysis of language, which allows insight into societal influences underlying behaviours and thoughts (Boutain 1999). My arguments against using this method reflect those made by Alison Pilnick, when she highlighted the need to move on from reconfirming the "*persistence of asymmetry*" of the doctor-patient relationship, or seeking interventions to overcome this that may be uncovered by discourse analysis (Pilnick, Dingwall 2011, pg. 1374). This was taken further by Sharrock et al, who suggested that when exploring the doctor-patient relationship, this form of analysis seeks to condemn clinicians rather than act as a form of inquiry (Sharrock 1979). Although I am unable to agree entirely with this critique, I had some reservations about the effectiveness of discourse analysis in this instance.

The interviews in the form of narratives, also lent themselves to analysis by a number of methods. Narrative analysis, for instance, could provide insight into how the women understood and made sense of their decisions (Thorne 2000). This is the approach adopted by Sandelowski in her studies on decision-making following diagnosis of a severe anomaly (Sandelowski, Jones 1996). The limitations of this analytical approach in addressing the question posed are reflected in the arguments made about the use of narratives as the sole data source (see section 3.3.2), where narratives represent a perspective of the events, which will likely be reconstructed over time. The question addressed would therefore become; 'How do women make sense of their decision to continue or terminate an affected pregnancy?'

Other forms of analysis, including framework analysis, thematic analysis and a grounded theory based approach such as constant comparative analysis were also

considered. The major difference between framework analysis and constant comparative analysis is that framework analysis maintains the integrity of the individual respondents' accounts, rather than intentionally breaking up the data (Green, Thorogood 2005). Although this may have proved beneficial when managing the interview data, use of framework analysis for analysis of consultation data would have been difficult as the interactive nature of the data would be lost (Rapley 2011). Thematic analysis has been defined as; "*A method for identifying, analysing and reporting patterns within data*" (Braun, Clarke 2006, pg. 79). This definition could also be applied to constant comparative analysis. The distinction arises from sampling, where thematic analysis can be undertaken on an existing sample whilst constant comparison requires an interaction between the collation and interpretation of the data (Braun, Clarke 2006).

3.5.2 *METHODS USED*

The constant comparative approach to data analysis, a form of analysis derived from the 'grounded theory' approach first described by Glaser and Strauss (1967) (Glaser, Strauss 2012), became my preferred method for a number of reasons. First, the datasets generated from consultations and interviews were combined and analysed together, thus enabling 'cross-checking' across participants. Second, analysing the data concurrently with the ongoing data collection enabled additional questions to be added to the interview schedule. The benefit of collecting and analysing data simultaneously was that emergent as well as anticipated themes were identified, with the opportunity to incorporate further exploration of the former. Third, in a severely under-investigated area such as this, an inductive approach to data analysis was essential. As I perceived an inductive approach as a priority, all the options considered were inductive to some degree.

Often in qualitative research, the analytical process runs concurrently with the data collection as the data already gathered are analysed and help direct the ongoing data collection (Pope *et al.* 2000). This proved the case within this study, where ongoing analysis of the data collected highlighted unexpected issues that were subsequently explored. One example was the identification of the 'making

sense of the decision' theme, where the post-termination period was perceived as an integral part of the decision-making process.

A systematic and iterative approach to analysis was undertaken. This was broadly based on the constant comparative method of analysis, where the method of comparing and contrasting the data was applied within and subsequently between each set of data. An initial phase of 'open coding' was undertaken (Strauss, Corbin 1990), where broad themes were identified. The open codes were then incrementally grouped into categories that reflected theoretical themes. These were modified and checked constantly as further open codes were added, and as new data presented negative or deviant cases. There is little literature to suggest how 'constant comparison' should be undertaken, nor which types of comparison should be made and subsequently distinguished (Boeije 2002). Within this study the methods of comparison for the parent generated data were formalised in a three-step process. First each item of data, whether interview or consultation, was coded and internal comparisons made in order to highlight difficulties or inconsistencies. Second, comparisons were made across data pertaining to the same participant. Initially this meant one consultation was compared to the next. The final set of data from each couple was the interview. This resulted in the most notable comparisons between what parents said they did (in the interview) and what they were observed to have done (in the consultations). Third, the process was repeated across the couples. Commonalities in responses were identified at this point that enabled the development of typologies. This is described further in Chapter 6, where typologies of decision-making are identified.

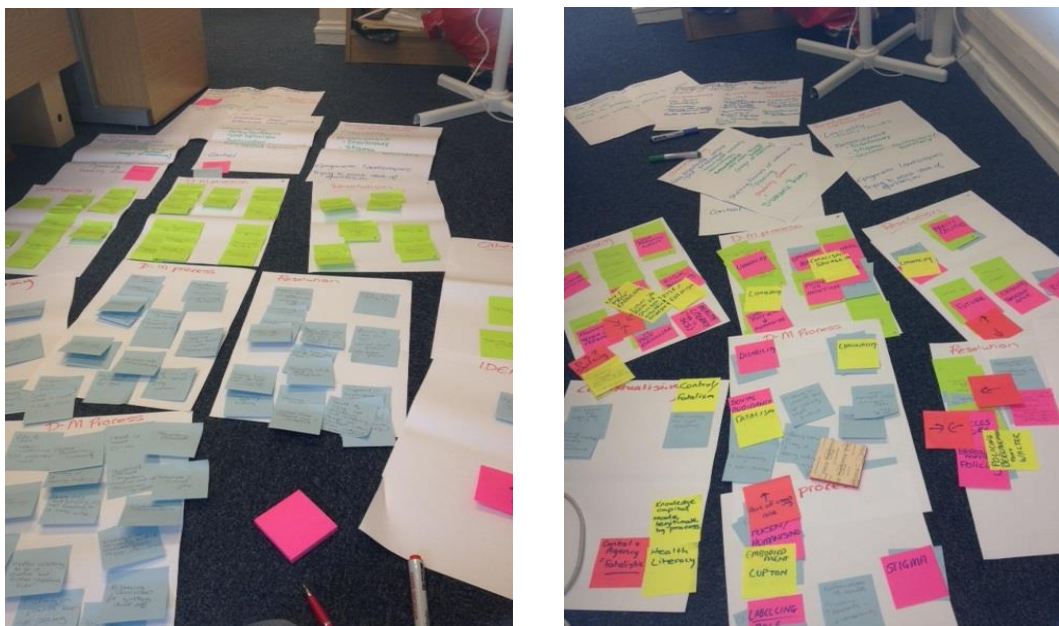
For clinicians, the same steps were applied. However, data generated from their interviews was specifically aimed at providing contextual understanding, rather than being related to any particular case. Internal and cross-case comparisons enabled an understanding of clinicians' expectations and desires in relation to the parental decision-making process. In turn, this was compared to practices observed in the consultations. However, this differed to the comparisons made in step two within the parent generated data, as it remained generalised and related

to expectations rather than actions. This becomes clearer when viewed against the findings presented in Chapter 5.

The process described above was cyclical, with the data generated by each new participant coded and then compared internally with the existing data. Eventually no new categories were identified, and cases were easily assigned to one of the pre-existing categories. At this point the categories were described as 'saturated' (Boeije 2002).

There is no software that can analyse qualitative data, or determine which issues should be coded and how to code them, but packages, such as NVIVO which was used in this study, can help with management and retrieval as well as recording memos and making links between sections of data (Tesch 1991). As a relative newcomer to NVIVO I have become aware of many of the additional functions that the software can provide. In particular the use of NVIVO to store and code literature was underused in this study, as I became aware of the function only after having completed the initial literature review. NVIVO also has limitations in respect of being able to visualise the coding tree. In order to overcome this, I amended the OSOP (one sheet of paper) method employed by HERG when managing data relating to patient experiences (Ziebland, McPherson 2006).

Figure 3-1 Data analysis adapted from OSOP



This involved reading through each section of data in turn and noting all the different issues raised by the coded extracts. These were then written on a single sheet of paper and linked with arrows. Instead of using a single sheet of paper, I used post-it notes. First, blue post-it notes, along with the respondent's ID, were grouped into themes that were labelled using green post-its, in essence axial coding (Strauss, Corbin 1990). Connections between the themes were identified and documented on pink post-its, and finally merged onto yellow post-its. This meant that ideas coded under one theme could also be coded in another. This differed slightly from the original OSOP method in that it was colour-coded for ease of identification, and that the post-it notes were moveable, making it visually easier than conveying ideas with lines between the codes on the single sheet of paper. In addition, it enabled me to separate the main concepts into chapters that have subsequently assisted writing.

In addition to this overall analysis, some linguistic analysis was embarked upon (Maynard, Heritage 2005). Issues such as word frequencies were explored, particularly in relation to the use of terminology relating to termination. Methods of analysis employed some of the functions of NVIVO, thus providing additional insight into the experience of the parents. Other quantitative aspects, including percentage of speaking each partner contributed to a consultation and how that changed temporally, provided additional insights that contributed to my overall understanding.

3.6 METHODOLOGICAL QUALITY AND RIGOUR

This section provides a description of the governance processes undertaken. In addition it addresses the 'quality' issues surrounding the study and presents the steps taken to ensure the findings are robust.

The ontological and epistemological debate is not isolated to choice of methodology and arguably has become more pertinent in the defining of 'quality' of research (Giorgio 1992, Mishler 1990). The concepts of reliability (would the same findings be achieved if the study was replicated), validity (how accurately it

reflects the phenomenon it set out to measure) and the resulting generalisability (ability to apply the findings to a different sample) are well established methods of judging quality in studies where the objectivity of findings is deemed essential. However, the suitability of these measures is fiercely disputed by those outside the positivist paradigm. Attempts to resolve this issue range between the extremes of adopting these quantitative criteria for validity to disregarding validity as an issue in qualitative approaches to research (Silverman 1993). These poles are representative of the differing underlying ontological and epistemological beliefs, namely those supportive of the realist ontology (on which positivist science is based) and those whose stance is more representative of the idealist perspective and tend towards “*releasing research from the stranglehold of validity as truth*” (Angen 2000, pg.379).

As stated earlier in the chapter, my ontological stance is consistent with that of subtle realism (Hammersley 1992, Silverman 1993) where an external reality exists (consistent with the realists) but like the idealist, we can know reality only from our own perspective on it (Ritchie *et al.* 2014). In terms of quality, acceptance of an external reality enables judgements on the credibility of the research to be made. Nonetheless, a distance from the validity associated with a ‘pure’ realist perspective must be gained if we accept reality can be known only through our own perspective, and not through an objective lens. One way of dealing with this has been through the use of a different set of terminology. A widely accepted set of criteria that runs parallel to that of the positivists has been proposed by Guba (Guba 1981). These are: a) credibility (in preference to internal validity); b) dependability (in preference to reliability); and c) transferability (in preference to external validity/generalisability).

All three concepts express aspects of the quality and trustworthiness of the research. Credibility refers to the confidence in which the data and processes of analysis address the proposed question (Polit, Hungler 1999). Transferability refers to “*the extent to which the findings can be transferred to other settings or groups*” (Polit, Hungler 1999, pg.717), while dependability “seeks means for taking into account both factors of instability and factors of phenomenal or design

induced changes” (Lincoln, Guba 1985, pg.299). The use of this alternative terminology has been selected in this instance as it offers a consistency in terms of the ontological and epistemological stance taken.

The concept of credibility deals with the question; “*How congruent are the findings with reality?*” (Merriam 2009, pg.213). Lincoln and Guba argue that ensuring credibility is one of most important factors in establishing trustworthiness (Lincoln, Guba 1985). In this instance I have utilised Creswell’s criteria for assessing ‘credibility’ in qualitative research. His criteria include prolonged engagement, triangulation, peer review or debriefing, negative case analysis, clarifying researcher bias, member checks, thick description, and external audits, with at least two of these required for a credible study (Creswell, Miller 2000).

A number of Creswell’s criteria have been met within this study. First, several peer review and member check activities were incorporated within the study. As previously discussed, an extensive PPI exercise was undertaken, with ongoing engagement throughout the process. Their input in the design stage ensured a smooth progression through governance processes. However, in terms of credibility, their input in relation to the interpretation of the findings has been most influential. Analysis and interpretation of findings was enriched through the informal presentation of the preliminary findings to members of the PPI group. Each member was able to identify aspects which ‘rang true’ to their own experiences, thus testing the proposed findings and subsequent recommendations. Discussions and feedback from some clinical staff, particularly senior midwives, were more stilted. This was principally true where aspects of care highlighted did not meet anticipated standards. Great care was taken to present findings in a non-confrontational manner, without attributing any blame. One recurring theme was the different priorities of midwives and the parents interviewed. Criticisms levelled at the recommendations by the senior midwives in particular, predominantly stemmed from their failure to engage with the parents’ alternative perspective to many issues. This issue was pertinent to other clinical staff, but to a much lesser extent.

In addition, in order to progress through the PhD process, a number of peer reviews were required. For example, the Advanced Post-Graduate (APG) process for transfer from initial registration to full PhD student status required that a presentation be given to colleagues, followed by a detailed discussion of my work to date and future plans. In addition, a number of other peer reviewed presentations have been given, including at the Medical Sociology (MedSoc) Conference⁸ and ARC.

From a methodological perspective, triangulation of data collection and analysis was central to the process of ensuring credibility (Kimchi *et al.* 1991), where use of consultation recordings alongside interviews enabled varying perspectives to be compared. A systematic difference between the way in which patients presented their views in the consultations and research interviews has been well documented in other studies utilising this triangulated approach to data collection (Barry *et al.* 2000). Discrepancies between what parents report and what is heard from the consultation recordings are informative (Shilling *et al.* 2011), as they raise issues surrounding parents' understanding and interpretation. The triangulation of the consultation and interview data is therefore a powerful method for understanding communication processes (Burkitt-Wright *et al.* 2004, Barry *et al.* 2000), and the impact of these on patient decision-making.

Although comparison was the predominant method of analysis, application of some 'linguist' approaches, such as word frequencies, offered a different perspective. One of the benefits of using the OSOP approach to managing the data is that deviant or negative cases that did not fit with the emerging story are immediately identifiable and explanations can be sought so that all data collected are accounted for (Ziebland, McPherson 2006).

⁸ MedSoc is a part of the British Sociology Association that promotes scholarship and communication in the field of the sociology of health and illness.

Credibility and dependability are tightly entwined, as demonstration of the former goes some distance to ensuring the latter (Lincoln, Guba 1985). When addressing dependability, techniques employed were aimed at ensuring the consistency of the findings. This is problematic in the qualitative field because of the changing nature of the phenomena studied. However, through careful documentation of the processes undertaken, the study could be reproduced even if the subsequent findings differ.

In positivist work, the convention of generalisability of findings is widely stressed (Winter 2000). Since the findings of a qualitative project are specific to a small number of particular environments and individuals, it may not be possible or even desirable to demonstrate applicability to other situations and populations. Nonetheless, by providing sufficient contextual information, it may be possible for a reader to make such a transfer (Shenton 2004). Within this study, peculiarities within each hospital centre may reduce the transferability of the findings across the UK. However, I took the strategic decision to incorporate a broadly inclusive sampling frame. In this way, I believe it is possible to argue that this study has a high degree of conceptual transferability and that insights may provide useful pointers for public health policy and practice.

3.6.1 RESEARCH ETHICS AND GOVERNANCE

Summarising the ethics and governance processes in a few lines significantly understates the preparation and work involved in planning and undertaking these. A comprehensive reflection of this work is therefore presented in chapter four.

The study was sponsored by the University of Leicester. Following submission of all the study documents to the University, formal ethical approval was sought and obtained from the Nottingham Research Ethics Committee 1, REC reference 13/EM/0293. Appropriate governance approvals were subsequently sought from the relevant NHS organisations.

3.7 SUMMARY

In this chapter I have endeavoured to provide an honest, detailed overview of the logic for my chosen methods of data collection and analysis. In addition, reference is made to how the planned approach was actually enacted in practice. As a 'subtle realist', my research perspective aligns with the belief that an external reality exists but the meanings attached to this reality are socially constructed. With the existence of an external reality, the degree to which the findings, arising from the methodology employed, can be manipulated through the adherence to principles, inform the quality of the research. Assurance of the credibility, dependability and transferability of the findings help to ensure quality. Mechanisms employed within this study include the triangulation of data sources, namely interviews and consultation recordings, and have enabled a cross referencing of perspectives from the same participant. In addition, the temporal aspect to the consultations provides a view on how the decision-making process emerges.

The next chapter provides further insight into the choices made through identification of ethical and methodological aspects specific to the conduct of sensitive health research.

4 ETHICS AND METHODOLOGICAL PITFALLS IN SENSITIVE HEALTH RESEARCH

As a novice researcher, designing a qualitative study around the ethically sensitive and emotionally charged area of termination of pregnancies affected by suspected severe congenital anomalies has proved challenging. This chapter provides a description and discussion of a number of issues that have arisen during the conceptualisation of the study, alongside ethical and methodological dilemmas encountered during the process of research.

4.1 SENSITIVE RESEARCH

Despite arguments that any research has the potential to be sensitive (Lee, Renzetti 1990) there are perhaps some areas that are likely to prove more problematic than others (Gibson 1996). Unsurprisingly ‘sensitive research’ has been used to describe a broad spectrum of subject matter (Dickson-Swift *et al.* 2008), with the precise definition being open to debate (Elmir *et al.* 2011). The seminal definition presented by Lee and Renzetti is that of research that *“potentially poses for those involved a substantial threat, the emergence of which renders problematic for the researcher and/or the researched the collection, holding and/or the dissemination of research data”* (Lee, Renzetti 1990 p.5). The value of this definition is perhaps the inclusion of potential risk to researched and researcher, something that remains a highly relevant and under-investigated issue (Fahie 2013). The topic investigated in this instance clearly encompasses these characteristics, where issues pertaining to bereavement, the loss of an infant and also the loss of the opportunity to experience a ‘normal pregnancy’, alongside the stigmatised area of termination, have been explored. Investigating topics such as this can pose considerable challenges for all researchers (Dickson-Swift *et al.* 2009) and particularly inexperienced ones (Bloor *et al.* 2008). Seiber expresses this eloquently, stating; *“although there is nothing that forbids research on sensitive topics, there are powerful forces against the conduct of such research”* (Seiber, Stanley 1998 p.61).

4.2 DETERMINING THE FEASIBILITY

The feasibility of this study relied heavily on engagement. Having spent many years working within the NHS as a clinical and subsequently research nurse, I have identified a number of strategies that have improved recruitment rates of research studies undertaken in the clinical setting. The main strategy implemented was early engagement of the key gatekeepers and stakeholders within the health service, namely the clinicians. Their influential role in recruitment is well established in the literature, with early involvement and good communication demonstrated to be essential in the access to and recruitment of patients (Fletcher *et al.* 2012).

Alongside this group of clinicians, a number of local and national parents' groups and charities were approached, with representatives providing some insight into the needs of the parents. 'Adverts' asking for parents to volunteer to advise on the study development were put out by a number of these local charities via social media. I was therefore able to identify parents who were not usually actively engaged in parents' groups, but rather those who sought support from time to time. This meant that the parents were generally less publically vocal and more detached. I hope that this enabled me to elicit a more informal and less 'corporate' view on the study. I have fostered these networks over a number of years, in relation to public engagement following national policy changes pertaining to care of severely disabled children. Although this proved to be a particularly successful mechanism, attempts to reproduce this may prove less effective unless existing networks can be identified and accessed.

A group of parents, broadly representative of the population of interest, was selected (see Appendix B for additional details). The parents were spoken to individually and it was made clear that this aspect of the research development would not constitute any part of the actual research, but rather was preliminary work to investigate the acceptability and importance of the research from the parents' perspective. As demonstrated in box 4-1, all the parents were very supportive of the research aims. There appeared to be a general consensus that

research in this area was lacking and an increase in awareness of issues would be beneficial to families and staff.

Box 4-1 Parents' reflections on the appropriateness of the research

"Telling his story is a celebration of his life"

"We felt very isolated after we chose to terminate, I mean you cannot really ask for help because it was our decision after all"

"I really want to share her story, it gives her a purpose"

There was a general feeling that talking about their experience legitimised their choices and gave meaning to their pregnancies. For those who terminated an affected pregnancy, difficulties in overcoming the stigma attached to termination were mentioned, along with a need to validate the baby's life.

As an additional method of gaining insight into the potential pitfalls of the study area, I contacted a number of researchers who have published articles on associated issues. Of those who responded, none expressed any specific problems with recruitment once initial barriers pertaining to access had been overcome. As had been noted in other areas of sensitive research, exploring these areas often provided participants with the opportunity to express their perspective, something that was otherwise often denied (Sque 2000). However, these sentiments drew attention to the sharp contrast between these perspectives and the insignificant body of literature investigating the views and experiences of this population. This perhaps suggests an avoidance of the topic by researchers, rather than lack of willingness to participate from the target population.

Other aspects of good practice were also highlighted. These included carrying a list of support organisations and contact numbers to hand out if required when interviewing, and sending 'Thank you' cards or messages a few days after the interview. These suggestions were both adopted in the study.

The involvement of clinicians and parents from the inception of the study proved to be immensely beneficial in terms of ensuring acceptability and feasibility of proposed methods and thus overcoming a number of the ethical and methodological challenges. In particular, navigation through the governance process was facilitated predominantly by undertaking this process of engagement. This will be discussed in depth in section 4.3 below.

4.3 GATEKEEPERS

4.3.1 *ETHICS COMMITTEE*

Four fundamental principles of ethics are usually identified: autonomy, beneficence, non-maleficence and justice (Beauchamp, Childress 2001). In medical research ethics, the principles of beneficence and non-maleficence translate into the duties to maximise benefits while minimising harm for the research subjects (Tangwa 2009), autonomy is regarded as making our own decisions on the basis of deliberation, and justice is often regarded as being synonymous with fairness (Gillon 1994). In England, all research proposals involving human participants recruited from the National Health Service are subject to scrutiny from a REC. Its role is to protect the rights and interests of research participants by balancing these potentially conflicting principles (Garrard, Dawson 2005).

Although qualitative research is becoming more widely accepted, data generation in studies of this type is often flagged by RECs as an issue (Hadjistavropoulos, Smythe 2001). These concerns are magnified when related to sensitive topics (Buckle *et al.* 2010). The rationale for the concerns frequently stems from the unpredictable nature of a narrative interview, leading to suggestions that the interview may result in distress (Hadjistavropoulos, Smythe 2001, Guillemin, Gillam 2004). Kellehear made this point quite sharply in relation to qualitative research; “*The interview is the creation of an unnatural social situation, introduced by a researcher, for the purpose of polite interrogation. It is this situation, which is ethically questionable*” (Kellehear 1993 p.10). This concern was reflected in suggestions that research proposals exploring sensitive issues were more likely to

be rejected by RECs compared to other proposals (Ceci *et al.* 1985). Although it is unclear whether this is still the case, more recent studies suggest that lack of understanding of qualitative research by RECs remains an issue (Daly *et al.* 2008, Larkin *et al.* 2008). RECs have an important role to play in safeguarding the interests of the most vulnerable in society in the research process (Guillemin, Gillam 2004). However, overly protective or paternalistic RECs risk excluding marginalised groups from having their voices represented in research (Hannigan, Allen 2003), and threaten their autonomy.

The lack of literature pertaining to women's experiences following diagnosis or suspicion of a severe congenital anomaly is perhaps a reflection of the ethical difficulties encountered, or perhaps even just feared, by the research community when approaching such a sensitive topic. Prioritisation of research enquiry of any one topic over another is likely to result in inequitable service development, as practice will be unsupported by evidence. These concerns are heightened when placed in the context of a group who may be considered to be disenfranchised. As previously discussed, the lack of research does not appear to be representative of a lack of desire to participate by the population of interest. Rather, this could reflect a sense of discomfort on the part of the research community. Certainly some of this may stem from the difficulties encountered, as suggested above. From her experience researching bereavement, Sque suggests a certain tenacity is required by the researcher in order not to be deterred when faced with difficulties of access to study populations or low participant response. However, alongside this there needs to be sensitivity about how any potential damaging outcomes may be mitigated, and an acceptance of the probable benefits of such research to the participants (Sque 2000).

Within this study, an attempt to demonstrate to the REC, as gatekeepers, a balance between these tensions was approached through careful engagement with key stakeholders, including the population of interest. Choice of methodology, timing of approach and interview, and skills required by the interviewer were all included in a careful assessment. Designing study documents alongside stakeholders was a novel experience. The final participant information sheets were marginally

unorthodox, and consisted of small coloured boxes containing word summaries of the main points (see Appendix G). This was the focus of a number of questions from the REC, but went unchanged following reassurance that they had been designed in collaboration with users. Other concerns raised by the REC stemmed from the appropriateness of the proposed approach, in particular the initial recruitment of parents. Assuring them of the involvement of parents in its planning provided sufficient reassurance for approval to be granted without any changes being required.

Once ethics approval was in place, NHS governance approvals were sought and obtained prior to starting data collection. Despite considerable effort to anticipate all ethical eventualities, ethical considerations are interwoven throughout the research process and dilemmas cannot always be anticipated (Morrow 2008). Effective and ethical research demands ongoing reflexivity on the part of the researcher (Noble-Carr 2006). In this instance no additional approvals or amendments were required or sought. It is likely that the additional time spent preparing and anticipating all eventualities, although frustrating at the time, meant that subsequent changes were not necessary

4.3.2 *CLINICIANS AND PARTNERS*

Two further groups of potential gatekeepers were identified during the planning stage of the study, namely clinicians and women's partners. As with the REC, these gatekeepers were keen to weigh up the ethical benefits and possible harms of the study before 'allowing' women or their partners to be approached. Although cautious assessment of the researcher and research is essential, the risk of being overzealous in their paternalism has the potential to force researchers to use inadequate samples, and raises not only methodological challenges but ethical dilemmas. This is a risk frequently discussed in the area of bereavement research (Hutchinson *et al.* 1994, Cartwright, Seale 1990, Sque 2000). Within the context of this study, stakeholder involvement proved essential in overcoming some of these threats. Apart from providing me with reassurance and reinforcing my conviction of the importance of the study, engagement of clinicians at an early stage allowed concerns to be expressed and solutions found. A number of simple problems had

been identified, namely concerns over video recording consultations, time requirements for the consent process, and the potential to cause distress. The first two issues were overcome by agreeing to record using voice recorders only and by making it clear that I would undertake the formal aspects of the consent process myself. The issue of inducing distress was far more difficult to overcome. There are suggestions that clinicians 'over worry' about this aspect, and people who are not ready to talk will decline the invitation to take part (Hutchinson *et al.* 1994, Cartwright, Seale 1990). Although there is little research in relation to the experiences of pregnant women following diagnosis or suspicion of a severe congenital anomaly, parallels can be drawn from bereavement research. Sque talks about families wanting to be helpful, interviews being therapeutic and providing an opportunity to reflect, and of intrusion even being welcome (Sque 2000). Furthermore, it is suggested that following bereavement many people "*are glad to find that their experiences, however awful, can be of help to others*" (Cartwright, Seale 1990 p.36). This was a sentiment shared by bereaved parents (Hyson *et al.* 2006). Experience from this study reflected altruistic perceptions, from the advisory group of parents, but also subsequently from participants, some of whom took the time to email and thank me for taking the time to listen. Convincing clinicians of the potential benefits, prospectively, was more complex, and I believe was primarily achieved through their involvement from the conception stage of the study, and subsequently through the development of trust with me as the researcher. My role as a nurse may have been influential at this point and will be reflected on next.

4.4 DUAL IDENTITIES

As a nurse with over 20 years' experience of working in the NHS, it is inevitable that I have developed professional biases, assumptions and values. The change in role from clinician to researcher has required adjustment that has not been linear. Some of this has been related to the way in which I have presented myself or been perceived by clinicians in the area. In some instances, it was useful to use existing clinical network relationships to gain access to interviews (with the clinicians themselves) and then through them to facilitate patient recruitment. Being

perceived as a student and novice researcher with no explicit personal academic record to support requests for access, it was necessary to use the reputation and status of supervisors to validate the research.

In practice, my nursing persona enabled me to access areas that may have been restricted to others. Whilst awaiting governance approvals, I was invited, as a nurse, to sit in on clinics and observe the workings of the fetal medicine environment. Not only did this enable a better understanding of the system, but also allowed time for trust to develop between myself and clinicians as gatekeepers. This was instrumental in overcoming methodological issues relating to recruitment and sample size. As one clinician stated: *"Oh, I know I can trust you, I've watched you, and I like the way you approach my patients."* Although my role as a nurse proved beneficial in addressing a number of the methodological issues, including access and recruitment, a number of other ethical questions arose from my explicit use of a dual identity, as will be discussed in section 4.5.

4.5 INTERVIEWING

Creation of data through the interview is a joint venture and is based on the ability of both parties (researcher and participant) to engage in discourse production where the participant is the expert narrator of her experiences (Leslie, McAllister 2002). Therefore the impact of my dual identity is reflected not only in the way I approach the participant, but also in the participants' expectations and perception of me, as a nurse and researcher.

Leslie and McAllister coined the term 'nursedness', referring to the uniqueness of a nurse's role that relates to the ability to make the extraordinary ordinary (Leslie, McAllister 2002). They argue that there is something about the way the role is socially constructed and perceived which gives people permission to talk about social taboos, and an intimacy within the relationship that encourages disclosure (Leslie, McAllister 2002). This enabled a reframing of the research from working on, to working with, and for, participants (McWilliam *et al.* 1997). Despite the unique perspective this may have generated in terms of data collection, a potential

risk lay in the participants' expectations of the interview. It has been suggested that difficulties may be experienced by participants in differentiating between a therapeutic and research interview (Kvale 1996). Both may promote understanding and change. However, research interviews emphasise changes in intellectual understanding, whilst therapy interviews consider personal change (Sque 2000). Despite this, enabling women to tell their story can be therapeutic and thus could be regarded as "*producing a positive outcome for them that is a beneficent act*" (Cutcliffe, Ramcharan 2002 p.1003).

Furthermore it has been argued that by persisting with a desire for firm boundaries between the roles of researcher and clinician or therapist, we deny the benefits this dual role can offer to participants (Crotty 1998), particularly in terms of empowerment (Leslie, McAllister 2002).

4.5.1 SUPPORT FOR THE RESEARCHER

Emotional engagement as part of the interview process forms a substantial volume of literature (McGarry 2010). However little is said about the emotional disengagement after the interview (McGarry 2010). Within the context of my study, I found talking with colleagues was immensely beneficial, but also found that not all colleagues felt comfortable discussing my experiences. Due to the sensitive nature of the topic, discussion outside work was often not feasible. The frequent discomfort expressed by others over my research topic gave me some insight into the reactions participants face in their everyday lives, something reflected in research literature relating to impact on the researcher when exploring women's views on termination (Goodrum, Keys 2007).

One of the unforeseen benefits to involvement of parents in the planning process was the opportunity to listen to their narratives, relayed to me with the benefit of passing time. This raised my awareness, from the outset, of the need to consider methods to manage the difficulties I was likely to encounter during the research process. These included access to counselling through a national organisation if needed, as well as timetabling my interviews in such a way that I had the chance to process each one, before being faced with another. Within the literature there are

a number of specific and practical strategies for managing some of the difficulties faced by researchers such as myself: these include self-care, social networks, support and reflexivity (Zurbriggen 2002). Being aware of the issues I was likely to encounter enabled me to instigate a number of these strategies. Unlike clinical work, where the healthcare professional is embedded within a team, researchers frequently work alone in the field. In this scenario, the dual persona left me on the outskirts of the clinical team. Hence, although I was privy to their chatter, the boundary between roles did not enable me to cross the border and become 'part of the team'. Thus the support offered to the team was not available to me as a researcher. However, I was often privy to news about the death of the neonates that were born to parents that had participated in the study. On one level, I found the 'completion' of the story beneficial, and was able to construct a positive narrative relating to the outcome. However, this information often brought immense sadness, when the outcome was worse than had been anticipated antenatally. This was particularly true in relation to a neonate born with spina bifida. Although the parents had anticipated severe disability, the death of the neonate within 48 hours of birth was unanticipated. Managing my emotions at this point was difficult, due to the relationship that I had developed with the parents. Had I been less 'involved' with the clinical team, it is unlikely that I would have heard the news. Both achieving closure (by knowing the outcome) and living with uncertainty (when the outcome is imagined) have their risks and benefits. However the blurring of roles between clinician and researcher can, as in this example, add to the stresses of managing a study investigating a sensitive topic, without the benefits of being part of the clinical team. The importance of ensuring support systems are in place is heightened.

Listening to the narratives of the PPI group also enabled me to desensitise from some of the emotional aspects with which I was to come into contact during recruitment and data collection. Frank discussion ensued with some of the more vocal research advisory parents, who suggested appropriate techniques to 'manage' parents who became emotional during interviews. Interestingly, over the course of the study, I found that I was able to process my own emotions much

more efficiently, the more stories I heard. This is a phenomenon reflected in the literature (Birch, Miller 2000).

There are suggestions that we, as researchers, should do more to imagine, discuss and implement strategies for managing the negative effects of research on sensitive topics (Zurbriggen 2002). However, it is unlikely that we will ever anticipate everything so; *"it is wise to remember the unexpected will probably happen"* (Zurbriggen 2002 p.255). Thus it is essential that we attempt to negotiate strategies to minimise negative effects not only for our participants but for ourselves as researchers as well.

Within the context of the study I found this blurring of identities difficult to manage. I had drawn on my clinical background in order to gain access to participants, and felt that my skills as a nurse were beneficial in 'managing' the emotiveness and suffering of the participants I interviewed. Nonetheless, I was always wary of the potential for the role of the interview to be misconstrued and remained aware of the power differentials that potentially existed in relation to the data generated, but also in terms of the risk of coercion. The impact of this dualism was not solely directed towards the participants, but also affected me as researcher. Alongside the feelings of immense privilege, at having incredibly personal stories generously shared with me, I also found myself feeling frustrated at times with failures in the system, or perceived episodes of 'poor care'. This phenomenon is reflected in literature surrounding the blurring of roles, when undertaking sensitive research (Lee 1993).

4.5.2 REFLECTIONS

Maintaining momentum for a PhD study over a prolonged period is undoubtedly a major challenge. After many years working within the clinical setting, the opportunity to 'make a difference' at another level has proved to be the driving force necessary for me to reach this point where I can present and discuss the findings from my study. In order to achieve my aim of making a difference, the next chapter provides my overview and description of the journey undertaken by parents following diagnosis or suspicion of a severe anomaly.

In September 2014 I attended an international conference where I was fortunate to get the opportunity to have a one-to-one session with the renowned sociologist Arthur Franks (Franks 2014). One piece of advice he gave was to write about what kept me awake at night. Reading back through much of what I have written, I've realised that there is a strong focus on the experiences of the women who terminated that, although not intentional at the outset, is perhaps a reflection of what remains prominent in my mind. It has personally been an emotional rollercoaster, not just in terms of the stresses associated with undertaking a PhD, but more particularly in relation to the subject matter itself. Many of the words spoken in the interviews, particularly by the women who terminated their pregnancies, still reverberate through my consciousness, and the image of the raw emotion drawn on their faces as they described their loss, remains etched on my mind. Perhaps one of the main differences between the women who terminated and those who continued was that those who continued had not yet had to face the physical reality of their loss; they were still pregnant and time was still their friend (Hedrick 2005). Despite the intense difficulties that they would subsequently face, a small glimmer of hope remained. The subsequent three chapters provide a temporal narrative of the experiences of these women and their partners following the suspicion and/or diagnosis of a severe congenital anomaly, with their words permanently committed to the pages of this thesis.

5 FINDINGS 1 - EXPLORING THE CONTEXTUAL FRAMEWORK

The findings that I will now present have emerged from the dataset created through the triangulation of data from interviews with parents and clinicians, and recordings of consultations. The decision-making processes are not enacted in a vacuum, but within a contextual framework constituting multiple interdependent layers including individual, social, healthcare and wider legal and professional factors. This chapter provides an exploration of these layers and the fluidity of their borders. This is extended in the next chapter where a conceptual model of decision-making is developed and the common characteristics of parents are explored. Tensions arising from the gap between the decision-making process applied by parents and that desired by clinicians are discussed. The final findings chapter focusses predominantly on those pregnancies ending in termination. It explores how the parents made sense of their decision, and the practical difficulties they encountered on their journey.

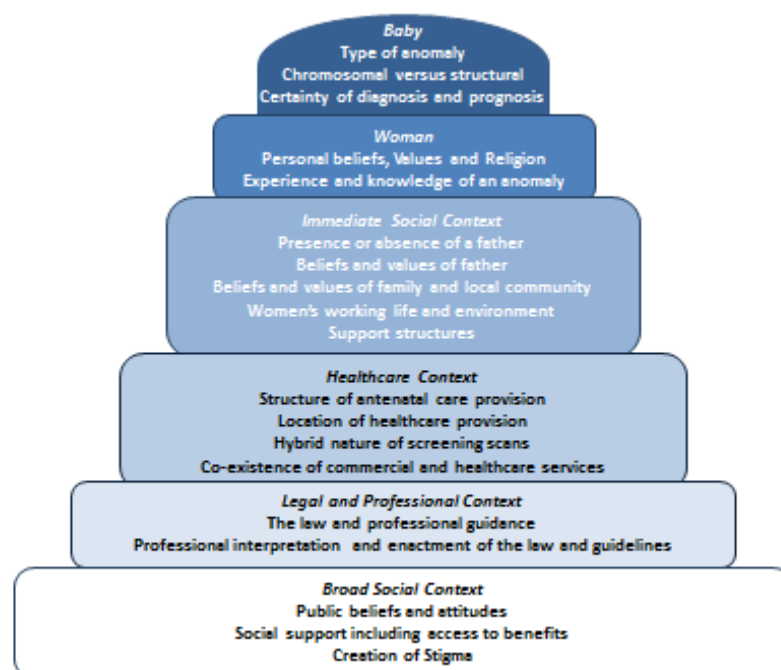
5.1 THE CONTEXTUAL FRAMEWORK

Contexts have existential, representational and denotative meanings which, through experience, inform and shape understanding of situations and events (Shedroff 2000). Exploring the contextual framework provides a starting point from which to examine the varying decision-making processes experienced by parents.

Through my analysis, five contextual layers were identified and are graphically represented in Figure 5-1. The pregnant woman has sole legal responsibility for the decision and the first contextual layer relates to her baby and the anomaly that affects them. Anomalies are perceived in different ways; for example, chromosomal anomalies are more likely to be attributed a lethal status (Zyblewski *et al.* 2009), whilst structural anomalies may be seen as repairable. This is further influenced by individual attributes of the mother, including personal beliefs, values and previous experience of a particular anomaly (France *et al.* 2012). These factors represent the second contextual layer. These are further influenced by her

immediate social context comprising of the effect of a partner's beliefs, family and local social support systems. These three layers have a direct impact on the decision-making process and have been labelled, for the purposes of this thesis, as the 'internal context' (represented by white writing on Figure 5-1). Each woman experiences the influence of these contextual layers in a unique way. As discussed in Chapter 2, much quantitative literature has been dedicated to isolating these factors or variables (Chenni *et al.* 2012, Feijen-de Jong *et al.* 2011, Reid *et al.* 2009), although the difficulty in reducing the process to a combination of variables is perhaps demonstrated by the conflicting findings within some of these studies.

Figure 5-1 Summary of factors contributing to the contextual framework



Underlying the immediate social context is the healthcare context, in which care is enacted. Analysis of the data highlighted a number of factors, including the way in which antenatal care was structured and located and parental perception and clinician enactment of screening, including the dual social and medical role of ultrasound scans. Risk and uncertainty and the framing of unknown anomalies against others added a further dimension.

The next layer has been entitled the legal and professional context. This encompasses the law alongside professional guidelines and principles. The way in

which these were interpreted by clinicians provided the framework in which they constructed their role and subsequently defined an ideal decision-making process.

In turn, the boundaries created by legal and professional guidelines are further constrained by public perception. Formal and informal mechanisms created by clinicians in order to distance themselves from the decisions of the parents are evident and can be attributed in part to the prevailing social environment of the broad social context where termination is heavily scrutinised and stigmatised. Together the healthcare, legal and professional and broader social context creates an 'outer contextual layer' (represented by the blue/black writing in Figure 5-1).

This chapter provides an examination of the outer contextual layers, with the intention of examining the common framework in which the varying decision-making processes were enacted. The importance of the individual attributes of the women and their babies, highlighted within the internal contextual layers, will be given consideration in subsequent chapters.

5.2 THE HEALTHCARE CONTEXT

5.2.1 *SCREENING*

The antenatal screening process creates opportunities for the identification of anomalies at various points in the pregnancy. A combination of tests is used, as described in detail in section 1.3. Some of these provide a 'risk' calculation (such as the nuchal translucency measurement taken in conjunction with a blood test) whilst others provide a combination of diagnostic testing and screening (such as the anomaly scan). These findings are subsequently used to direct care and inform decisions on when to offer, or accept, further diagnostic testing. There is increasing evidence to suggest that parental understanding of serum screening differs significantly to that of screening undertaken via ultrasound, where serum blood tests are associated with identification of abnormalities (Santalahti *et al.* 1998). Two issues thus arise, both well documented in the literature. The first is that of the construction of scanning as a social and medical event (Lupton 2013,

Williams *et al.* 2005, Mitchell 2001, Roberts 2012), and second is that of risk and interpreting risk (Pilnick *et al.* 2004, Williams 2006, Georgsson Öhman *et al.* 2009).

5.2.2 *JUST A PRETTY PICTURE*

The hybrid nature of the antenatal ultrasound scan has long been acknowledged and debated (Roberts 2012). The medical and social roles of the scan are firmly embedded in routine clinical practice (Mitchell 2001, Roberts 2012), but their boundaries are fluid and blurred (Mitchell 2001), and potentially impossible to differentiate (van Dijck 2005). In addition, evidence suggests that women frequently do not have a full understanding of the clinical purpose of ultrasound (Garcia *et al.* 2002, Thorpe *et al.* 1993).

Data from this study reveal the varying perceptions of parents on the purpose of the anomaly scan⁹ in particular. Some parents stressed the social nature of the procedure for ‘meeting’ the new baby whilst others stated that their objective was more clinical in terms of having a ‘look to see if there were any problems’. There was also a spectrum of understanding between parents over the purpose of the anomaly scan that perhaps reflects the shifting boundaries of the social and medical worlds of the ultrasound scan. Although not universally expressed, many parents saw screening consultations as enjoyable days out and opportunities to welcome new family members. Speaking of their anomaly scan:

*We took [daughter] with us to meet her new brother
[talking about attending the anomaly scan] (Mother16 –
Terminated)*

Another mother talked about the importance of the photograph taken at the anomaly scan and combined this with a medicalised perspective:

⁹ The anomaly scan is performed between 18-20 weeks gestation, in order to examine for any structural abnormalities, or soft markers for chromosomal anomalies.

The measurements were all fine and I'd read up on it myself extensively so I was quite comfortable with the whole scan myself. (Mother20 – Terminated)

There is some evidence derived from an ethnographic study that highlights the healthcare professional's role in reinforcing the social nature of the ultrasound scan, through the humanising of the fetus (Thomas 2014). In the event that an anomaly is suspected or identified, this enactment abruptly stops. They distance themselves from parents and refrain from engaging further about what is seen on the ultrasound image, in an attempt not to contribute further to "*the enactment of the image on the screen as a living child*" (Schwennesen, Koch 2012 pg.290). Most of the parents interviewed demonstrated a high level of "*cue consciousness*" (Ramsden 2003 pg.182), where they sensed that something was amiss during the scan. For some, this was the change in tone of the sonographer or clinician scanning, for others it was the apparent focus or rescanning of a particular area, or the call for a second opinion. Nonetheless all the parents involved made sense of the changing atmosphere with hindsight:

And you know don't you, there are tell-tale signs ... you can tell by somebody's face and you can tell that something's not right 'cause they go quiet. And then they put you in that counselling room and you think; "This is not good". You know. (Mother14 - Terminated)

5.2.3 COMMUNICATING RISK AND UNCERTAINTY

Much of the screening and diagnostic process revolves around the concept of risk and uncertainty, be that in relation to communicating risk by clinicians, or parental understanding and tolerance of risk. Acceptance or rejection of screening by parents, weighing up the risks of additional testing against the potential for a definitive diagnosis, along with ongoing uncertainty relating to prognoses, requires skilful navigation and management. For the clinicians, caring for parents when options are clouded by uncertainty is equally complex. In a speciality where litigation is increasingly common, communicating these risks whilst managing

parents' expectations within the confines of the law and professional guidelines results in unresolved tensions.

Although the concept of screening is based on risk assessment, many parents appeared uninformed about the function of screening tests until after their status changed to that of 'high risk'. This is a status endowed on parents when the risk of an anomaly was perceived as being raised. This could stem from the identification of a 'soft marker' on any scan, or a raised risk profile calculated following the nuchal translucency measurement¹⁰. Hospitals vary according to the cut-off points used when defining high risk. Therefore, in one hospital, a high risk result could be more than 1 in 100 chance of having an anomaly, whilst in others the risk would need to be greater than 1 in 150. This risk would then entitle parents to access free diagnostic testing. Although the diagnostic tests would provide a guaranteed result for some chromosomal anomalies, accessing this knowledge comes with its own risk, with amniocentesis and CVS carrying a risk of around 1 in 100 chance of miscarriage. In addition, confirmation of a diagnosis of a condition cannot be directly translated into an outcome or prognosis, and further risk and uncertainty must be balanced with the potential for suffering. Balancing the likely outcome of the tests and the risk associated was a difficulty encountered by many of the parents:

That [risk] was difficult to get your head round. We couldn't get our head round that at all. Because it did not add up at all. So percentages are supposed to be out of 100 aren't they? And [the clinician] was like there is a 60% chance it is chromosomal, then [the clinician] broke it down and it was like, I'm sure that added up to 120. (Mother19 – Terminated)

¹⁰ The nuchal translucency (NT) is a collection of fluid beneath the fetal skin at the back of the neck. The fluid collection is increased in many fetuses with chromosomal abnormalities. A calculation incorporating maternal age and NT measurement creates the basis of the risk figure is derived.

Some parents sought to dismiss the risk as ‘only data’, or sought additional information which disproved the risk:

I knew they were checking for hormone levels in the blood and if it was high and plus the NT and my age goes into a computer and then I get a percentage and it is only data and it's never 100% accurate. And I was reading stuff online and they were saying that they had a really high NT and things were ok. (Mother12 – Terminated)

Others appeared more resigned in their interpretation and acceptance:

I got the odds of 1 in 5. I knew 7 people who were pregnant at work and they were all fine, and I just knew that I was the one they had everything come back clear so it was bound to be us. (Mother9 – Terminated)

The difficulties encountered by clinicians and parents alike in managing risk and uncertainty were widely discussed by clinicians:

...actually weighing up the pros and cons of probability and risk is probably quite a hard thing to do (Clinician05)

Assessing parental understanding of risk in relation to initial screening was generally a task delegated to midwives. Therefore clinicians’ experience of managing data in the form of probabilities and risk was generally associated with events further along the decision-making process. They suggested that data were often used as a protective mechanism for themselves, something that has become more common in recent years due to increases in litigation and expectations of parents. Maternity claims represent the largest total amount and second highest number of litigation claims in the NHS (NHS Litigation Authority 2012). In relation to congenital anomalies, there are a growing number of wrongful birth claims,

where parents argue that had an anomaly been identified, or the true extent of the disability understood, they would have terminated the pregnancy (NHS Litigation Authority 2012). It appears that when clinicians did not know what to say, they provided something objective as a protective mechanism. This was seen as being particularly pertinent when providing information on prognosis:

I think we have to be careful and we often use data to cover, I mean if you're not sure what to say, then you blind someone with numbers and a paper and hope that that will be fine. It's also providing something objective. I think that this is something that has happened more so. It's something that has changed over recent years [due to increased scrutiny]. (Clinician06)

The practice of defensive medicine as a response to scrutiny or litigation is not a new concept and perhaps unsurprisingly is well established in the literature stemming from the US (Elmore *et al.* 2005, Nash *et al.* 2010, Studdert *et al.* 2005, Weisman *et al.* 1989). However, little literature relating to practices adopted in order to moderate this is available. One such study from Australia highlighted the management of risk in terms of greater disclosure of uncertainty and communication of risk, as primary responses to mitigating issues of scrutiny (Nash *et al.* 2010). This fear is seen to arise not only out of the latter, but also when parents seek objectivity in order to make their decisions when only probability is involved. Interpreting data may then be perceived as being directive, something clinicians are required to avoid. This is discussed further in section 5.3.2, in relation to roles and responsibilities of the clinicians. In one sense this erodes the professional authority of the clinicians and leaves some parents feeling deserted, as suggested below:

Well at first they just gave us all these figures and percentages and stuff. But I mean, you know I'm not a mathematics genius. And all I wanted was them to tell us what was best for our baby. (Father12 – Terminated)

A variation in the way risk was understood and interpreted was common amongst parents, with many failing to understand the nature of probability. Some responded by dismissing or attempting to disprove the data provided. Others sought direct guidance from clinicians. Due to the constraints imposed by wider contextual factors, including non-directive counselling, the desired direction was often unforthcoming. An unresolved tension was created between the needs of parents and the boundaries imposed by the context in which the interactions were enacted.

5.2.4 *FRAMING ANOMALIES*

The way in which unknown anomalies were framed against those which were known traverses a number of the contextual layers. First it is influenced by individual parental experience or understanding of the anomalies, as well as that of their immediate social context, and it is also reinforced by the way in which screening for the anomalies is offered and enacted within the healthcare context. Finally it is influenced by wider social understandings and enactment of these by the clinicians within the legal and professional context.

What became apparent from analysis was the lack of knowledge and understanding of the FASP anomalies screened for during pregnancy. Down's Syndrome was used by the majority of the parents to benchmark information provided by healthcare professionals on the chromosomal anomalies. The few notable exceptions to this had previously encountered the anomaly concerned. Framing techniques can be used to reduce the obscurity of an unknown subject by contextualising the information in a way that enables people to associate it with what they already know (Goffman 1986). In this case, public perception of Down's Syndrome became the frame in which other anomalies were encountered. It also became apparent that work undertaken to increase public understanding of screening may have shifted the social framing of the screening process to incorporate Down's Syndrome. The early scan (nuchal translucency scan) and associated serum tests constitute the Down's Syndrome screening programme, although soft markers associated with chromosomal anomalies noted on the anomaly scan may trigger referral for invasive testing for chromosomal anomalies

(including Down's Syndrome). The social reframing of screening is perhaps more relevant to the early scan and serum testing, with the impact on the anomaly scan less noticeable.

The excerpts below highlight a number of issues: first, acceptance of the routinisation of screening; second, the lack of understanding that anomalies other than Down's Syndrome were being screened for; finally, the generally accepted views on Down's Syndrome and age related risks:

So yeah, I sort of went to the scan to check for Down's. I mean it was a routine scan and I normally have the Down's test¹¹ because the older you get the more the risk is anyway. It's just because you're past it. (Mother3 – Continued)

The next excerpt highlights similar sentiments regarding the risks associated with older mothers and Down's Syndrome. In addition, it highlights the lack of awareness of what was being screened for:

You know you read all this stuff about older women and the risk of Down's Syndrome but they just do not really mention all these other things. It's sort of weird because I went into the test worrying about Down's Syndrome and now I really wish it was just Down's Syndrome. (Mother6 – Terminated)

In addition, reassurance about Down's Syndrome was actively being sought through testing. However, the results did not provide the necessary assurances, as she became aware of the unanticipated diagnosis of a lethal chromosomal

¹¹ "The Down's test" referred to by this mother is a screening test that gives parents a risk based on the combination of a blood test, a scan measuring the nuchal translucency, and maternal age along with other risk factors.

anomaly. An assumed understanding of Down's Syndrome was widely stated, albeit a variable one:

..it sounds awful but something like Down's where you know what you're dealing with, but it was this element of not knowing, knowing it was going to be bad but just not knowing how bad. (Mother12 – Terminated)

There are a number of possible explanations for the lack of appreciation of the diversity of the screening offered. These include the rarity, high termination rates and the low survival rate of babies with chromosomal anomalies tested under FASP; for example Trisomy 13, 18 or Triploidy. Indeed, the likelihood that they would have encountered children with anything other than Down's Syndrome is low (based on the incidence of these anomalies presented in Section 1.2). In addition, the length of time that the FASP programme has been operational is significantly less than that of the Down's Syndrome screening programme, with much literature still referring to tests in terms of Down's Syndrome screening only (NHS Choices 2013). One respondent highlighted this when she remarked:

I don't really remember whether they said anything about those other problems... I knew about Down's and that's what I was sort of listening to. (Mother14 – Terminated)

The excerpt above suggests that the parents may have been given information about other anomalies. However, little of the information beyond her existing frame of reference was absorbed.

Generally there was a greater awareness of the FASP structural anomalies than chromosomal anomalies, with the marked exception of CDH, which none of the parents affected had heard of. In respect to the other FASP structural anomalies, this may relate to the tangible nature of a structural anomaly and conversely the difficulty in comprehending an overarching chromosomal anomaly. More recently, CDH has formed a major storyline in a national soap opera, *Holby City* (BBC 2015).

Public awareness may therefore have increased with regards to this particular anomaly. A number of the parents affected by a CDH diagnosis discussed the innocuousness of the term ‘hernia’ which is socially framed as a minor affliction, easily corrected by surgery, often not even requiring a general anaesthetic. The association of this term with a lethal condition was difficult to adjust to:

I know obviously that a lot of babies are born with heart defects and stuff.... But no I hadn't heard about the diaphragmatic hernia.... And a hernia just didn't sound like something to worry about (Mother2 – Terminated)

Despite the broader awareness of structural anomalies, there was frequently an expectation that these were things that happened to others, particularly those who failed to adhere to the responsible behaviours required of a mother:

Well I mean obviously I knew that babies could have problems with their hearts but it's sort of one of those things where you don't think it's going to be you. You know what I mean? I mean we did everything right, you know I don't smoke ... (Mother15 – Terminated)

Where parents had experienced a particular anomaly, either personally during a previous pregnancy or that of a close friend, the impact of the understanding gained was positive, in that it sped up the decision-making process:

Yes, it's really rare, but it happened to a friend of ours just before I was pregnant with our little one. So I think. So we already knew a fair bit about it, not exactly everything but we knew it wasn't really compatible with life. I think that made the decision a little easier, knowing something about it already. And when we got pregnant with our last one, I was really worried that the same thing would happen. (Mother19 –Terminated)

5.2.5 LOCATION OF CARE

This section continues on the theme of framing, exploring how not only personal or socially constructed knowledge frames understanding, but also location of a service. In this instance the provision of multiple services, namely antenatal care, screening and fetal medicine, in a single location potentially misrepresents the primary functions of the varying services. This has resulted in unresolved tensions between the practical and emotional aspect of care delivery. The joint location of care provision masked the realities of fetal medicine, and the potential for parents to arrive at the start of the decision-making process unprepared was heightened.

The influence of the context and setting on the consultation has been documented (Weinmann 1997). In relation to the setting studied, fetal medicine clinics were run consecutively with the other antenatal services. This was generic across all centres studied and appears to be standard practice nationally. This may in part be attributed to the use of the same specialised equipment for all services. Given the expectation of screening as an integral and normal part of antenatal care, when an anomaly was identified the parents felt out of place, that they no longer belonged and that they were different, and issues of location and place suddenly became significant. The provision of antenatal care, screening and fetal medicine in a joint location may have added to parents' confusion over the primary aims of services, with parents requiring fetal medicine services feeling scrutinised and labelled as deviant:

It was hard going into the antenatal clinic sometimes. I mean even now.... I don't want to feel like I'm different to anyone else you know. Um, but seeing people that are obviously pregnant... that was so difficult. (Mother2 – Terminated)

Being surrounded by large numbers of 'normal' pregnancies was particularly difficult, and increased the sense of isolation. Parents felt they were singled out and labelled as 'abnormal'. Many highlighted the tacit belief amongst parents in the clinic relating to the use of the counselling rooms. In the excerpt below, the mother

describes how parents shown into particular counselling rooms were immediately identifiable as having a problem:

It was hard going into the antenatal clinic at [hospital]. It just felt like everyone was looking at us. That was horrid. And those counselling rooms! I mean they may as well have put skull and crossbones on the door! And then when you come out having to face all those people who are staring at you and putting a smile on your face and pretending everything is ok. (Mother15 – Terminated)

Some parents extended this illustration to suggest a sense of deviance:

I mean they used the same consultation room for everything that went bad, everything that was bad. It was like the naughty cupboard. (Father08 – Continued)

Despite the initial distress caused by being in a shared location, the majority of the parents who continued subsequently expressed a desire to reintegrate with parents experiencing 'normal' pregnancies:

I'd rather be sat with everybody else because I think if you're going to be sat on your own then it's like..... I mean maybe not when we first got the news, but now... (Mother13– Continued)

Staff appeared sensitive to the difficulties of a combined location, and attempted to address this by restricting interaction between parents waiting for a follow up fetal medicine appointment and those attending antenatal clinics. However, this was frequently interpreted by parents as an attempt to single them out, and label them as deviant:

I don't know whether our names had a black mark, or something. But as soon as we arrived we were whisked

into one of the counselling rooms. (Father09 – Terminated)

For those that terminated the pregnancy, the desire to be away from other parents became stronger, particularly after the termination process had been initiated. This is explored in further in Chapter 7.

5.3 THE SOCIAL, LEGAL AND PROFESSIONAL CONTEXT

Due to their inter-dependence, the two outer layers of the contextual framework are explored concurrently in this section. Actions by clinicians are framed by the law and their professional knowledge and authority. However, this is not entirely straightforward. Despite consensus on a theoretical definition of severe in terms of anomaly, individual clinician variation was evident in relation to when a termination would be offered with regards to the timing and type of anomaly.

The wording of the termination laws allows for individual interpretation by clinicians. Although many complained about the lack of clarity or ‘wooliness’ of the law and the resulting pressures that are placed on individual clinicians within the system, the difficulties in creating a universally accepted definition of severe was widely acknowledged. In addition, the negative impact of a clearer definition on the options open to parents when faced with a decision to terminate or continue an affected pregnancy was raised.

Failure of society to engage with this discussion, yet scrutinise the actions of those involved in the decision-making process, has resulted in corporate interpretations of the law. This in turn may contribute to the creation of variations in access to termination, where some parents are forced to travel significant distances to hospitals to access care, along with the associated financial and emotional implications.

5.3.1 THE LAW

As highlighted in Table 5-1 below, terminations for congenital anomalies are performed under Section E of the Human Fertilisation and Embryology Act.

Table 5-1 Current sections of the termination law

Date	Law	Section	Interpretation
1990	The Human Fertilisation and Embryology Bill	Section C	Termination up to 24 weeks where the risk to the mother or her family of continuing with the pregnancy is greater than termination
1990	The Human Fertilisation and Embryology Bill	Section E	No limit where there is a substantial risk that the child would be born with severe abnormalities

Analysis of the data highlighted two issues pertaining to the limitations of the law. First, the difficulties encountered by clinicians in the interpretation of terminology used within the law, and second, the unintended impact of the 24 week limit for Section C terminations on application of the law for Section E terminations.

The terminology employed by the law is subjective and open to interpretation. Terms such as; “*extreme, substantial risk, grave permanent injury, seriously handicapped*”, (Department of Health 1990) are used to inform the practice of clinicians. As a starting point to this study, it was essential to gain some insight into how clinicians defined severe, and how this translated into the offer of termination in cases where a severe congenital anomaly was suspected. The difficulty in defining, and gaining consensus on definition, between fetal medicine clinicians was widely acknowledged:

You can't! ... so when we debated at the British Maternal and Fetal Medicine Society, whether [a chromosomal anomaly] met the criteria for Clause E, severe, the audience was completely divided ... So we cannot agree on that. (Clinician09)

This became particularly pronounced at 24 weeks, the point at which terminations under Section C are no longer permitted. Furthermore, the methods by which termination data are recorded also changes at this gestation and clinicians are responsible for submitting additional documentation to the Department of Health. This created a sense of unease:

There is a sense that the data are submitted into a black hole and it's unclear who gets to see this. (Clinician04)

After 24 weeks individual clinicians' thresholds for offering a termination also appeared to become more stringent. When discussing the possibility of offering a termination for ventriculomegaly¹², one clinician commented:

Before 24 weeks a termination could be offered for a ventriculomegaly of 14mm for instance, but after 24 weeks that wouldn't necessarily fulfil the definition [of severe](Clinician13)

The lack of clarity of the termination law and the difficulties this raised for clinicians, particularly after 24 weeks, was frequently highlighted:

... that critical time after 24 weeks after that it becomes much more difficult doesn't it? And I'm having to try and muddle through the law's pretty bloody woolly and it's completely unhelpful (Clinician08)

¹² Ventriculomegaly is a term used to describe the dilatation of the lateral ventricles in the brain.

The way in which the law is interpreted is open to challenge, with the issues created by a relatively recent legal case (the Jepson case) in particular still resounding in many clinicians' ears (Dyer 2005). The Jepson case revolved around an Anglican curate who sought a judicial review of the decision by police not to prosecute doctors who terminated a pregnancy for a bilateral cleft palate at 28 weeks. The challenge was based on the premise that a cleft palate was not a serious handicap and therefore did not fit under Section E. Although the case was finally dismissed in 2005, it has been suggested that ensuing scrutiny resulted in making an already distressing situation for expectant parents and their doctors even more difficult (BPAS 2013). Referring to the Jepson case, the avoidance of practice which might invite scrutiny was highlighted in the quote below:

There have been a number of well publicised cases of people performing um late terminations [for an anomaly]... was that right was that wrong? I don't know, but I do not want to fall into that. (Clinician10)

This cautiousness on the part of clinicians appeared to restrict the options they felt able to make available to parents, and in some cases resulted in time constraints being applied to the decision-making, in order that termination could be performed before 24 weeks. Many of the clinicians expressed anxiety over public scrutiny and judgement over their role in facilitating terminations:

I have an ongoing degree of anxiety ... that what we are doing is legal but there is a huge tranche of the population who would say ... "it's wrong what they are doing." And ... mostly [from] people who haven't had to make these choices (Clinician04)

Late termination (after 24 weeks) invoked particular fear of public scrutiny. As a result some clinicians suggested that they would encourage parents to make a decision before this point:

I would ... encourage people to make a choice ... before 24 weeks gestation because I don't think either myself professionally, personally or for them as individuals we particularly want anybody to be more carefully scrutinised as to why they've chosen a termination beyond. Nobody will look a second time if it is less than 24 weeks ... (Clinician10)

The strength of public opinion was acknowledged by all clinicians interviewed. They often expressed frustration over this, suggesting that the emotiveness of the issue had resulted in a failure of society to discuss openly and reach a consensus on where the boundary of acceptability should lie. Instead they felt that individual clinicians are subjected to intense scrutiny and responsibility is placed on them to enact the law:

...well I think again it's about society having a proper debate... sometimes I think society should take more responsibility because it's something that society really doesn't want to talk about so it puts quite a lot of pressure on the individuals providing the service because there is no guidance. (Clinician09)

One mechanism observed for overcoming these pressures and standardising practice within a centre was the use of a 'corporate' approach to interpreting the law. In practice this involved regular multi-disciplinary meetings, as well as formal peer support processes within the fetal medicine team. This served to remove the responsibility from an individual by placing it on a group, but also theoretically overcame some of the differences between clinicians' practice, as it allowed for discussion:

As you can see from this, I have my own spectrum, my own gradation and certainly in those kinds of cases where it's not obvious, then we sit together as a group of consultants and listen to one another's opinions on this.

That has altered the initial thoughts that one of us might have had about whether we would offer a late termination. (Clinician10)

Alongside this has been an increase in the number of ethics committees, who review and advise clinicians on an appropriate course of action. These committees consist of a number of clinical and ethical/legal advisors who discuss individual cases presented by clinicians. It remains the responsibility of the clinician whether or not to refer to the committee. Guidance is given by the group, but is not legally binding, although some countries insist all late termination cases are reviewed by this kind of group (Woodrow 2003). As is the case with research ethics committees, due to the subjective nature of the decision, consistency of opinions is not always guaranteed between committees (Angell *et al.* 2006). Although these mechanisms appeared to provide reassurance to individual clinicians, they were ineffective in reducing variation in practice between centres. Instead some centres were labelled '*more termination minded*' than others by the clinicians themselves, highlighting the differing interpretations of the law. Parents were referred to these centres for second opinions when a late termination was requested but clinicians were unwilling. Clinicians recognised that the same issues were not encountered before 24 weeks:

... before 24 weeks ... I could have organised that ... but after 24 weeks I felt that I could not organise a termination, even though they [the parents] felt quite strongly they wanted it ... it was my obligation to refer them on to somebody else [who would offer a termination]. (Clinician03)

These informal '*second opinion*' pathways appeared well embedded in practice. None of the centres participating within this study fell within the '*more termination minded*' group, although a number of clinicians had worked within such centres during their training. One of the risks associated with having a corporate perspective on termination is that this leads to potential variations in access to treatment. Although the option for a second opinion was available for all

women, the cost emotionally, financially and in terms of time may result in a barrier to access for some parents.

5.3.2 ROLES AND RESPONSIBILITIES

Within the interviews, a shared, 'corporate' understanding of professional roles and responsibilities was expressed by the clinicians. Their over-riding function was seen to be that of facilitator in assisting parents to make the right decision for them. In order to achieve this, a number of roles were perceived as being adopted in a chronological order. First was that of information bearer and interpreter; second, assessor of the decision-making process; and finally, supporter of the decision.

Clinicians emphasised the importance of their role as an information giver, stressing the need for the information to be imparted impartially, in a non-directive manner:

So the objective of antenatal counselling is to give them all the facts so they can make a decision what they want. But we never ever try to influence them one way or the other. (Clinician14)

A clear distinction was made by the clinicians between the decision-making process and the decision itself, where the 'right' decision could be achieved by engaging actively in the decision-making process:

There isn't a right or a wrong decision ... it's making sure you have all the information, and weigh it up so that you make the right decision for you, and your family at that time (Clinician11)

Their second role was expressed as an assessor of the decision-making process, where clinicians sought evidence that the parents had actively engaged and had been transparent in their deliberation over the best course of action for them.

[I have] a duty to explore how she [the mother] has reached her decision (Clinician16)

Finally, the importance of their role as supporter of the decision was universally endorsed by the clinicians interviewed. One clinician summarised this perspective as follows:

Because it is a huge decision to terminate your baby and if they've chosen to, then it's important that you make them feel as good as possible about their decision. And if they've chosen to carry on then it's also important to talk about the good side of that... (Clinician18)

As suggested above, the role of supporter of the decision was perceived as reinforcing the chosen option once a decision had been agreed upon. As will be discussed later in Chapter 6, reinforcing the chosen option could result in tensions between parents and clinicians.

5.3.3 RATIONAL DECISION MAKING

The roles and responsibilities of parents were equally well defined by clinicians. In order to reach the 'right' decision, a 'rational' decision-making process needed to be employed:

...it's important that they demonstrate a rational decision-making process [which will ultimately] result in the right decision (Clinician16)

Theoretically, rationality has been defined as; "a style of behaviour that is appropriate to the achievement of given goals, within the limits imposed by given conditions and constraints" (Simon 1976 pg.405). From the perspective of the clinicians, requirements of this 'rational' process involved parents actively engaging, weighing up relevant information, deliberating over the future impact of the decision, balancing risk in terms of likelihood as well as severity of predicted

disability and finally implementing the decision. Clinicians expressed concerns over parents who did not engage in this ideal decision-making process:

... but I would prefer if it were an active choice rather than a passive choice and I'm not always sure that it is.
(Clinician10)

Lack of engagement was interpreted as a passive choice by clinicians, resulting in them questioning the decision. This was particularly the case when parents continued with the affected pregnancy. This will be explored further in section 6.8. The ideal decision-making process sought by clinicians exhibited similar stages to that represented in the decision-making literature, namely; accessing information, deliberation and implementation (Charles *et al.* 1999).

Table 5-2 **Decision-making process; theory, ideal and clinicians' role**

Theory	Ideal, as defined by clinicians	Clinicians' roles
Accessing information	Active engagement	Information bearer and interpreter
Deliberating	Weighing up relevant information Deliberating over the future impact Balancing risk	Assessor of the decision-making process
Implementation	Implementing the decision	Supporter of the decision

In order to demonstrate the close correlation between the theoretical, ideal and clinician role, I have brought the three aspects together in Table 5-2. This illustrates an approach to care, which suggests clinicians' expectations are founded in evidence and can be seen as a rational approach to their role. Furthermore, drivers from the outer contextual layers such as professional guidance on

informed choice contribute to clinicians' need for a transparent decision-making process.

For clinicians a 'successful outcome' of the decision-making process was one where clinicians were satisfied that a rational decision-making process had been employed and clinicians and parents were in agreement on the final decision made:

If we reach an agreement that she is satisfied with the information, and I am satisfied with the decision she has made, there is a successful relationship and a successful outcome. (Clinician09)

As the extracts above describe, the decision-making process was perceived by clinicians as a two-way partnership, with clearly defined roles for clinicians and parents alike. Adherence to the perceived roles by both parties constituted a 'successful outcome'.

As a consequence, tensions could arise when parents were not perceived to fulfil their role in the decision-making process. Where parents requested a termination, and the process and/or decision was not perceived as rational by clinicians, barriers were put in place to ensure that the expected processes were undertaken by parents prior to action (in relation to a termination) being sanctioned by clinicians:

... 'cause ultimately I don't have to offer a termination. I have to offer them the opportunity of a second opinion elsewhere... but mostly if I say that I'm not 100% comfortable with this, at the very least I want you to listen to what someone else has to say. (Clinician07)

Conversely, for women who continued with their pregnancy, clinicians expressed concerns over either their willingness or ability to engage actively in the decision-making process:

... there is concern that some people... their default position is just to continue with the pregnancy because to actually go through that process of thinking where might I be, what might the consequences be of this action or that action, they're either not willing to do that, or they really just do not have the skills to do that. (Clinician10)

5.4 SUMMARY

This chapter has provided an examination of the wider contextual factors instrumental in creating an environment in which decisions are made and enacted. Clinicians encountered difficulties navigating the complexities and ‘wooliness’ of the law, which resulted in variations in individual clinician interpretation. Fear of scrutiny impacted on the clinician-parent relationship and the dynamics of the decision-making process shifted. Parents became responsible for the decision, with the clinicians theoretically detaching themselves, and the decision constructed as a matter of personal rather than professional concern (Latimer 2000).

Along with an increased awareness of Down’s Syndrome itself, attitudes and understanding of early Down’s Syndrome screening have progressed. The same change does not appear to be present with regards to the FASP anomalies. Although awareness of the structural anomalies was more widespread than the chromosomal anomalies, cause of the anomalies was often attributed to behaviour. This risked subsequent apportioning of blame.

In order to gain a greater insight into the decision-making process, it is necessary to explore the ways in which parents navigate the process and the mechanisms they employ to make a decision. A greater understanding will benefit not only future parents, but also clinicians. As one clinician said:

[It] will be great for us to have an insight into how they make their decisions. Not so that we can manipulate them, I don’t want to manipulate them, but we want to be as sure as possible that this woman is making the choice that will turn out to be the right choice for her. And at the moment I have no way of knowing.
(Clinician08)

6 FINDINGS 2 – DECISION-MAKING PROCESS

Having set out the wider context in the preceding chapter, this chapter provides an in-depth exploration of the decision-making process from the clinician and parent perspectives. Six themes pertaining to decision-making emerged from analysis of the substantial data set. Examination of the spectrum of responses within each theme enabled me to develop a conceptual model through the grouping of shared characteristics of the decision-making process exhibited by parents. Examination of each category within the model highlighted tensions created between the ideal and the reality in practice, alongside mechanisms employed by clinicians to overcome these. Evaluation of these mechanisms from the parents' perspective suggested mixed success, with clear divides between the categories.

The chapter covers the themes that emerged from the data, the evolution of the model description of the four typologies that were established, together with the shared attributes of each. Tensions between clinicians and parents which arose are also explored.

6.1 DECISION-MAKING THEMES

Six inter-linking themes emerged from analysis of the data relating to decision-making.

- *Information seeking*
- *Acceptance of diagnosis/prognosis*
- *Values and beliefs*
- *Future consequences*
- *Weighing up options*
- *Engagement with healthcare professionals*

A brief description of these themes follows. These were subsequently used to develop the conceptual model presented later in this chapter.

6.1.1 INFORMATION SEEKING

This theme reflects the different ways in which parents approached the need for information and how they collected it. Two main categories of information seeking activities were identified: activities such as internet searches or accessing organisations online (none of those recruited actively sought information through personal contact with organisations during the early decision-making period), and participating in monitoring or testing in order to obtain additional clinical information or provide confirmation of the potential diagnoses or prognoses. Parental approach and attitudes to the two categories generally differed.

Self-directed information gathering was independent of information provided by the clinicians. Information was sought as a means to gaining more insight, or as another perspective on what they had been told in clinic. A clear spectrum of responses from avoidance to active searching was noted in relation to self-directed information seeking behaviours. For some, the need for information was paramount. As one respondent stated:

...we went straight on the internet (laughs). Um I think at one point my step-mum was with us in the appointment and she was on the internet (Mother2 – Terminated)

In addition, validation of the information provided by clinicians was seen as essential, prior to enacting their decision:

We organised between them telling us what it was and going in and we went on the internet, and I mean we just wanted to know for ourselves that it wasn't going to, I mean that she wasn't going to live. (Father10 – Terminated)

Alternately, others responded to the identification of a potential anomaly by avoiding all information:

I don't want to read anything on the internet, I don't want to go on any websites I don't want to look at any pictures. (Mother1 – Continued)

As was acknowledged by many, the internet was a source of all information, and evidence to support any outcome was possible to locate:

I mean you can read what you want to hear from it [information on the internet]... you hear these miraculous stories you know. People carried on and the child was fine and this that and the other. And then you can read other stories and you can think, my God. (Mother6 – Terminated)

Although parents seeking information generally understood the importance of assessing the source of the information, those who engaged in ongoing information seeking behaviours appeared to find more difficulties in identifying reliable sites:

they had given me a list of it could possibly be this... you go home and you type in google and you click on the first link and the first link is never the proper one to look at, you need to go to google scholar or something like that to get the real ones, but you just do, don't you? You google you click on Wikipedia you get the worst possible story for that you click on the images and you get the worst. (Mother10–Terminated)

The variability of the information identified potentially created the need for ongoing searches to validate their understanding. This became a vicious circle.

Parents also sought additional information by participating in monitoring or further testing. This was generally clinician-led and involved the decision to undergo further invasive or non-invasive testing as recommended by the clinician.

Here, the spectrum varied from unquestioning acceptance to informed decision-making and subsequent acceptance or refusal of the recommended tests. Invasive testing procedures such as amniocentesis or chorionic villus sampling (CVS) carry a risk of miscarriage. Although none of the women refused further non-invasive testing (namely ultrasound scanning or Magnetic Resonance Imaging (MRI) scanning), some chose to delay invasive testing until the end of the pregnancy, thus eliminating this risk. However, these parents then had to manage a degree of uncertainty throughout the pregnancy.

This theme is concerned not with the decision to accept or reject the testing, but with the mechanism through which the decision was made. Within this sample a further spectrum of responses emerged. First, those who unquestioningly accepted the testing “*because the doctor said so*” through to those who made an ‘informed’ decision to accept or reject the testing, as the quotes below highlight:

So it just felt like for a 1% risk which is virtually zero it was worth it to find out. (Father2 – Terminated)

Conversely, faced with the same risks, other parents decided that the risks posed outweighed the potential benefit:

So you have the second test then your % goes up so it's not 1% its then 3 or 4 % and we decided even 1% was too much. (Father3 – Continued)

The decision to refuse invasive testing at this point suggests that the decision to exclude termination as an option had already been made, and confirmation of a severe anomaly would not influence the decision. Furthermore, the option to undergo invasive testing at a later gestation when the risk of miscarriage was no longer pertinent was accepted. This reinforces that rejection of invasive testing was not an avoidance of information, but an active decision to avoid the associated risk of miscarriage.

6.1.2 ACCEPTANCE OF DIAGNOSIS/PROGNOSIS

This theme encompasses the difficulties encountered by parents in coming to terms with the diagnosis and prognosis of the congenital anomaly. The elements underpinning these difficulties included parents' ability to cope with uncertainty, alongside the level of uncertainty created by the anomaly detected. In addition their degree of understanding influenced the ability of parents to reach a level of acceptance. The interwoven nature of these factors made them difficult to differentiate, hence they have been considered together under an umbrella theme of acceptance. This section will demonstrate examples of how parents managed the interaction between understanding and uncertainty and whether they reached acceptance of the given diagnosis and prognosis.

Unlike scans that can be interpreted by the operator at the time of the procedure, invasive testing such as amniocentesis, where a sample is taken and sent to the laboratory, requires time (up to three weeks) to culture and analyse. The impact of this waiting time is significant, therefore tests such as the FISH¹³ test have been utilised to provide quick (24 hour) results. For many parents, this provides a rapid end to uncertainty through a reliable test with a zero false positive rate. However, in the event that a mosaicism¹⁴ is identified, the FISH test may present a false negative result, resulting in further uncertainty for the parents. As a father explained:

The results from the very first tests came back normal, so we thought there wasn't a risk. So it wasn't until [a few weeks later] that we were told that the full culture had come back positive [to a mosaic chromosomal

¹³ FISH (fluorescence in-situ hybridisation) an add-on test that tests uncultured cells taken during the amniocentesis.

¹⁴ Mosaic is an incomplete chromosomal anomaly, where not all the cells are affected

anomaly], I mean what are you supposed to think?
(Father07–Continued–Consultation4)

In this instance the parents had received negative results from the FISH tests. The positive full culture results were then perceived as questionable. Lack of understanding or tolerance of the uncertainty resulting from these conflicting results appeared to prevent these parents fully accepting the diagnosis or prognosis.

The visibility of the anomaly also contributed to parental acceptance. In particular, for anomalies where absence of all or part of an essential organ was visible to the parents on the scan, acceptance of the diagnosis was easier:

We could see the big hole [in the head]; I mean it was obvious really. He was never going to survive
(Mother06– Terminated)

However, this was not generalisable to all structural anomalies. Where clinicians were required to interpret the tests (in the case below, an ultrasound scan), acceptance was variable:

I've no idea how they see anything on those things [scans]. I mean I guess we just have to trust them [the clinicians] (Mother02-Terminated)

In addition, some anomalies were associated with significant uncertainty in terms of prognosis or long term outcome, even where diagnosis was clear. This was something that the clinicians found particularly difficult to manage:

...it's an evolving speciality and the imaging is evolving as well, um it's not unusual to be in a position where we say we've seen this on your baby's brain scan, and they say what does that mean and we do not know!
(Clinician06)

For others, the impact of the diagnosis and prognosis never appeared to be fully absorbed or understood. The quote below was taken from an interview with a pregnant woman three days before she delivered. Her membranes had ruptured two days previously. Her baby had been diagnosed with a lethal condition that presented as an accumulation of fluid:

*But I had a scan yesterday and I'm pretty sure they said
that the fluid was all gone [from inside the baby] Yeah
I'm pretty sure that's what he said. Yeah, yeah.
(Mother04 – Continued)*

A miscommunication had occurred between clinician and mother, where the mother had understood that the fluid referred to that in the baby, not her membranes (as meant by the clinician). This misunderstanding further suggests that the mother had failed to grasp the poor prognosis associated with her baby's condition.

6.1.3 VALUES AND BELIEFS

This theme relates specifically to the beliefs and values expressed by participants and the impact these had on their decision-making. There were a variety of values and beliefs expressed amongst the participants. These pertained particularly to the issue of termination. Some expressed fundamental beliefs (not necessarily related to religion) that termination was wrong under any circumstances. Others articulated an understanding of the importance of the option to terminate:

*I could see how that would have been, not the easy way
out, cause I don't think that would have been easy, but I
could see how that would have been the 'best' thing for
some people, if you know what I mean. But that just
wasn't an option for us... There was no way I would do it
[termination]. (Mother01-Continued)*

Termination and disability are particularly emotive topics. The strength of opinion and influence of belief systems varied considerably between parents. For some, as

the excerpt above highlights, existing beliefs and values meant that termination was never an option. However, a few found themselves questioning fundamental assumptions they had grown up with. As one parent stated:

I've always been brought up believing termination is wrong, but with all the things wrong with her, I just couldn't put her through [being born]. I mean the suffering would have been terrible. (Mother06-Terminated)

Despite being brought up with strong beliefs, these parents found the reality of the situation overwhelming. Their subsequent decision to terminate the pregnancy resulted in the parents feeling isolated and cut off from potential support networks:

I haven't been back to church since... You get all these things drummed into your head then you cannot get them out... I've been too afraid to go back because I've committed a sin, which goes against the church's teaching (Mother19- Terminated)

Although religion was a strong factor in relation to predetermined beliefs and values, it was not a prerequisite. For others, attitudes towards termination played an essential role in their decision. Most parents who expressed fundamental beliefs in relation to their decision-making process continued with their pregnancy but as highlighted in the quote above, there were exceptions.

6.1.4 IMAGINED FUTURES

A further theme that emerged in relation to decision-making behaviours stemmed from the consideration parents gave to the future consequences of their decision. Some talked about making their decision based on the potential long and short term consequences of their actions, not only for the baby but also for themselves and family.

Short term issues around the potential suffering of the baby were frequently raised:

It was the thought that the baby would suffer and be in pain... (Mother02–Terminated)

Long-term, the impact on themselves and other family members was considered:

But we have got to think about the other children. I mean we managed to get our heads round all the practicalities of getting over here for the operations and how we were going to manage with all that sort of stuff it's just the rest you know the chance that I hand her over to someone and then never see her again and saying goodbye then would just be so hard for all of us you know when you've had that time to bond (Mother09 – Terminated – Consultation2)

Others did not feel that the future was theirs to decide, or found they were unable to separate themselves from the emotional aspect of the decision in order to look ahead:

I was, not in denial, but did not want to think of the end circumstances that came at the end of it. (Mother13 – Continued)

For others still, they were unable to look further than the immediate future:

I haven't really thought any further ahead, I'm just thinking of the first operation and getting that out of the way ... That's all I'm really thinking about. (Mother05 – Continued)

As these statements suggest, parents placed differing emphasis on the importance, and potential, of evaluating the future impact of their choice as part of their decision-making process.

6.1.5 *WEIGHING UP OPTIONS*

The 'weighing up options' theme encompasses the way in which a decision was made. Two extremes were noted. The first suggested an analytical, methodical process to decision-making, while the second appeared haphazard, disorganised and unsystematic. More colloquially, the differences could be compared to 'heart' or 'head' led decision-making styles (Cohen 1993). The 'heart' led approach was, as it suggests, often emotion led. Practicalities of the decision were overlooked in favour of feelings or 'gut reactions' about what was the right thing to do. Conversely the 'head' led approach supported a rational/practical approach to weighing one option over the other.

The excerpt below illustrates the 'head' led approach where a practical and balanced approach to the decision-making process is clearly demonstrated:

We were really, um, we did look at the practical, we looked at what [anomaly] was and the difficulties if he did survive, and the quality of life ..., we took all sorts of things into consideration really, but looked at it from a practical position. (Mother09 – Terminated)

At the other extreme, some parents found it difficult to overcome the emotiveness of the situation, and were 'heart' led. As highlighted in the excerpt below, the parents identified that they were already grieving for their lost dream of a 'perfect baby', and were unable to engage in the weighing up of options:

...we kept swinging from one side to the other... there wasn't any sort of real process to deciding what to do, weighing everything up was just so difficult because we were grieving (Mother15– Terminated)

6.1.6 ENGAGEMENT WITH HEALTHCARE PROFESSIONALS

The next theme reflected the different dynamics involved in the engagement between clinicians, midwives and parents, where a number of preconceptions and stereotypes were identified. These informed future interactions:

... you know some consultants have an air about them that makes you feel like they're on a level above you another plane almost. He wasn't like that. He explained everything thoroughly and made you feel at ease and all of those things, and he had a ... confidence without an arrogance so you kind of you trusted in him with what he was doing and saying (Mother19– Terminated)

Reflections on the experience of the parent-clinician relationship highlighted a spectrum of responses by parents. Some saw the clinicians as protectors and supporters:

... They [clinician] have been so supportive.... Just listening to us and giving us time whenever we needed to talk... (Mother06 –Terminated)

The interaction between clinician and mother expressed within this excerpt suggests one of mutual respect, in which the mother felt fully supported in her decision to terminate her affected pregnancy.

On the other extreme the interaction between clinician and parent was perceived as being unsupportive, even pressurising. This was more frequently the case when parents declined invasive testing, or they felt judged and had to stand up to clinicians:

You know every time we come in it's do you want this test, do you want this test, do you want this test. We've said no. We've said NO. But they keep on, and every time

they say it over and over again. And you think "Oh Christ" and that really does mess with your head.
(Father08– Continued)

Other parents felt completely isolated in the decision making process:

We just felt so deserted... I mean just so alone...
(Mother03–Continued)

Engagement with midwives appeared to offer a middle ground for a small group of these parents, with strong relationships developing between fetal medicine midwives and the mothers:

... I love going to see [midwife]. S/he just lets me rant like this, and then says "Do you want a scan? I'll get someone to give you a scan" And that is really lovely...
(Mother01–Continued)

Clinicians suggested that this could be related to the midwives' role and status, where midwives were viewed purely as a supporter of the parents, rather than as information givers:

And sometimes they open up to the midwives something they wouldn't necessarily tell a doctor. Why? I don't know but they can... feel like the information giving, which is a lot of what we do, has happened. And so this person is just there to listen to what their thoughts are rather than to be imparting information. (Clinician16)

They may also be perceived as more approachable than clinicians as they are potentially more similar to the parents in terms of age and socioeconomic status:

...and [the parents] think "Who is that old man? What does he know?" The midwives are often closer to their

age and I guess they can be seen as more approachable because they aren't doctors" (Clinician09)

Relationships between midwives and women who had decided to continue their pregnancy often appeared particularly strong. Midwives and these parents had the opportunity to create a rapport over a period of time, as they were cared for in the fetal medicine unit throughout their pregnancy.

6.1.7 SUMMARY

Many factors influenced parents' ability to accept the diagnosis and prognosis of a severe congenital anomaly. Conflicting test results, lack of easily identifiable structural abnormalities and a failure to grasp the implications of findings all contributed to some parents' inability to accept the diagnosis. The themes arising relating to decision-making are summarised in Table 6-1 below.

Table 6-1 Summary of decision-making themes

Information seeking	Approach and need for information
Acceptance	coming to terms with the diagnosis and prognosis
Values and beliefs	expressed belief systems including religious beliefs
Imagined futures	consideration given to for future consequences of their decision
Weighing up options	the way in which the decision was made (head or heart led)
Engagement with healthcare professionals	dynamics of the relationship between parents and healthcare professionals

6.2 CATEGORIES AND COMMONALITIES

A key finding in this study relates to the ways in which parents navigated the decision-making process. Four major categories of decision-making, which emerged from the themes identified, will now be discussed. The value of categorising lies in simplifying the complexities inherent in data, whilst recognising that in doing so one might lose some of the richness of that complexity. Nonetheless, through categorisation it was possible to highlight shared behaviours that ultimately could be amenable to intervention, or provide explanations or reassurance to clinicians when caring for parents.

This section provides a description of the evolution of a conceptual model alongside a description of the typology of the parents categorised within each behaviour group. By applying the model it was possible to illuminate tensions created between clinicians and parents during the decision-making process.

6.2.1 *EVOLUTION OF THE CONCEPTUAL MODEL*

The conceptual model evolved through an iterative process. First, as the recorded data were collected, they were transcribed, re-read and themes relating to decision-making were identified. Second, six themes were derived, as outlined in section 6.1. Third, a grid was then developed comprising a range from high to low/yes to no, represented horizontally, and with each individual theme listed vertically. Each case was allotted to this grid and patterns soon emerged, from which it was possible to group behaviours. Finally these were then translated into the model presented in Figure 6-1 overleaf.

Table 6-2 **Response spectrum within the themes¹⁵**

	High/Yes	Medium/Maybe	Low/No
Info. Seeking One			
Info. Seeking Two			
Acceptance			
Values/ Beliefs			
Imagined futures			
Weighing up			
Engagement			

Exploration of shared attributes within each behaviour group was then undertaken and a descriptive matrix was compiled comprising the behaviour groups on the vertical axis, and a set of variables (Table 6-3). The latter refers to the outcome of pregnancy, gestational age at diagnosis and outcome, the type of anomaly and socioeconomic status and maternal age upon which the sample was drawn. The procedure described suggests a clean, precise process. However the reality was far more ‘messy’. Ensuring clear definitions for each theme and demarcating boundaries proved invaluable.

¹⁵ Due to the relative rarity of the anomalies described, in order to maintain anonymity this grid has not been populated, but the data will be disclosed as a group typology later in this thesis.

Figure 6-1 **Conceptual model of decision-making typologies**

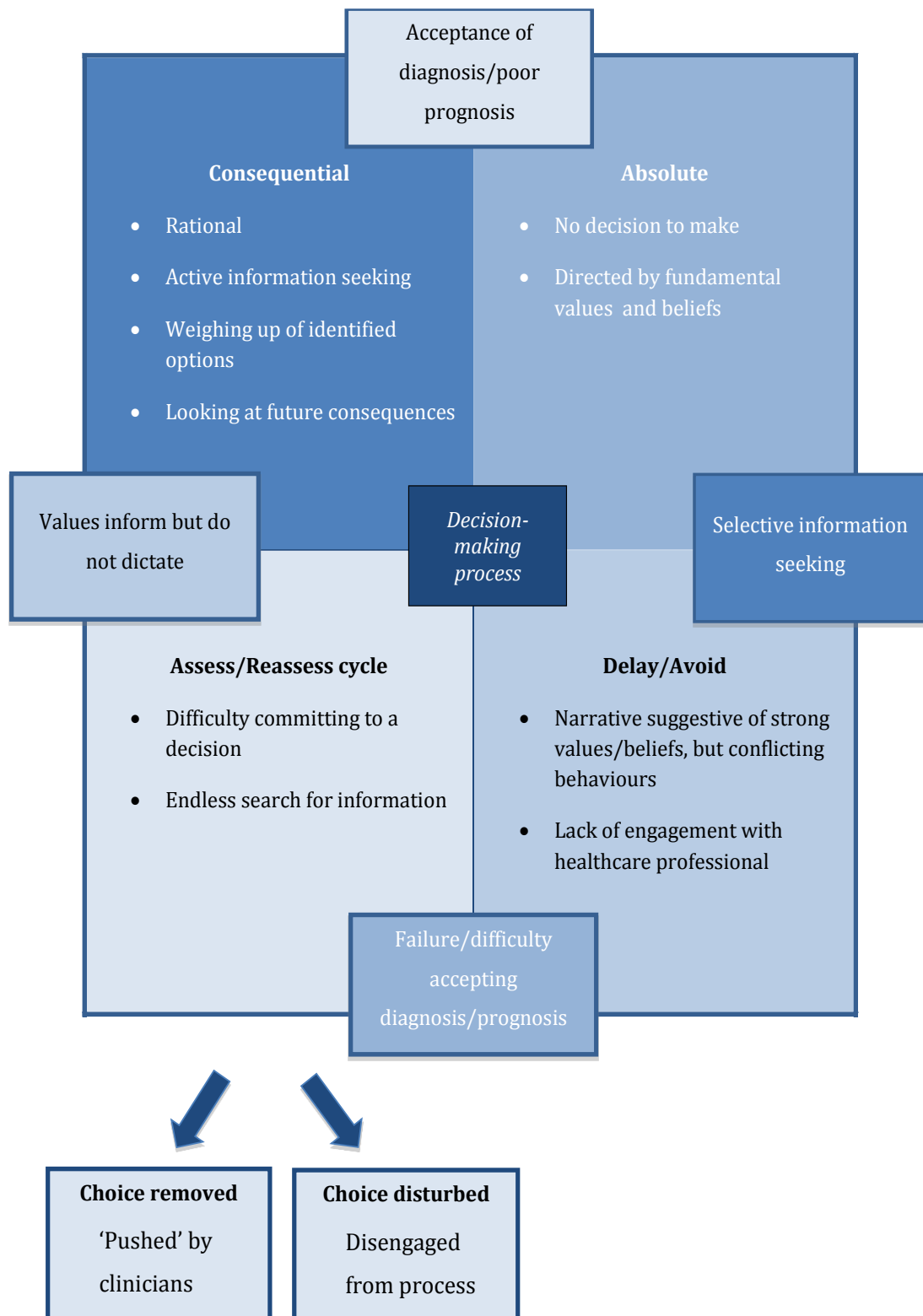


Table 6-3 **Matrix of decision-making groups and attributes**

	Number and Outcome	Gestation at suspicion	Gestation at diagnosis	Gestation at TOP	Type of anomaly	Amount of certainty	SES	Maternal Age
Consequential	5 TOP	9-18 weeks	13-19 weeks	13-20 weeks	Mainly Chromo	Certain	High	36-42
Absolute	4 Continued	9-20 weeks	9-20 weeks	N/A	Structural /Chromo	Variable	Low	26-38
Assess/Reassess Choice removed	5 TOP	9-20 weeks	13-20 weeks	20-25	Structural /Chromo	Variable	Low/ Medium	27-35
Assess/Reassess Choice disturbed	2 Continued	9-13 weeks	14-20 weeks	N/A	Structural /Chromo	Variable	Low/High	Late 20's to mid 30's
Delay/Avoid	4 Continued	11-21 weeks	16-21 weeks	N/A	Structural /Chromo	Variable	Low	19-29

6.3 CONSEQUENTIAL

The first category has been labelled Consequential or Analytical decision-making. The use of the title 'Rational' has been avoided, although the term reflects the characteristics of these parents' decision-making process. This stems from the frequent use of the term within the empiricist decision-making literature, where it refers to a correlation between the expected decision and the actual decision made, in other words the 'right' decision or outcome. The inappropriateness of employing terms that may be misinterpreted in this way is clear.

The cases allocated to this category shared the following demographics:

Table 6-4 **Shared attributes of the Consequential Group**

Sample size and outcome	Gestation at suspicion	Gestation at diagnosis	Gestation at TOP	Type of anomaly	Level of certainty	Socio-economic status	Maternal Age	Partner Present
5 TOP	9-18 weeks	13-19 weeks	13-20 weeks	Mainly Chromo	Certain	High	36-42	All

The parents were all from areas of high socioeconomic status. In addition, they had all completed a higher education qualification. The majority of the anomalies were suspected at an early gestation, and many related to chromosomal anomalies with little uncertainty attached to the diagnosis and prognosis. The maternal age range of this group was marginally older than the other groups and may reflect the higher proportion of chromosomal anomalies identified. All the women accepted the offer of invasive testing at an early gestation.

In this group, the timeframe between suspicion of the anomaly and diagnosis tended to be between three and four weeks, while the average time from diagnosis to termination was a few days. This indicates that the decision to terminate had frequently been engaged with before the diagnosis was confirmed. None of these women continued an affected pregnancy.

6.3.1 CONSEQUENTIAL- BEHAVIOUR CHARACTERISTICS

In the context of the themes identified, this group of parents actively sought information in relation to their baby's diagnosis as well as the prognosis:

... we did a lot of research on the internet about what it was. It was explained to us but sometimes you need time to ... take it in.... (Mother09 – Terminated)

Substantiation of information from multiple sources helped support their decision-making:

Well the information from [clinicians], it sort of corroborated. I guess that gave us the confidence to make our decision. (Mother20- Terminated)

Despite the confidence that added information gave parents, clinicians highlighted some concerns over the use of the internet to access information:

...if you put this into google you'll find the extremes. Like "My baby had this and they were absolutely fine" and then you'll get things like, you know, the completely opposite ends of the spectrum. So you won't be any wiser. (Clinician03)

The excerpt above suggests that clinicians had reservations over the ability of some parents to be discerning about information, or interpret what they found. In addition, identification of information that contradicted that offered by the clinicians would be likely to result in tensions. However, parents within the Consequential Group suggested greater ability to differentiate between data sources in their information seeking behaviours than the other groups:

... I mean, we sort of knew where to look. We've got some friends who are doctors as well and they gave us papers to read... (Mother06– Terminated)

Others in this group explained the search for information as a way of checking what the clinicians had said, to ensure all eventualities had been evaluated prior to making a decision:

We really needed to know for ourselves that it was there wasn't any hope, so we looked online (Mother20 – Terminated)

In addition to undertaking information seeking activities, all the women in this group underwent early invasive testing. This was seen as the responsible action, and essential for ending uncertainty and providing sufficient information on which to make an informed decision:

I just don't do uncertainty. I have to be able to plan for the future. I guess all my life is like that. I try not to leave things to chance. I need to know. The amino was something we ... sort of saw it as part of our antenatal care. It was ... our responsibility. We have a responsibility to our family and to our baby. (Mother06 – Terminated)

Within this group a sense of taking responsibility for their actions, along with accountability for the outcome, was expressed:

If you do that then you kind of blame, if you rely on the doctors to tell you it all then the doctors don't tell you something ... then in some ways you will blame the doctors for leading you down that path... (Father20 – Terminated)

Conversely, failure to inform one-self was perceived negatively and as avoiding taking responsibility:

If you are educated enough about something then you can make an informed decision... it's a lack of responsibility [not to do so]. (Mother09 – Terminated)

A further identifying characteristic of this group was the apparent levelheadedness applied to the decision-making process:

... we both just had our rational heads on. I guess that's just what we tend to do in those situations, you just think really rationally and the emotional sides just came through afterwards. (Mother20 – Terminated)

In addition, the consideration given to future consequences for the parents themselves, their family and their baby, was raised. Evaluation of the long-term consequences was enacted in a number of ways, and was generally dependent on the certainty or uncertainty related to the diagnosis and prognosis. In the first excerpt, the implications of placing the responsibility of long-term care onto existing or future children were contemplated:

That is something that we took into consideration as well ... it's the long term. I mean if in years to come if something was to happen to us and she was to live I mean they [the other children] would then be responsible for her and that's a big responsibility for anybody. (Mother10– Terminated)

The second excerpt highlights the evaluation of the timing of the loss of the baby:

It was either have a termination and lose her then before we got to hold her and know her, or wait and then have to deal with all the heartbreak of losing a child. (Mother06 – Terminated)

A number of the participants echoed these sentiments and suggested that termination was perhaps a personal grief, while loss of a baby was a shared grief.

Another couple demonstrated the dilemma involved in weighing up the consequences for the baby against the impact on themselves:

You have to make a decision. What was right for us, or what was right for him? I mean what was right for us was to keep it going. What was right for him was to stop it there and then. (Mother09 – Terminated)

There was a consensus in this group that acceptance of the diagnosis, and the subsequent prognosis, was essential to making a rational decision:

I think it's the acceptance part; you've got to accept that it's really there. You've got to accept that your child has got a problem and is not going to be a healthy baby and you've got to accept, the minute you accept that and you don't think to yourself that it will all be fine and he or she will be fine. (Mother20 – Terminated)

6.3.2 TENSIONS

Conflict and tensions that arise during consultation are frequently a result of conflicting needs and expectations of the two parties (Stavropoulou, Glycopantis 2008). As highlighted in section 5.3, the expectation of clinicians was that the parents would actively participate in a 'rational' decision-making process, characterised by their engagement in weighing up of relevant information, deliberating over the future impact of the decision and balancing risk in terms of likelihood as well as severity of the anomaly identified. Where these expectations matched the actions of parents, a 'good outcome' was perceived by all involved. For the majority of parents in this group, the decision-making process appeared to be uncomplicated, with a number of parents highlighting that clinicians had commented not only on this, but also on their preparation:

We recently had our post counselling consultation with [clinician] and s/he did say how impressed s/he was

with how well informed we had both been... (Mother20 – Terminated)

However, for one couple in this group tensions did arise in relation to information gathering over what constituted sufficient information in order to make an informed choice (where clinicians felt the parents were under informed). Where this was the case, clinicians enacted their role as gatekeepers to treatment, in this instance termination:

... we always include the offer of counselling from a specialist paediatrician I would be very unhappy to offer a late termination until those individuals had spoken [to the neonatologist or paediatrician] (Clinician01)

As the excerpt above highlights, clinicians maintained authority over the parents either by refusing to 'sign off' a termination, or referring the parents for additional testing or counselling before reconsidering. Within the group of participants recruited, this authority was enacted on only one occasion. In that instance the couple was invited to attend a multi-disciplinary meeting and offered additional invasive testing. Both of these were accepted, although the decision to terminate remained unchanged. The gestation at the time of termination was 18 weeks. Whether the clinicians would have complied with the request had the gestation been greater than 24 weeks is purely conjecture. Furthermore, as the pregnancy was under 24 weeks' gestation, the parents could have requested a termination under Section C without meeting the clinician's demands. However, the parents were emphatic that they would not proceed under Section C. This perhaps suggests an attempt to demarcate themselves as different to other parents undergoing termination under Section C. This will be explored further in section 7.4, when examining the impact of stigma and trying to make sense of the decision.

6.4 ABSOLUTE

'Absolute' stems from the Latin 'absolutus' meaning unattached. It portrays the concept of an unconditional reality. In relation to decision-making this can be interpreted as a course of action that is true in all situations regardless of circumstance or context. This group has been entitled 'Absolute' as a result of their shared perspective that they could not proceed with a termination under any circumstances.

6.4.1.1 ABSOLUTE – BEHAVIOUR CHARACTERISTICS

This group of women believed there was no decision to make. Fundamental value or belief systems directed them along a particular course. Two of the four based this on religion:

*Allah, he performs miracles. We will wait and see.
(Father17 – Continued)*

*... you know it is big and it is scary and it is daunting and
it is a lot to take on. But, God knows how big my
shoulders are, and he wouldn't put anything too big on
them. (Mother01 – Continued)*

However, for the other two, their strong belief systems were based on a personal moral code rather than decreed through religious beliefs. Where these were related to termination, the possibility of interrupting the pregnancy was excluded:

*I was adamant from the beginning that there was no
option for it [termination]. And if that were the case I
would still go full term, still have the baby. (Mother08 –
Continued)*

6.4.1.2 SHARED ATTRIBUTES

The demographics of this group are summarised in table 6.5 below.

Table 6-5 Shared attributes of the Absolute Group

Sample size and outcome	Gestation at suspicion	Gestation at diagnosis	Gestation at TOP	Type of anomaly	Amount of certainty	Socio-economic status	Maternal Age	Partner Present
4 Cont.	9-20 weeks	9-20 weeks	13 weeks	Structure and Chromo	Variable	Low	26-38	3

All the women in this group continued with the affected pregnancy. Three out of the four were supported by their partner, with one couple separating due to disagreement on how to proceed. They were all from low socioeconomic groups, with three couples coming from minority ethnic groups including Afro-Caribbean and Pakistani origin. Not all the parents expressed strong religious beliefs. Despite the differences in cultural and religious beliefs, this group held a shared perspective, which was expressed in their adamant belief that there was no decision to make.

With regards to the anomaly itself, there was no pattern in relation to the type of anomaly or the gestation at which it was identified. This group were, however, universally risk averse in relation to the uptake of invasive testing for chromosomal disorders. Although all the couples went ahead, they delayed until after 32 weeks' gestation when the risk of miscarriage no longer applied, and were able to obtain a conclusive diagnosis prior to the birth of the baby.

6.4.1.3 TENSIONS

The parents in the Absolute Group did not actively seek information on which to base their decision as the decision had, in essence, already been made. However, they did make an active decision to either delay or forego further invasive testing, with the majority of the parents in this group opting for late amniocentesis (after 34 weeks). This resulted in tensions between parents and clinicians where clinicians feared that parents were *insufficiently* informed, either due to parents'

initial refusal and subsequent delay in undergoing invasive testing, or over their perceived lack of understanding of the diagnosis and subsequent prognosis.

Clinicians placed the acquisition of information and the subsequent knowledge that this engendered in high regard as it provided the cornerstone to a rational decision-making process. The contradiction arising from “*knowledge is always good in itself*” and the “*right to remain in ignorance*” is hotly debated in the genetic ethics literature in relation to autonomy and informed choice (Harris, Keywood 2001 pg. 436). Where parents rejected accessible information in the form of invasive testing, clinicians deemed this illogical:

I have concerns that they really don't understand what they are going to let themselves in for. I mean if it's [information] available, why wouldn't you want to know? (Clinician17)

Although tensions pertaining to the seeking of information were highlighted in the Absolute and Consequential Groups, the tensions that arose and the mechanisms through which they were addressed differed, predominantly as a result of the decision to continue or terminate the affected pregnancy. The decision to continue the pregnancy, by the Absolute Group, altered the dynamics between the two parties. In essence, continuing the pregnancy was the default position and required no action to change the outcome. Therefore the legal-professional responsibility of the clinicians to the Absolute Group was reduced to a professional responsibility as the legal responsibilities, in terms of enacting the termination law, were not pertinent in these cases, nor was the ensuing fear of scrutiny. This resulted in a power shift in the parent-clinician relationship, where the clinician had no leverage (the ‘allowing’ of a termination) to control the volume of information that the parents should access. Despite the lack of sanctions available to clinicians, an attempt to regain control was evident where parents stated that they continued to feel pressurised into accepting invasive testing, as ‘responsible parents’, in order to provide additional information to support the diagnosis and predicted prognosis:

And then every time we come back and you get this, “Oh well you should have the amniocentesis” It’s like he’s trying to force you into going for it. (Mother08 – Continued)

This sentiment was reflected in a number of the consultations, as the excerpt below demonstrates:

Clinician02 *Just one question quickly for you now [mother] did [clinician] talk to you about the amniocentesis?*

(Mother08) *The test?*

Clinician02 *Yes the test about the chromosomes*

(Father08) *Yes about 4 times or 5 times. Every time we go in*

Clinician02 *I understand it is something that you did not really want? (Consultation 5)*

The clinician proceeded to inform the parents of the benefits of undergoing the test. These were framed around the benefits for the baby after birth:

...it is always worth considering having a test, so that we can treat the baby with dignity and respect after baby is born, and we can make the best decisions (Clinician02)

The quote above exemplifies the unique difficulties encountered by clinicians in fetal medicine or obstetrics generally. Until birth, the baby remains the responsibility of the mother. However, after birth, the baby becomes an individual. The differing perspectives on the need for information between the parents and the clinicians created a tension where clinicians believed that the needs of the baby after birth could not be met without a full understanding of the diagnosis. This is at odds with the parents’ needs to protect the child from the risks associated with gaining that information. Resisting the pressures applied and

rejecting the proposal of invasive testing is perhaps an indication of the consideration given by parents to the decision to delay or decline testing.

In relation to more general information relating to the anomaly, the parents in this group were selective in their information seeking behaviours where the provision of further information had the potential to cause further distress, with little gain. This was something that was widely acknowledged by clinicians:

No matter what, a lot of them will stick with it [the decision to continue]; because they've made up their minds and in their heads that's fine, they can cope with it. And if you say to them, "There is more information to come then". And they are saying; "Okay well what next?" there is a lot of worry and the ideas become confused.
(Clinician08)

However, prior to arriving at this point, clinicians wanted reassurance that an informed decision had been made. Perhaps as a reflection of this, a common feature of parents in this group was the sense that their decision was not being heard:

... and yeah I just felt like [any of the clinicians] I spoke to at some point put termination, the termination option in there. And I know it had to be done and I know they have to give you all their options and it's all part of their job and I fully understand that but it's not something you want to be hearing when you're having a baby; you know what I mean? (Mother01 – Continued)

Midwives played a particularly important role with this group. Amongst the midwives, a sense of admiration for those who continued affected pregnancies was expressed:

it makes it easier when they totally come to terms with things that you've said to them, totally accepting, some people say, God's given me, well whatever God, and that's nice and I like that because it is so different to the society where everything has got to be perfect ... and they're happy with that, and that's what I like about that because they're happy whereas other people aren't happy (Midwife3)

Midwives and mothers within this group had the opportunity to create a rapport over a period of time, as the women were cared for in the fetal medicine unit throughout their pregnancy. Acknowledgment, by parents, of the sense of acceptance by the midwives was reflected in a number of extracts:

... to be absolutely fair I love it, I love going to see [midwife]. Um not under the best circumstances obviously ... And I know when I go to the hospital and I no longer belong to [midwife] my life still needs to carry on. Um somehow we have to deal with it, but I just do not know how. (Mother08 – Continued)

A large proportion of the midwives working within the fetal medicine units studied actively practised a religion. Whether the nature of the job attracts particular attributes or whether this was chance is unclear. However there is some evidence to suggest that either personal attributes determine the choice of specialism, or that particular specialisms attract certain personalities (Kluger *et al.* 1999, Hojat, Zuckerman 2008).

I've grown to really like them. Particularly [midwife] and I don't know if that is because we both have that element of Christianity in us. ... but kind of I've got God and s/he was kind of; "I've got God too." And so it was like ok, we've got something together. And so it feels like she knows a little more about where I'm coming from

because I've said "God has put this on me" and she's just accepted that. Not like "you weirdo, what sort of excuse!" (Mother01 – Continued)

The excerpt above highlights not only the acceptance shown by the midwife caring for her, but the influence of religious beliefs in the support that was perceived by the mother.

6.5 ASSESS/REASSESS

This group was the largest and most complex of the four categories. Although initial behaviour characteristics exhibited were consistent, at some point in the decision-making process the group was split into two. These sub groups have been entitled Choice Removed and Choice Disturbed. The shared attributes of the two groups will be presented separately. However, the initial behaviour characteristics are discussed jointly in section 6.5.1 below.

6.5.1 ASSESS/REASSESS BEHAVIOUR CHARACTERISTICS

The decision-making process of this group of parents was cyclical; information was highlighted, a tentative decision made, further information given and their position re-evaluated (the assess/reassess cycle). For this group, there appeared to be an endless search for information, associated with difficulty committing to a decision. The changing landscape and uncertainty associated with diagnosis and prognosis impacted significantly on this cycle in the example below:

... when we went to one of the scans we had a load of questions ready, didn't we, [about the anomaly]. And then this woman she just told us all this other stuff. Then she turned around and said have you got any questions? Well I was like this is something new now; we don't even know what it means, so we had to start all over again ... (Mother02– Terminated)

A number of the clinicians also highlighted this circular process:

*I had to terminate the consultation. It was 2.5 hours ...
Every question generated a question and every answer
generated another question. And we were going in this
big circle... (Clinician14)*

Some of this related to the uncertainty of the prognosis and the need to wait for the baby to grow in order to assess the severity of any structural anomalies that had been identified. This waiting and uncertainty often proved particularly distressing:

*But then we had to keep waiting for the baby to grow
some more. 'Cause the chromosome tests came back all
clear... then we had to wait till 16 weeks. And then that's
when they picked up the heart defect. And then they
wanted to compare everything with an MRI scan; so we
had to wait to 20 weeks. The situation was always, you
know, we suspect this, we suspect that ... dealing with
the uncertainty, that was the worst bit really. (Mother02
– Terminated)*

Irrespective of the subsequent decision to terminate or continue the pregnancy, many of the women talked about putting their pregnancy 'on hold' and suspending decision-making during this time, as a protective mechanism for themselves and their families. The excerpt below highlights this common need to distance themselves from the pregnancy:

*You know people don't even know I'm pregnant. Look
my dad, my real dad who only lives in ..., doesn't even
know I'm pregnant ... it is being kept so small ...
(Mother03– Continued)*

Another problem expressed by parents in this group was the difficulty in accepting the diagnosis and prognosis:

I mean sometimes it was quite difficult to talk about it because sometimes I was.... not in denial but did not want to think of the end circumstances that came at the end of it. I was trying to hide away from it, if I was honest (Mother15– Terminated)

Although an ongoing assess/reassess cycle is potentially an inevitable process in response to uncertainty, issues arose when some of the parents found difficulties moving on in the cycle from tentative decisions to final decision and subsequent action. At some juncture during the decision-making process, the group divided. In an attempt to explore the decision-making process of these parents, two response typologies have been identified, based on subsequent actions and interactions with clinicians.

6.6 CHOICE REMOVED

This group constituted five couples, all of whom eventually terminated the affected pregnancy.

6.6.1 SHARED ATTRIBUTES

As demonstrated in Table 6-6, this group of parents were heterogeneous in relation to type of anomaly and gestation when the anomaly was suspected or when diagnosis was made.

Table 6-6 Shared attributes of the Choice Removed Group

Sample size and outcome	Gestation at suspicion	Gestation at diagnosis	Gestation at TOP	Type of anomaly	Amount of certainty	Socio-economic status	Maternal Age	Partner Present
5 TOP	9-20 weeks	13-20 weeks	20-25	Structure and Chromo	Variable	Low/Medium	27-35	All

All the couples came from middle range socioeconomic backgrounds, which were reflected in the midline maternal ages (27-35). The time between gestation at suspicion and termination represented a larger range than the previous groups. This reflected the extended time taken to make a decision.

6.6.2 CHOICE REMOVED – BEHAVIOUR CHARACTERISTICS

The interaction between parents who fell into this group and the clinicians involved differed from consultations with other groups, both in the way parents approached the clinicians and the way in which clinicians responded. The parents on the 'choice removed' route appeared more ready to seek the opinions of healthcare professionals and spent more time deliberating about their decisions than those in any of the other groups. In turn, the clinicians responded by making the counselling more 'directive'. In relation to the continuum described by Williams, the boundary between choice and coercion was shifted significantly (Williams *et al.* 2002). The parents were clearly undecided on the best course of action and furthermore found difficulties in moving from decision to action. The following extract was taken from a counselling session with a couple and two clinicians. (Consultation3)

Clinician31: *So this is usually the scenario for a [specific anomaly] patient and we have one dying horribly on ward X at the moment... So to start with this is what will happen if you carry on with the pregnancy, otherwise termination is always an option, I'm giving you all the facts here, I'm not swaying you towards any way.*

Mother02: *Crying*

Clinician31: *Yes you are sitting on a bomb. And then of course they are that much older and you get attached to the baby and their loss is that much more painful. But they might not reach any of those points because with*

the pulmonary hypertension, there is not one surgeon who is going to want to take them or touch them.

The clinician repeatedly expressed concerns for the suffering of the baby; in addition suggesting that the option for surgery originally offered would be removed, in essence closing the option for intervention. Finally s/he expressed concerns for the parents, by stating that their grief would be greater if they continued. At this point the second clinician re-directed the consultation and re-established the boundaries and responsibility for the decision, while reiterating the need for a 'rational' informed decision.

Clinician27: *We've talked a lot. It's a lot to take in. You've probably been explained enough. And the other thing is we're not expecting any decisions from you. All we're trying to say to you is how things are. But at the end of the day you just need to digest it and get it in your head. Then you need to make a decision about what you want to do. And whatever you want to do we are here to support you. And it doesn't have to be a rushed decision.*

The consultation continued with the risk of suffering to the baby being repeated by the first clinician.

Clinician31: *But you need to be thinking of the suffering of this baby as well ... of course it is a challenge for you and you have the support from your family, but it is the suffering of this baby.*

The affected pregnancy was eventually terminated at 24 weeks' gestation. Five weeks after the termination, the parents stated their gratitude towards the "straight talking" clinician, saying this was what helped them make their decision. In this instance directiveness was perceived as "good care" by the parents (Schwennesen, Koch 2012 pg.282).

I think in the end of it all was kind of, he was going to be that poorly that they probably wouldn't be able to operate on him, which would cause him to die anyway and um I think that was what helped us make our decision. (Father02 – Terminated)

These parents had been deliberating for a number of weeks and did not appear to be able to move on to a decision. In contrast, clinicians involved in the care of parents in this group became increasingly insistent in their communication of the risks of continuing the pregnancy that appeared at odds with their statements of neutrality.

Despite the 'push' towards making a decision, difficulties in committing to the subsequent course of action were expressed:

...we mentioned that we might be having the termination, and then we were booked back in 2 days later, on the Thursday. Like booked back in and we can do it (Laughs) Then it was like (deep breathe in) ...we just ran a mile. (Mother12 – Terminated)

... Yeah like that just freaked us out. So although we felt like we'd made the decision, when someone said "Ok come back in 2 days and we'll do the termination" it was like, right, ok, hold on... (Father12 – Terminated)

6.6.3 TENSIONS

In general, the parents in this group felt well supported throughout the decision-making process. The major tension between parents and clinicians stemmed from the ongoing process of assessment and reassessment of the situation by the parents that increased the likelihood that the decision was tentative and would change.

Second, clinicians highlighted the potential for tensions between themselves and some parents, where parents were perceived as being overly engaged. Although this was not apparent in any interactions between clinicians and parents in this study, this was regarded as a risk by clinicians. A tension became apparent, where clinicians perceived information seeking as a positive and essential attribute, yet highlighted concerns when parents sought information outside of the clinicians' control. Talking of the negative aspects of parental information gathering, one clinician highlighted:

[with the internet] you won't be any wiser. So it's best if you stick to the tests ... have the MRI scan and let the consultants that are the experts on these conditions talk to you about it, or the paediatricians about the children they have seen, rather than put it into google because they come out with all sorts. (Clinician03)

Another clinician expanded on this, acknowledging the needs and rights of parents to gather information whilst raising the potential dangers:

I'm not saying keep people in the dark because people will pursue it and quite rightly pursue information. I would. Um but then, they you know they can get way, way, way out of where they need to be. (Clinician07)

Novel procedures and techniques were occasionally identified by parents whilst seeking information. For some clinicians this caused ethical difficulties when making judgements over what was physically feasible and what was appropriate:

... what patients now expect, they read stories showing that we can do lots of things and we can, we keep babies alive ... but one of the difficulties is now to know when to and which babies ... yes you can keep them alive, but is that appropriate? (Clinician08)

Attempts to control access to information were highlighted by clinicians, including reporting of findings in broad terms without committing to a specific diagnosis that could easily be googled:

[clinicians] have got very much better at using words that are harder to google. It makes a difference.
(Clinician07)

Tensions surrounding accessing information existed on a number of levels. Although clinicians universally discussed the requirement of informed choice, tensions were apparent where parents were required to be informed, but not too informed. Parents were encouraged to take ownership of the decision through accessing information on which to base this. This enabled clinicians to distance themselves from the decision itself, and protect them from accusations of eugenics. On the other hand, clinicians wanted to retain ownership of the information with the power to determine what, and how much, to provide to the parents.

If clinicians misjudged when a decision was final, the support offered to the parents backfired and instead became a source of distress. Clinicians talked of the concept of 'leaning' towards an option in support of the parents:

But without actually leaning any way until they've leant and then try and facilitate sort of their enjoyment of that decision. You know, it's difficult (Clinician18)

'Leaning' in this way appeared responsive to the calls by some parents for clinicians to support them, by reflecting the positive side of the decision made. However, misreading the decision could result in added distress. An example of this is reflected by one woman talking about her experience following a counselling session with one of the specialist clinicians:

And that [clinician] said that [the baby] wouldn't have known no different because he wouldn't have ever been no different. [Baby's] quality of life, it would have been

*just [his/her] life because [he/she] wouldn't have known
no different. And that made me feel bad then. (Mother12
– Terminated)*

In this scenario, a well-intentioned attempt by a clinician to support an initial decision to continue created an additional level of grief and guilt when the parents subsequently decided to terminate the affected pregnancy.

Tensions did not arise solely between clinician and parent. When caring for parents in the Assess/Reassess Group, clinicians demonstrated a response to their needs by giving the 'push' that was required for a decision to be made. However, by acting in the perceived best interests of the parents, clinicians experienced a tension between this and the idealised requirements for non-directive counselling. One clinician encapsulated this tension stating:

Sometimes, they [the parents] are asking you, "Is that right, is that wrong?" They are wanting you to be involved in their decision-making... it's hard to see them like that and I want to call out and say "Well if it was me this is what I would prefer" (Clinician08)

This supports what has been found within the literature, where the "taken for grantedness" (Williams *et al.* 2002 pg.341) and subsequent difficulties in achieving non-directive counselling and informed choice have been well documented in relation to antenatal screening and testing (Clarke 1997, Williams *et al.* 2002, Schwennesen, Koch 2012).

As previously discussed in Chapter 5, the principles of non-directive counselling and informed choice have been framed as mechanisms to distance clinicians from the discourse on eugenics, as well as to protect them from the emotional responsibility of the decision (Clarke 1997). Nonetheless the failure of theoretical ideals to submit to "the messiness of mundane practices" (Mol 2008 pg.43), is a finding aligned with a growing body of literature (Anderson 1999, Petersen 1999, Pilnick 2008, Schwennesen, Koch 2012, Rapp 2000, Williams *et al.* 2002).

Schwennesen goes as far as to suggest that the concept of non-directiveness is “*absurd*” (Schwennesen, Koch 2012, pg.285), as the creation of knowledge is an intervention in itself, and as non-directiveness by definition is the lack of intervention, the two cannot co-exist. Others suggest that by clinicians distancing themselves through the rhetoric of non-directiveness or informed choice, patients are left feeling deserted (Bosk 1992, Corrigan 2003). One father’s reaction exemplified this argument when he stated:

And all we get everywhere else is “We respect your decision” And then you think “Oh my God is it the wrong one?” (Father08– Continued)

As became clear in these scenarios; “*situations of indecision emphasize the need for a collaborative, or even at times a delegated decision-making whereby the professional assumes a greater role*” (Madrigal et al. 2012, pg.2880).

6.7 CHOICE DISTURBED

This was the smallest of the groups. Originally classified within the Assess/Reassess Group, a series of events resulted in the group dividing after some parents became disengaged from the process.

6.7.1 SHARED ATTRIBUTES

The two couples in this group shared commonalities in behaviour patterns relating to decision-making with the Choice Removed Group. However, the groups diverged at some point, resulting in the outcome of the pregnancies differing and the Choice Disturbed Group couples both continuing with the affected pregnancy. Both pregnancies were affected by a severe structural anomaly. In addition, one couple was given a subsequent diagnosis of a mosaic chromosomal anomaly. Unlike any of the other groups identified, these parents came from sharply contrasting socioeconomic backgrounds.

Table 6-7 Shared attributes of the Choice Disturbed Group

Sample size and outcome	Gestation at suspicion	Gestation at diagnosis	Gestation at TOP	Type of anomaly	Amount of certainty	Socio-economic status	Maternal Age	Partner present
2 Cont.	9-13 weeks	14-20 weeks	N/A	Structure and Chromo	Variable	Low/ High	Late 20's to mid-30's	Both

6.7.2 CHOICE DISTURBED – BEHAVIOUR CHARACTERISTICS

Parents in this group often spoke of the need to “*prove clinicians wrong*” or picked up on issues that they were subsequently unable to let go. The changing landscape of diagnosis and prognosis resulted in trust between clinicians and parents disintegrating and finally disengagement from the process.

Initial behaviours within this group included the seeking of information from other sources, in particular seeking a second opinion and searching for testing external to the point of contact in the hospital. There was frequent fixation on technicalities, often which seemed irrelevant to the diagnosis or prognosis. In some cases these were used to ‘point score’ against clinicians. Finally parents disengaged, with no active decision to either continue or terminate made. By ‘default’ these women continued their pregnancies. Extracts from a consultation demonstrate the frustrations felt by parents after seeking a second opinion:

*... it [second opinion] turned out basically pointless... it wasn't the fact ... for them to confirm that we've got a [diagnosis] because we've read it, we've seen it and we know all about that ... The idea ... was to run some more tests ... but obviously we ended up that nothing was done because they've gone with your guys diagnosis; so in essence it was kind of a pointless exercise...
(Father07–Continued–Consultation4)*

In this instance, the parents had highlighted an issue pertaining to the invasive test and had become fixated on this. The father subsequently expressed his frustration

that the clinicians to whom they had gone for a second opinion had not repeated the test and had instead 'colluded' with the other clinicians.

Events relating to issues of managing risk and uncertainty were prominent in the data derived from these parents. Both sets of parents discussed the impact of the imperfect science of antenatal diagnosis. The resulting changing landscape created issues of trust, where parents felt that the option to terminate became questionable as they perceived the projected diagnosis and prognosis could not be guaranteed. Tensions developed as the parents became more aware of the difficulties of interpreting tests, and of the range of alternative approaches to problems. This is a well-rehearsed story within the risk management literature (Taylor-Gooby, Zinn 2006).

In the excerpt below, the parent explained how the results of initial tests had been reported as negative before an unequivocal result finally being given. He also reiterates his understanding of the risk of a chromosomal anomaly being present:

... we were told that two of the tests came back and they were perfectly fine, there was a bit of an issue with the third result, then it went away for laboratory exploration At the moment, we were told that it was sort of a 66 percent likelihood of him having [severe chromosomal anomaly]. (Father07-Continued-Consultation4)

A clinical explanation of the results suggests a different picture, where rather than a 66% likelihood of the baby being affected, there was a 100% surety of the baby being affected, but only 66% of the cells:

It's unusual but it can happen that the FISH test comes back normal, but the full culture shows a mosaic [chromosomal anomaly]. It's because the full results give the cells time to grow... (Clinician04 – Consultation)

However, the parents interpreted this as the clinicians 'giving up' on the baby:

I mean this is like, it's like 66 percent, and it's not good enough to just say fuck it, is it really? (Father07–Continued–Consultation5)

This lack of a common understanding resulted in a number of noticeable changes in the dynamics of the consultations. First, was the adjustment from maternal to paternal led discussion. Initial consultations with each of the couples consisted of a 50/50 divide between mother and clinician, but this changed over the course of the pregnancy. In the final consultation between clinicians and one of the couples, the dialogue makeup changed to 40% clinician, 60% father, with the mother contributing four words in the 45 minute consultation. This change in dynamics was acknowledged by one of the fathers, as highlighted in the excerpt below:

.. see I don't want to be seen as if I'm talking over everything, because I'm not, but obviously if I've got stuff to say I'll say it like, you know, don't think I'm talking over [wife]. (Father07– Continued – Consultation4)

In a subsequent interview he explained that they had lost trust in the clinicians, and felt his wife was being pushed to terminate the pregnancy:

I mean they got things wrong, like the first test [FISH test]. There were just too many 'what ifs'... they just tried to railroad [wife]..... (Father7 – Continued)

Second, changes in the way the clinicians approached the parents were noted. The couples were labelled as “difficult to manage”, and interactions became defensive. The extracts below were taken from a 30 minute pre-consultation meeting between clinicians from fetal medicine, genetics, neonatology, and paediatric surgery, with the intention of establishing a common and consistent line:

After they had gone for the second opinion they phoned back again and asked why the amniocentesis hadn't been repeated. It was explained that there was no point

in repeating the test. He said they are a very difficult couple and we need to be very clear in what we are saying to them (Clinician09)

We need to make sure we complete an intra-partum care plan as I think she is likely to be quite difficult to manage (Clinician08)

The second set of quotes, taken from the same pre-consultation meeting, highlighted additional mechanisms for managing the parents' expectations, namely supporting the worst case scenario and removing hope:

But I'm not happy to consider offering anything at this point [in terms of surgery] (Clinician35)

My worry is that we are giving them a ray of hope and that it is only going to make things worse. (Clinician06)

Ultimately the parents detached from the process, as this extract from the subsequent consultation demonstrates:

... I'd sooner not come here again, that's where I'm at, I'm fucked off with it, ... I know that you guys cannot tell us any more than you're telling us and you have to make us aware of this, that and the other, but I'd sooner not fucking know, like forget the lot, forget it, forget, forget it, do not want to hear any more about it, do not want to hear any more about [chromosomal anomaly]. I do not want to hear any more about [structural anomaly]. We know what we know; we know what we need to know... Every time we go away from here there's shit going on in my head. There's shit going on in her head, ... It's like every time you talk to the boy he kicks you, and it's like,

*there's nothing wrong with him mate.... (Father07–
Continued – Consultation6)*

The disengagement from the process was demonstrated in the data derived from both couples. The uncertainty created by the difficulties in interpretation and explanation of antenatal testing, permanently scarred the relationship between the parents and clinicians. Counselling became defensive in an attempt to 'manage' the parents. The directive tactics used successfully with parents in the Choice Removed Group only served to increase the tensions between the two parties in this case, with the parents finally withdrawing completely.

6.8 DELAY/AVOIDANCE

The final group was entitled delay/avoidance. These women did not make an active decision, rather they continued their pregnancy by default.

6.8.1 SHARED ATTRIBUTES

The shared attributes of this group are presented in the table below;

Table 6-8 **Shared attributes of the Delay/Avoidance Group**

Sample size and outcome	Gestation at suspicion	Gestation at diagnosis	Gestation at TOP	Type of anomaly	Amount of certainty	Socio-economic status	Maternal Age	Partner present
4 Cont.	11-21 weeks	16-21 weeks	N/A	Structure and Chromo	Variable	Low	19-29	2 present

All of these women continued with their pregnancies. Parents in this group were all of low socioeconomic status and had the lowest average maternal age of all the groups. Two of the women did not have a partner. Educational attainment was particularly poor within this group, with the age of leaving education ranging between 15 and 18. The gestation at identification of a problem and gestation at diagnosis was variable, as was the type of anomaly present.

6.8.2 DELAY/AVOIDANCE BEHAVIOUR CHARACTERISTICS

Parents in this group drifted towards continuing the pregnancy without making an active decision. They perceived the role of the clinicians as a paternalistic one, and expected direction when necessary. This category was the most difficult to define because delaying coming to a decision could be seen as making a rational decision. However, four specific behavioural characteristics could be identified that suggested they should be categorised separately in the Delay/Avoidance box, namely: avoidance of information, difficulty accepting the diagnosis, misinterpretation of risk and uncertainty, and lack of engagement with the clinicians. Unlike the other group comprising of women of low socioeconomic status, the 'Choice Removed' Group, the doctor-patient relationship revealed little engagement from the outset. Failure to establish a solid relationship was perhaps the underlying cause for the different approaches used by the clinicians to the Delay/Avoidance and Choice Removed Groups.

The first behavioural characteristic exhibited by this group was an avoidance of information seeking:

*To be honest I haven't really looked for any more
information because my mind's not really in a place at
the minute (Mother05–Continued)*

Second, where a definitive diagnosis was given, a lack of understanding or acceptance of the outcome was often demonstrated. The extract below was taken from an interview with a parent whose baby had been identified as having a severe chromosomal anomaly with a number of associated structural problems also suspected:

*They go on and on about this [anomaly] and all the
problems and stuff ...I think they make it up half the
time... (Mother13– Continued)*

The extract suggests that the severity of the chromosomal anomaly had not been accepted.

All the parents were asked in the interview what advice they would give other parents who faced a similar scenario. Most expressed regret at not having understood what was being said and not questioning:

I'm not good at explaining like. I think what went wrong really was like we did not really say that we did not say when we did not understand like. (Mother04 – Continued)

Nonetheless, generally a very passive stance was taken regarding the outcome, suggesting they felt they had little control over the consequences:

Well there isn't much point in worry like. I mean what happens, happens really. (Mother11 - Continued)

Third, there was often a lack of understanding of the uncertainty that can come with antenatal diagnosis. Medicine was viewed as a perfect science. Parents interpreted clinicians' attempts to keep them informed of potential problems as being negative and looking for non-existent problems. One father added a little humour to his analysis of the situation stating:

I'm sure he's got a book and he just sticks a pin in it every time we come in. So next time ooh, green parrot disease today? (Father13– Continued)

Finally, relations between clinicians and parents were often stilted. The lack of engagement and poor communication between the parties is clearly visible in the consultation abstract below. (Consultation3)

Clinician07: *And your due date?*

Mother04: *30th May*

Clinician07: *And is it a boy or a girl?*

Mother04: *A girl*

Clinician07: *I did not know either way, so, have you got a name for her?*

Mother04: *Angel*

Clinician01: *It's not a big baby. Do you eat well, do you smoke?*

Mother04: *Yes, I smoked with Jamie too and she's fine.*

Clinician01: *How big was your last baby?*

Mother04: *5lb 6*

Clinician01: *Ok so it wasn't a big baby either*

Mother04: *No*

Clinician01: *Have you got anything else to ask?*

Mother04: *No*

Mother04: *[Laughing with friends]*

Clinician01: *Sorry what did you say?*

Mother04: *I was speaking to my friends*
[Laughter and whispering]
[Talking to friends and daughter]

Clinician01: *[Inputting details into computer]*

Clinician01: *So 4 weeks' time; any concerns with the baby's movements and you need to come in. And if you can reduce the smoking that will be great.*

Mother04: *[No response]*

What was notable was the attempt to engage with the mother at the start of the consultation. After a failure to get any interaction, the clinician stopped trying although the conversation between the second clinician and mother continued, in an equally stilted way.

During her interview she graphically described her feelings about the interactions with clinicians, and her perceived role as a patient:

Because doctors are so, you know they know such big words. And we're so young and stuff and we sit there and go, "What are you on about?" ... I mean you know you have to wear something nice and you have to go and say yeah yeah... (Mother04 – Continued)

The authority divide was clearly demarcated in the relationship, with the clinician's role perceived as a paternalistic one. When asked about why she had decided to have an amniocentesis, one mother responded:

The doctor said to have it [amniocentesis]. (Mother13 – Continued)

The issue of differing status and the assumption of deference to the clinician's authority were acknowledged by the clinicians themselves:

(Speaking of a 17 year old mother-to-be) Maybe she's not asking for a termination for reasons she cannot verbalise, but they are reasons that we could assure her about and she's closing up an option. And "this old man sitting opposite me, what does he know?" (Clinician10)

Midwives frequently proved a particularly valuable agent in the process of navigating between the two parties. Discussions between parents and midwives following consultations often revealed the need for clarification of practical points. For example, one mother was asked by the consultant to contact the secretary to make a follow up appointment for a scan. Although a deferent response was given, it became clear on subsequent discussion with the midwife that she did not understand what she had been asked to do:

... she, in there, she said that I need to ring her and make an appointment. What am I actually ringing them for?
(Mother05– Continued – Consultation4)

Although this ‘translator’ role was helpful in assisting towards a better understanding of issues for the parents, the roles and responsibilities of the midwife are significantly different to those of the clinicians and do not include ‘offering options’ but rather ‘supporting decisions’. Poor engagement with the clinicians had the potential for parents to miss out on the options available to them, with the affected pregnancy continued by default.

6.8.3 TENSIONS

The characteristics portrayed by the Delay/Avoid Group led to high levels of concern amongst clinicians:

... yeah there is that concern that some people may be, their default position is just to continue with the pregnancy because to actually go through that process of thinking, “Where might I be, what might the consequences be of this action or that action?” They’re either not willing to do that, or they really just don’t have the skills to do that. (Clinician 10)

The demographics of the women whose decision-making processes were categorised within this group were often very different to those of the clinicians who were caring for them. Some clinicians suggested that this could be problematic:

... some younger people and some people who are less used to dealing with professionals who will be a bit more withdrawn. They just want to be told what to do. (Clinician02)

As discussed, the midwife's role as a 'translator' became essential when caring for women in this group. However, one risk was that the options offered to women by the clinicians could not be presented again by the midwives due to their differing role. For these women, this may have resulted in a choice lost. Clinicians frequently highlighted requests from parents to direct them:

...you often get what should I do, what is the right thing to do, what would you do? I don't know and you cannot answer that. (Clinician09)

Here again the ideals of non-directiveness and the needs of parents appeared to be at odds. Unlike the parents in the Choice Removed Group, no relationship between clinicians and parents had been established here. This may explain the subsequent difference in behaviour of the clinicians, where a paternalistic pattern of behaviour was exhibited when caring for the Choice Removed Group and yet a rigid non-directive approach was adhered to when caring for the Avoid/Delay Group.

In this and similar situations, the tension between patient and clinician was tangible. On one side the parents were looking for direction, but were potentially unable to express this. On the other side, clinicians were fearful of directing the discussions in a situation where a relationship had not been established.

6.9 REVIEWING THE MODEL

This model has been developed through the application of data generated from 20 pregnant women and 18 partners, facing a decision following identification of a severe congenital anomaly. Care was taken to ensure that the model was generated from the data up, rather than by fitting a model to the data. Despite this, the model portrays hard boundaries, suggestive of a clear classification. This may be a reflection of the purposive sampling approach adopted where a broad and extreme spectrum of participants was sought. Application of the model to a larger sample may highlight parents who cross the boundaries of one classification to another. From this sample, only one couple appeared to do this.

In this instance, one couple presented characteristics that lay across those described in the Consequential Group and the Absolute Group. In terms of categorisation, the couple has ultimately been included within the Consequential Group, as their characteristics are representative of the group as a whole. However, this couple initially expressed strong religious beliefs, suggestive of those in the Absolute Group. The parents subsequently decided to terminate the pregnancy, which left them feeling isolated and cut off from support networks:

It's ironic really. At a time when I really need the [church community] support, I daren't tell them what I've done. So that's it really. It just adds to it all... (Mother19-Terminated)

This study has been able to explore tensions that arise as a result of the behaviours characterised within each group. Unique tensions, such as that expressed above, are likely to exist for parents who do not fit neatly into a defined category. This model is not intended to provide a classification for all parents but to highlight the range of responses. A larger sample may possibly highlight a spectrum rather than the absolute nature of the categories. The categories remain as guides rather than guarantees.

6.10 SUMMARY

This chapter has provided an in-depth exploration of the decision-making process from the clinician and parent perspectives. A series of six major themes were derived from the data collated. These were: information seeking, acceptance of diagnosis/prognosis, values and beliefs, future consequences, weighing up options, and engagement with healthcare professionals. Within each theme presented, the variability of responses of parents was highlighted. Each of these correlated with specific behaviours demonstrated by parents in the data, either reflectively within the interviews or observed in the consultations.

The subsequent patterns of behaviours that emerged formed the third part of the chapter, and were captured in the form of a conceptual model of the decision-making processes of the parents involved. A series of tensions arose between parents and clinicians. Although these tensions were not always specific to one of the model categories, interaction and techniques applied by clinicians to 'manage' the situations resulted in identifiable patterns or processes engaged in by clinicians and parents.

7 FINDINGS 3 – THE AFTERMATH, AND MAKING SENSE OF THE DECISION

This chapter represents predominantly the perspectives of the women and their partners who terminated an affected pregnancy and the enactment of the decision and making sense thereof. Whilst analysing the data it became evident that my initial definition of decision-making could have been broader, as the ‘making sense’ of decisions emerged as an integral part of the process. When the study was formulated, little consideration was given to this aspect. Interviews were conducted at different stages with those who continued and those who terminated their pregnancies. With hindsight it would have been useful to have undertaken further interviews with the women who continued their pregnancies, post-delivery. However, comparison of data between women who continue and those that terminate will always prove difficult temporally, as the outcome of surgery or procedures offered to babies born with severe anomalies is likely to influence parents’ narratives.

Ten pregnancies ended in termination; five of the women were categorised in the Consequential Group and five in the Choice Removed Group. The characteristics of the women in these groups are reiterated in the table below.

Table 7-1 Shared attributes of Consequential and Choice Removed Groups

Model Category	Gestation at suspicion	Gestation at diagnosis	Gestation at TOP	Type of anomaly	Level of certainty	Socio-economic status	Maternal Age
Consequential	9-18 weeks	13-19 weeks	13-20 weeks	Mainly chromo	Certain	High	36-42
Choice Removed	9-20 weeks	13-20 weeks	20-25 weeks	Structural / chromo	Variable	Low/ Medium	27-35

The impact of the multi-layered contexts will be explored in relation to the implementation of the termination process and with regards to the wider reaching social implications.

7.1 PREPARATION FOR TERMINATION

Following identification of a congenital anomaly, the care pathway was directed away from antenatal services to fetal medicine, although the co-location of this service may not make this obvious to parents. Parents and their babies were then cared for by a dedicated fetal medicine team. For those who decided to terminate the affected pregnancy, preparation for the procedure was undertaken within the fetal medicine setting before parents were transferred to a local maternity service to deliver the baby. In the majority of cases, the maternity service was in the same trust as the fetal medicine services, but this was not a pre-requisite. This pathway of care appears to be standard across England and Wales, with one notable exception in the North of England (Fisher 2013). Analysis of the data highlighted a number of key events that will now be examined in turn.

7.1.1 *CONSENT*

A great significance was placed by parents on the formality of signing a consent form for termination, and its integral part in the grieving process. All the women who terminated discussed the emotional impact that signing the consent form had on them:

I struggled to sign it. Although you've made the decision, you're still signing your baby's life to be stopped ... and that is really hard. (Mother12 - Terminated)

A coping mechanism employed by three of the women was to request that their husbands also sign the consent form. When interviewed, one of them explained that she had made this request in order to protect her relationship with her husband by ensuring future blame could not be apportioned individually. Although she understood the legal underpinnings of the consent form, she felt that the symbolic importance attached to the joint signing made it more of a shared decision:

I made [my husband] sign the consent form too.... I wanted to be sure that it was our decision, not just mine.

I know that legally it had to be my signature ... we both wanted to sign it.... That just felt right and it sort of shares out the guilt a bit. [Silence](Mother15 - Terminated)

For her, the symbolic significance of signing the consent form formalised the acceptance of responsibility for the decision.

7.1.2 FETICIDE

Feticide is offered in the NHS where gestation is 22 weeks or more (see section 1.5). This was actively adhered to in all the centres studied, as per RCOG guidelines (RCOG, 2010). This meant that none of the women in the Consequential Group required a feticide as the decision had been made to terminate before 22 weeks' gestation. Although the anomalies had been identified at similar gestations, as discussed in section 6.6 the women in the Choice Removed Group had taken longer to decide on a course of action. The unintended consequence of this was that many were faced with an additional decision over whether or not to undergo a feticide. Three women from the Choice Removed Group were offered a feticide. One refused and, whilst later acknowledging the risk of the baby being born alive, stated that the feticide procedure crossed her boundary of acceptability:

And the reason I did not want the needle in the heart was because I did not want to kill him while he was inside me. (Mother12 - Terminated)

The role of pregnant women as protector and nurturer of the unborn baby was widely expressed in the interviews. Within the literature there is a growing discourse in public health as well as medical forums reflecting this trend (Ettore 2002, Lupton 2013, McNaughton 2011).

Although terminations performed under Section E are exempt from the 24 week time constraint, the influence of the feticide time guidelines may equally impact on the urgency of the decision-making process in order to avoid the added trauma of undergoing a feticide. For the women and their partners concerned, the emotional impact of the procedure was immense:

Then it was doing it. When they injected the stuff to stop the heart that was just awful. I think that was the worst bit. (Mother15 - Terminated)

The finality of the feticide was difficult to manage for parents and clinicians alike. One woman suggested that the process was “*too clinical*” when the procedure was undertaken in near silence, while another found clinicians’ attempts to communicate increased her sense of guilt:

that [the clinician] was telling me what [name of the baby] was doing, and saying he was putting his arm around his chest and that got me, that upset me, because I did not want to know that. Cause to me he [the baby] is trying to stop it (Mother02 - Terminated)

The excerpt above highlights a tension, where the clinicians were criticised whatever action they took. The emotional impact of the feticide on the parents was clearly significant, with concerns over the procedure also echoed by the clinicians:

It's personally a difficult thing to do [feticide] because you are really blurring the edges between termination, and infanticide (Clinician10)

After the feticide, the women were rescanned about 30 minutes later, to ensure the fetal heart beat had stopped. At this point they were given the first dose of medication to induce the termination.

7.1.3 TAKING TABLETS

All women undergoing a medical termination were given a dose of medication in the clinic. They were then sent home for 36-48 hours, before being admitted to the labour ward for completion of the induction of labour. A number of midwives voiced concerns over the impact on parents of sending them home:

I always worry about them then [after they've taken the tablets]. We just send them off, and God knows what is going through their minds... (Midwife30)

Responses from the parents indicated that the relationship with the clinicians became very clinical once the termination process started:

... you just take that tablet and you're just sent away, that's pretty much how it felt. A very clinical process, so you come in and you take your tablet and then you are out (Mother19 – Terminated)

However, reactions of parents varied when asked about the act of taking the tablets:

And I thought that the actual doing of you know the taking of the tablet, I thought I'd feel guilty, you know I was expecting all these other emotions but I did not at all, I think we just knew that the decision was the right decision. (Mother09 - Terminated)

The excerpt above was fairly representative of the sentiments expressed by the women in the Consequential Group. One suggested that they found taking the tablet “a relief” as it meant that the waiting was over and they could “get on with it”. (Mother20)

A number of women in the Choice Removed Group continued to seek direction from the healthcare professionals, including in relation to procedures such as swallowing the pills. When offered the option to take the tablets home, one mother stated:

... if I went home and had to take them then I might not be able to. Just having someone there staring at me to

make sure I took them was better. (Mother12 - Terminated)

Medicalising the process, along with the desire to relinquish control to the healthcare professionals, appeared to be a mechanism employed by the parents, particularly those in the Choice Removed Group, to distance themselves from the process. Anxieties continued after taking the first tablets. Attempts at reassurance by healthcare professionals were ineffectual and perceived as dismissive:

I was worried something was going to happen while I was at home Yeah I was really worried about that, 'cause it was for 2 days ... Yeah and then when we asked [what we should do] the midwives said "Just carry on as normal" I mean, what does that mean? (Mother10 - Terminated)

Variability between the Consequential and Choice Removed Groups was again apparent at this point. Women in the Consequential Group, although expressing the emotional nature of those two days, conveyed a much more positive picture, recounting the various ways in which they had spent the time. For example:

In between the days between taking the tablets, we'd actually been to a funeral parlour to arrange everything, so we had already done it all in between. (Mother20 - Terminated)

All the women in this group responded by 'doing'. All activities focused, in different ways, on the pregnancy. These included arranging the funeral, washing nightdresses with the soap provided in the Bounty packs in preparation for the delivery, packing up any baby things that had been bought or sorting scan pictures. Although their actions varied, they all represented symbolic as well as practical aspects to managing the process. They appeared to have come to terms with their decision and, although this did not lessen their grief, they suggested their strategy

was that of 'moving on'. Conversely women in the Choice Removed Group recounted their experience of the waiting as anxiety filled and emotional:

*We just didn't know what to expect. I was so scared ...
We just sat at home and cried. I couldn't do anything or
think about anything (Mother02 – Terminated)*

7.2 DELIVERY FOLLOWING THE DECISION TO TERMINATE

This section provides a description of issues highlighted during the interviews relating to the delivery itself. Issues highlighted included lack of preparation, the impact of low staffing levels, and poor pain control. Forced disclosure, where parents had to disclose in front of others that they were having a termination in order to gain access to facilities, is also discussed.

I have found the following section difficult to write. Emotionally reliving the narratives has been distressing, but also frustrating as systematic failings in the service provided were identified. Although there was much praise and thanks to individuals for their support and care during the delivery period, overall the women who decided to terminate 'fell through a gap' where the system failed them all in one way or another. Recent literature reflects many of these findings at a national level (Fisher, Lafarge 2015). Forced disclosure and poor preparation for the physical aspects of termination, compounded by inadequate staffing levels resulted in women feeling deserted. This further reinforced their sense of guilt and self-blame. Narratives justifying acts of poor care, such as inadequate pain control, re-emphasise this perspective.

7.2.1 THE PRACTICALITIES OF TERMINATION OF PREGNANCY

The first issue to be addressed is that of forced disclosure, along with real or perceived judgement. One mother described her encounter with administrative staff, when she was forced to disclose that she was having a termination in front of other patients, in order to get access to the delivery suite:

And then had to say that I was booked in for a termination. And so there were people around and you could hear the (sharp intake of breathe), and so that was a bit uncomfortable. (Mother09 - Terminated)

Questioning by staff over the decision to terminate was also perceived as implying disapproval:

And I remember the anaesthetist actually came in and asked me why I was terminating ... and I thought that that was really insensitive at that point and I thought they should have known that anyway. (Mother12 - Terminated)

Couples sought acknowledgement from healthcare professionals that their termination was 'different' from other terminations. Failure to gain this left parents feeling judged:

It was almost as if she was there and thinking that there are these two young people who have had a bit of sex and did not want the kid, but that wasn't the story... We weren't two young scallywags who had been the back of a bike shed, we weren't that, we wanted this baby so much but we couldn't. And it was as if she was like, get out, get out you're taking up a bed. (Mother12 - Terminated)

This idea of 'good' terminations and 'bad' terminations stemming from 'good' and 'bad' reasons for having them has been previously reported (Norris et al., 2011). In terms of stigmatisation of termination, women who have had terminations for congenital anomalies may be both the stigmatiser and the stigmatised, believing they had 'good abortions' and distancing themselves from others who had 'bad abortions' (Rapp, 2000). One father expressed this succinctly:

Those people have a choice, but we did not have a choice. They had that choice. We had ours taken away, we did not get that choice (Father14 - Terminated)

The use of the phrase 'those people' conveys an immense power in the extract. Some parents actively attempted to create a divide between themselves and others essentially undergoing the same procedure, for reasons that they may not approve of. The use of terminology played an important role in this. In the example below, a clinician recounted the story of a previous patient who requested a termination for a severe anomaly at 28 weeks. However, she requested that the consent form was changed from the term termination, to the term induction, where induction indicates a medically induced labour. As such, women who are overdue may be induced; hence the connotations associated with the term 'termination' do not apply:

... she wanted to terminate at 28 weeks but wanted it to be called an induction and not a termination ... (Clinician10)

As with many of the parents interviewed, the use of the word termination, and even more so, abortion, was abhorrent. Analysis of the data using word frequency searches highlighted only seven occasions when the word 'termination' was used by parents, and only two occasions when 'abortion' was said. This occurred once in the context of describing a colleague who had had a number of abortions for apparently 'social' reasons, and again by a mother describing the connotations and stigma associated with the term abortion. The word 'termination' was used predominantly by parents who continued the pregnancy (and hence had not chosen this option). Silence and unfinished sentences frequently represented parents' way of communicating the word. Discussions surrounding the physical process of termination often prompted use of the word 'procedure' to express their meaning. Again this suggested a need to medicalise the process whilst distancing themselves from the implications associated with termination. Clinicians used the term termination, rather than abortion. Although there was a high frequency of use of the term in the interviews, use in consultations was rare,

with discussions phrased around 'choice', 'not continuing' or 'stopping the pregnancy'.

7.2.2 STAFFING

Staffing levels and access to care from an allocated midwife were highlighted by the parents as key factors in providing 'good care'. A number of guidelines supporting best practice have been published at national and local levels, including guidance from the National Institute for Health and Care Excellence (NICE) (NICE 2014). However, women frequently portrayed themselves as undeserving of care, universally expressing sympathy towards those who cared for them during delivery:

I kept on thinking at the time that it must be really tough for you to be the person on duty who had to deal with that [termination]. (Mother02 - Terminated)

Furthermore, when midwives were unable to attend to them, parents suggested that the needs of other women should rightly take priority over their own:

Then we did not hear from her, but she was with another patient and obviously that is her assignment and her priority.... (Mother10 – Terminated)

In the extract above, the parents highlighted the difficulties encountered when one midwife was allocated to care for more than one woman. Where the needs of the other patient conflicted with their own, the 'live' birth was perceived as the priority.

Individual midwives and clinicians were frequently praised for the care provided. When problems were encountered, these were subsequently, with a couple of notable exceptions, framed as systemic challenges with staffing numbers and provisions:

The nurse was absolutely lovely but I think it was just provisions for it, I mean they were just too busy. I mean it's the provision side of it. It's a systems failure really (Mother20 - Terminated)

The need for dedicated staff to care for women undergoing termination was discussed and the advantages of a dedicated midwife widely appreciated:

...it would be nice to have more support for them. I mean having a dedicated midwife for their care so that they can actually be with that midwife and post-natally having bereavement counsellors available as well... (Mother02- Terminated)

Nonetheless, lack of resources repeatedly made these ideals unachievable and women frequently reported delivering alone:

And the midwife who was on, I guess she has quite a few patients at the same time, but she was actually on her break at the time, she had gone on her break when the baby arrived so my husband actually delivered the baby 'cause there was nobody there. (Mother20 - Terminated)

Subsequent justification for the midwife's absence was made by the parents in each case:

But maybe they weren't expecting it all to happen so quickly and you cannot just have someone sitting there for 3 hours while they are waiting for the tablets to work. But that was really tough (Mother16 - Terminated)

For women who had previously experienced childbirth, the shock of the delivery process following a termination was no less traumatising:

I think in my mind I thought it would be like when you delivered a baby but just on a much smaller scale. So you might be in bed or wherever. I wasn't expecting to be on a commode. (Mother19 - Terminated)

For a late medical termination, as in this instance, delivery on a commode may be surprising. No midwife was present at the time to advise and the side effects of the drugs, such as diarrhoea, may have been the reason for delivering in that way. For all, the physical process was incredibly distressing, and intensified the emotional turmoil they were already experiencing:

And you know it's just sort of it's just a bit degrading and horrifying to be in that situation really. (Mother14-Terminated)

Recent studies have suggested that partners were often perceived by women as their primary supporter (Lafarge *et al.* 2013, Fisher, Lafarge 2015). However, lack of preparation and understanding of the process led some women to exclude their husbands from the delivery process, resulting in further isolation and distress:

I was just so scared, it was horrific. I had to send [husband] out, because it's just not something that someone else should see...so I delivered on my own, in the toilet ...and then there's the guilt of what you've done (crying) (Mother19- Terminated)

The ongoing sense of guilt expressed by many mothers further compounded their sense of desolation.

7.2.3 PREPARATION AND DELIVERY

Absence of information on the physical aspect of terminating was highlighted by all the women. Lack of access to antenatal preparation was a major issue, as antenatal care had focussed on the identification of the anomaly and subsequent

decision-making process. The narratives of parents suggested a universal lack of preparedness for the emotional and physical aspects of the delivery:

We were really in the hands of the people at the hospital and all that, and we really did not know what was happening, what it would be like. Because obviously we had no antenatal or anything like that. (Mother12 - Terminated)

Although the parents generally spoke highly of their care in fetal medicine, there was a sense of being deserted at this point. The positive impact of brief visits to the delivery suite by clinicians and midwives from fetal medicine was highlighted by a number of the women:

... they [fetal medicine clinician and midwife] came up to see me when I came in [to deliver]. That was so reassuring; I mean I just felt so lost up there [delivery suite]. (Mother02 – Terminated)

Although there was no expectation of seeing the fetal medicine clinicians again once the termination procedure had commenced, these visits provided reassurance not only for the immediate situation but also hope and ‘permission’ to start to plan for the future:

[fetal medicine clinician] was fantastic. I mean s/he even came up to us afterwards and s/he came in after the birth and said how you are. I mean s/he did not need to. But s/he was great. And s/he said we would see you again [for screening in subsequent pregnancies]. (Mother02 – Terminated)

Little consideration had been given to practicalities of termination, and what to expect. Although this was highlighted as a problem during the period after taking the first tablets, the impact during delivery was significantly greater. The excerpt

below highlights this problem, as it describes one woman's experience as she waited in the maternity unit to deliver:

... So one of the side effects of taking the drug is that you get really bad diarrhoea. But I did not know that and while it was happening I did not know whether that was normal, I kind of went into a bit of shock, and I was shaking and being sick. (Mother19 - Terminated)

The realities of the delivery itself were similarly poorly prepared for. One mother described her surprise, when she found herself delivering the placenta following the arrival of the baby:

... I did not have a clue what I was doing... I did not realise that I was going to have to do that ... it would be good if someone talked you through it all because you don't get lessons or anything. (Mother09 - Terminated)

The positive relationship between preparation for hospital episodes and subsequent perception of pain and recovery rates has been widely discussed within the medical literature for decades (Egbert *et al.* 1964, Janis 1958, Kiecolt-Glaser *et al.* 1998). Speed of accessing treatment, as well as the immense emotional context in which the procedure was being undertaken, is likely to have impacted on the amount of information given, as well as retained. Poor pain control was also an issue raised by all the women:

And then it took about a good hour for them to get the anaesthetist in to give me any pain relief ... by that point I was in excruciating pain. (Mother19 - Terminated)

The few complaints about individual staff members all stemmed from issues pertaining to accessing pain control. As described in the excerpt below, when an anaesthetist finally arrived to set up the pain control, the parents felt they were causing an inconvenience, by delaying the end of the shift:

And then the anaesthetist s/he was really mardy¹⁶. She was just about to go off a shift so she really did not want to be there. (Mother12 – Terminated)

For many, access to pain control was compounded by feelings of isolation and lack of preparation:

It wasn't just the pain relief it was somebody there, just having somebody there. Because we did not know what. At no point were we told this is what is going to happen (Mother20 - Terminated)

7.2.4 FACILITIES

Attempts by the centres to care for parents who had delivered post termination away from others compounded staffing issues. The facilities offered differed in each of the centres studied. Some had bereavement suites, others private facilities away from the main delivery suite. At the time of writing, one of the centres still cared for the parents on the delivery suite alongside other labouring women, although plans were in place to develop a separate bereavement suite in the future. Despite these intentions, the bereavement facilities had restrictions on use. Where women opted for epidurals for pain relief, they were placed in a delivery room in the main facility. Post-delivery, centres aimed wherever possible to continue to care for the women in the room they had delivered. However, a number of the participants were transferred to the main postnatal or antenatal ward due to bed shortages in the delivery suite. This proved distressing for those involved, as one mother described:

When I was giving birth, it was ... with all the other people giving birth and then afterwards I went onto a

¹⁶ Mardy is a regional word that stems from mar meaning to spoil. It is an adjective meaning sulky, complaining or petulant.

ward with other people and their babies. And obviously people see me and think, "Where's her baby? She's never with her baby." (Mother12 - Terminated)

The women described themselves as being left in "no-man's land" (Mother 15), neither an expectant woman, nor a mother. This evoked feelings of being judged and scrutinised by others around them. These feelings were reinforced by the physical space in which they received care:

There is nowhere to put people in that situation you are in that sort of in between the place where yes you are a pregnant woman and you need to be treated like one but at the same time you are grieving. (Mother14 - Terminated)

Where fetal medicine and antenatal care were combined, delivery within an environment whose primary role was to accommodate women during delivery meant the women felt out of place and deviant. Sharing the space with expectant mothers was a further reminder of what they had lost:

So in a way you're mourning but then you're still, when you've got to go into the delivery suite, you're mourning already and again you see all the expectant mothers.... (Mother16- Terminated)

For those who were placed away from other mothers, in general, the distance from the sights and sounds of newborns, was appreciated:

The only good thing was ... we did not see a single baby, which was nice, especially when you know you are losing your baby. You're in a separate part...it was delivery but at completely the other end (Mother10- Terminated)

Despite acknowledging the benefit of being cared for away from the main delivery suite, some interpreted this as a strategy to protect the other women rather than themselves:

But it was as if it was like, "keep out the way" because these other women don't want to see you. (Father10-Terminated)

The use of the phrase "other women" implies a perception of being different to other labouring women. Delivery suites are designed for delivery of babies. Commonly women are cared for on the antenatal ward prior to delivery, until they are in labour. Women admitted for delivery following a termination are given the final medication to induce labour at the time of admission. Depending on factors such as number of previous deliveries, the length of time they were in hospital varied considerably. On occasions they would wait for a number of hours before labour started. The lack of provisions to support the parents during this time was noted, particularly by the fathers:

It does feel a bit like they forget the father sometimes you know. It was like the bed in the hospital and there was no bed for me. You know, not even a blanket, and she said there wasn't enough pillows even for the patients. And the blanket was a sheet and that was it. So I wrapped up my jumper (Father10 - Terminated)

For the majority of parents, this lack of facilities was highlighted as the major problem encountered by fathers. This included lack of an additional bed or space for the father to sit, blankets or covers and food. This was particularly an issue at the weekend when the hospital shops and café were closed. In addition, some fathers complained about the lack of acknowledgement from members of staff:

Yeah I know that I'm not actually carrying a baby but it is my baby as well. But she did not even ask my name.

[Talking of the midwife caring for his wife.] (Father16- Terminated)

7.2.5 SEEING THE ANOMALY

Where a diagnosis of a visible anomaly had been made, parents frequently expressed a fear over what the baby would look like once delivered. These concerns were compounded where the women found themselves delivering without midwifery assistance:

The midwife wasn't even there. I mean she came back and it was all done...We did not want to see the baby; really I did not want to. (Mother12- Terminated)

Fear of how the baby might appear led a number of the women and partners to avoid contact with the baby:

... we did not know what he was going to look like. So that scared us even more. So we wanted them to take him away and then ask them what he did look like so we could prepare ourselves (Mother16- Terminated)

Midwives played an important role in preparing the parents and dressing or wrapping the baby. For some parents, viewing the anomaly was reassuring. In the example below, the visible deformities provided some comfort to the parents, knowing that they had made the right decision:

... and I guess that as soon as we saw our little [baby] we knew that we'd, you know [made the right decision]. We could physically see the abnormalities you know she had a cleft lip even though [the] fingers and toes were all formed perfectly, [the] brain was not, and half [the] skull was missing. So we were aware with the complications of the abnormalities. (Mother20- Terminated)

Unlike those in the previous excerpts, the parents in this instance were prepared for the physical defects. The potential for added distress was turned into a positive. Although there was not a clear divide between the Consequential and Choice Removed Groups in terms of how the anomaly would present, the Consequential Group had generally sought out information on the likely appearance of the anomaly, whilst parents in the Choice Removed Group were more likely to be unprepared.

For those delivering a baby with an asymptomatic anomaly (internal structural anomalies not visible externally), concerns about terminating a healthy baby were compounded by the normal appearance. Unlike the parents delivering a baby with a visible anomaly, post-mortems were requested by all these parents, and played an essential role in confirming diagnosis:

I think once we've had the results and they say "Yes we were right on this, we were right on that" I mean I think just at the moment it's still not knowing 100%. Really we're just taking the doctors' word for it [the diagnosis] at the moment. (Mother02- Terminated)

Ongoing fears were voiced by these parents as they searched for confirmation that they had made the 'right' decision. Parents in the Choice Removed Group, in particular, expressed high levels of anxiety that the clinicians may have been wrong. This may have related to the way in which their decisions were made. Unlike the Consequential Group, who had accepted the poor prognosis and subsequently taken responsibility for an active decision, the parents in the Choice Removed Group had largely relied on the clinicians to direct them. Their decisions had been based on the recommendations of the clinicians rather than their own research. The impact of the lack of visibility of the anomaly at birth may have compounded any pre-existing doubts. Delays in post-mortem results further increased the distress experienced.

The desire for a post-mortem was not shared by all the women. For some, religious beliefs did not permit post-mortems, and others felt they could not endure the thought of their baby being “cut up”:

Just seeing him there. And they said they would have to cut him up and take bits of his organs and stuff but they might not be in the right place and I just couldn't deal with that... (Mother12- Terminated)

Within this study, all the women who declined a post-mortem had delivered a baby with an external, visible defect, and therefore the reassurance a post-mortem would give was less urgent.

7.2.6 LEAVING THE HOSPITAL

The finality of leaving the hospital without their baby provided another point of immense stress for the parents, as it further emphasised the differences between themselves and others:

But I think out of everything, leaving the hospital was probably the worst possible thing... it was... heart wrenching. (Mother12- Terminated)

Parents responded very differently following delivery. Some chose to stay in hospital a little longer, in order to be near their baby:

We stayed an extra night. I just needed to be near [the baby] (Mother06- Terminated)

Others discharged themselves against medical advice in order to get away as fast as possible:

As soon as I'd had [the baby] I left, I just couldn't stand to be there. I needed to be home with my family. (Mother14- Terminated)

Women who went home frequently highlighted simple reassurances offered by midwives that their baby would be cared for as rationale for not staying in hospital:

*And to be fair to the midwife, she made me feel better by saying that she would take care of him and make sure he had all his stuff you know like his teddy and things.
(Mother12-Terminate)*

Conversely, those who stayed frequently expressed a lack of trust that the midwives would care for their baby:

*I don't know ... it's because when she's in the hospital they're not going to treat them nicely [the baby]
(Mother20- Terminated)*

It is unlikely that this and similar comments suggested that midwives would intentionally harm a baby. More likely is that the women had concerns that the midwives would not perceive their baby as 'human' but rather as 'expelled contents of conception'.

The disjointed care pathway between fetal medicine and obstetrics was clearly visible. Maintaining contact with the fetal medicine service provided comfort and hope for parents. Care provision for those who terminate a pregnancy appears to lack the necessary flexibility to ensure that this group of parents' specific needs are met. The establishment of approved care pathways that incorporate an extension to the fetal medicine role may perhaps provide the foundations to bridge the gap.

7.3 SOCIETAL SUPPORT

The societal support and rituals that are then engaged in once they leave the hospital contribute to the social understanding of the journey they have embarked on. As highlighted in Table 1-4, the legal definition of a birth is associated with varying degrees of financial and social support. Societal support comes in the form of tangible resources such as financial support or time off work as well as emotional resources that arise as a result of broader issues such as public perception and subsequent stigma attached to termination.

The gestation at which a termination is performed can have significant financial implications. Women who deliver before 24 weeks, and where the baby shows no signs of life at birth are excluded from maternity benefits, including time off work or financial assistance. Women undergoing a feticide before the end of their 24th week of pregnancy would fall into this same category of late miscarriage and lose any entitlements to maternity leave or pay. This created additional difficulties for some women. As one clinician explained:

It can have enormous financial repercussions... But I don't think women make decisions on that basis in the main, though I will often talk to them about it, because for some women, being in receipt of maternity benefit will make a big difference in terms of how long they can have off work. And if they are going to need a considerable amount of time to get over something, as many women do, that can be quite advantageous.
(Clinician17)

For those who terminated before 24 weeks, the level of support offered by employers was variable. Although legally they were entitled to take sick leave for as long as required, their need for time to recover was often overshadowed by financial concerns. Even in the event of a sympathetic employer, a number of parents raised the issue of finances and the difficulties they had encountered in respect to loss of income:

'Cause you still worry about it because you do a lot of overtime and that, don't you? (Father16 - Terminated)

The impact on women reliant on additional payments such as overtime or those on zero hours contracts were most likely to be affected. These tended to be women from lower socioeconomic groups. The financial implications for these women had the potential to be greater. For many of the women, the lack of official recognition that they had had a baby was compounded by the subsequent financial implications:

I find it ironic that if I had carried on with the pregnancy for just over another 2 weeks I would have been able to take maternity leave and get maternity benefit and all those things. I mean I would then have had a baby, but I just had, you know..... It feels like no one cares for my baby because she never existed really. (Mother06 - Terminated)

Seeking recognition of the baby was a theme heard throughout the interviews. Society "polices" bereavement by putting in place requirements and rules that must be adhered to (Walter 2001, pg.123). Without a baby to grieve for, parents cannot access the social rituals designed to bring comfort. These include aspects such as burials or funerals and permanent reminders of their existence in terms of graves and headstones and allow their grief to be shared. Supported by organisations such as SANDS, midwives in fetal medicine appear to have played an important role in enabling recognition of the baby:

I'll never forget what she said. She said that I was making the first decision as a mum. (Mother10 - Terminated)

7.4 STAYING MUM – DISCLOSURE AND STIGMA

The stigmatisation of those involved in termination is well documented (Kumar *et al.* 2009, Norris *et al.* 2011). For the parents, this could result in social isolation where disclosure to peers was avoided, and hence support was not sought, due to fear of stigmatisation. Analysis within this study highlighted a clear demarcation between women in their willingness to disclose. Women in the Consequential Group expressed no anxiety over disclosure. They appeared more confident in their decision, as well as the acceptance of this amongst their social group. Conversely, the women in the Choice Removed Group expressed severe reservations over disclosing information about their decision to others. Fear of stigma and judgement from others was widely expressed. Perhaps as a mechanism to overcome this, they attempted to separate themselves from women undergoing terminations under Section C. Previous work has highlighted the correlation between stigma and poor health outcomes, including psychological or mental health (Link, Phelan 2001), thus placing those in the Choice Removed Group at high risk. As discussed in section 6.6, this group consisted predominantly of families from low to middle socioeconomic status, and as such were at higher risk of experiencing regret and poor health outcomes than those from higher socioeconomic groups (Statham *et al.* 2000).

The participants were divided on their decision whether to disclose, partially disclose (to certain people only), or not to disclose. The decision to disclose to others was based on the perceived risk of stigma. Stigmatisation is a deeply contextual, dynamic social process. It relates to the degradation of a person through a particular attribute which violates social expectations (Norris *et al.* 2011). Goffman described stigma as “*an attribute that is deeply discrediting*” that results in the bearer transferring “*from a whole and usual person to a tainted, discounted one*” (Goffman 1963, pg.3). Abortion stigma has been defined as “*a negative attribute ascribed to women who seek to terminate a pregnancy that marks them, internally or externally, as inferior to ideals of womanhood*” (Kumar *et al.* 2009, pg.628). One of the peculiarities of this is that it can be considered a “*concealable*” stigma (Norris *et al.* 2011, pg. S50). It is unknown to others unless

disclosed, although none are fully in control of whether their status is revealed (Quinn, Chaudior 2009). Consequently, those stigmatised by termination have to manage the stigma once revealed, but also the decision on whether or not to disclose (Quinn, Chaudior 2009). Amongst the women, there was a clear demarcation between the Consequential and Choice Removed Groups in their decision to disclose or not. The excerpt below comes from a mother in the Choice Removed Group. She decided to partially disclose by telling some people and not others, based on how well she knew them, and their likely reaction:

I struggle with that a lot. Um, like I've had to tell the girl at work that we lost it rather than what we decided to do, to end the pregnancy. (Mother02 - Terminated)

The decision not to fully disclose came with an additional set of guilt:

I mean I did not tell them the truth, well only one person, the rest think I had a miscarriage. So I go between feeling guilty because they were being kind to me because they thought I'd lost my baby while actually I had decided to abort her. I did not really deserve their sympathy. (Mother15- Terminated)

The predominant reasons for not disclosing arose from fear of judgement and stigmatisation due to perceived strong societal disapproval over termination:

Abortion has such strong, you know people have such strong views about it and I just don't want to have to justify my decision to other people. (Mother12 - Terminated)

Conversely the women from the Consequential Group all disclosed:

I know the group (of work colleagues) well. And there are going to be people who disagree with what we have done. But it was only us in that situation and so no that

[disclosure] really did not [concern me]. I think because of the age we are as well it was different. I think if I had been in my early 20's you know. You've got different pressures and you've not lived have you? I mean I think because we are older we looked at things differently.
(Mother09 - Terminated)

Women in this group appeared more confident with their decision. The understanding that others may potentially disagree was widely expressed. The parents in the Consequential Group also expressed an understanding that there was no right or wrong decision, and regret was likely for those who had continued as much as for those who terminated:

Don't listen to what other people say, go by your own instinct. There is no right or wrong answer, there isn't. Whatever decision you make ... there will be moments when you regret things, I'm sure that people who carried on with their pregnancy sometimes regret that they did.
(Mother19 - Terminated)

Unlike the mothers, irrespective of the decision-making group, all the fathers found it difficult to disclose:

[Talking of disclosure] Not really I guess. I mean I don't tend to tell anyone unless we know them really well. I mean you know some people have really strong views and I don't feel that it's any of their business what we did and I don't really want to talk about it to them.
(Father12 - Terminated)

Within the interviews, it was frequently the women who responded to the question of disclosure on behalf of their partners:

... he's [her husband] not told anyone at work ... he went back to work and the chap [work colleague] said "Have you had a week, a good break" And [husband] just went "No" and that was the end of the conversation! (Mother09 - Terminated)

The difficulties experienced by fathers' in terms of disclosure appeared to arise from their universally perceived role as supporter for their wife. Organisational factors such as consent practices reinforced this sense that their needs came second to that of the mother. Support for the fathers was also perceived as lacking. This was reflected in many of their responses to taking part in the interview, where many fathers initially assumed that their participation was not required. Others continued with that stance during the interview, allowing the mother to take the lead. Questions directed to the father ensured a more equal participation. This subsequently generated discussions between the couple. Much of this revolved around perceptions that the mothers had about the way in which the fathers had coped:

He just seemed to be able to distance himself somehow. It's, it's... I don't know, sort of like he could look at the problem from a distance while I just felt like I was part of it, in the middle of it, I don't know. (Mother15 - Terminated)

In situations where the mothers felt that disclosure was acceptable, they expressed surprise at the lack of disclosure from their partners. For some fathers, talking to strangers offered an acceptable option. In the excerpt below, one father discussed his disclosure to taxi drivers:

Taxi drivers! I've told a few people but I'm bottling it all up. I'm trying to be strong for everybody. Because you're struggling aren't you? (Father15 - Terminated)

Irrespective of the decision to continue or terminate, the fathers universally saw their role as being the strong one, there to support everyone else, even though they acknowledged that lack of emotion may be interpreted as lack of care:

Yes so I've got to be strong and carry on. But it may look like I don't care. But when I sit [at work] you cannot help but dwell on it can you? (Father03 - Continued)

One father provided further insight when questioned:

I did not give it much thought. I mean from my thoughts it's more about how my wife is, so that's all I've done really. I've not really thought about what I've needed... I don't feel I've really thought about myself (Father19 - Terminated)

Many mothers suggested that their partners often reached a decision before them, while they sometimes had ongoing doubts:

[husband] always thought it was the right thing to do, but in my mind, was it? I mean as well as I knew what we were going to do, I had that doubt. (Mother16- Terminated)

This could lead to tensions between the couple:

[husband] got there much quicker than I did. He just seemed to know what was best really. I hated him for it at the time. (Mother15 - Terminated)

Many of the parents spoke about the difficulties it placed on their relationship. Irrespective of the decision to continue or terminate the pregnancy, a huge stress was applied to the couple:

*I can imagine that this could break a lot of relationships.
I guess that was where [husband] was really good. He
never blamed me (Mother02 - Terminated)*

The excerpt above suggests that added to the stress of the decision was the guilt attached to the diagnosis. In cases of spina bifida particularly, where primary prevention in terms of folic acid tablets are known to reduce the risk, women frequently blamed themselves. Communication difficulties were often perceived as a major cause of arguments:

*But we have fell out about it a few times. And sometimes
I feel like since we found out about it our relationship
has kind of broke down a bit as well. And it is taking its
toll because he doesn't want to talk about it and I can be
quite opinionated. (Mother05 - Continued)*

The excerpt above was taken from an interview with a couple who decided to continue with the affected pregnancy, and highlights the toll taken on the relationships of all the couples. For some it became too much and one of the couples recruited broke up within a few weeks. The mother continued with the pregnancy alone.

7.5 SUMMARY

The narratives of the women and their partners who terminated a pregnancy affected by a severe congenital anomaly contribute to the major findings within this chapter. Their experiences of the processes preparing for the termination, the delivery itself, and the immediate aftermath are examined. This chapter has also provided a platform for the important role of the midwives whose voices have remained relatively silent throughout this process. This has highlighted the move to medicalise the care of women and their babies where a severe congenital anomaly has been identified.

Preparation for the termination was managed in the fetal medicine unit. A number of key events were identified from the data. These included signing the consent form, feticide and initiating the termination. Great symbolism was placed on processes often construed as 'routine' or incidental by clinicians. In particular the signing of the consent was perceived as a highly emotive event by parents. Legally, the responsibility for the decision is placed on the women. Meanwhile the drive for 'informed choice' has shifted the clinicians' perception of the signing of the consent form from the legality of consent to a formality at the end of a process. The result is a tension between the legal and practical requirements of consent.

The feticide procedure was especially harrowing for parents and clinicians alike. A constant tension existed whereby clinicians were seen as either 'too clinical' when they undertook the procedure in silence or 'insensitive' when they talked through the procedure.

As with the signing of the consent form, taking the tablets was a symbolic event, signifying the death of the baby. The location of counselling rooms within the antenatal setting increased parents' distress. Parents managed the process in a variety of ways. The characteristics of the groups identified in section 6.2, were evident in the various mechanisms employed to cope.

In relation to the delivery, findings suggest that these parents 'fell through a gap' in care services, when they were passed from fetal medicine to the maternity services in which they delivered. Issues relating to their lack of preparation for the procedure, inadequate staffing levels to support their individual needs, poor pain control and facilities that met their physical but not psychological needs were highlighted throughout. Although parents' concerns were generally levied at the system rather than individual staff, there appeared to be a lack of understanding amongst staff in the delivery suite of the unique needs of this group of parents.

In the wider context, parents universally sought acknowledgement, and subsequently an identity for the baby. This enabled parents to engage in socially recognised rituals to mourn their baby. In addition, it allowed their grief to be a

shared experience. Fathers encountered their own unique difficulties. Primarily they perceived their role as protector of their partner. This frequently led to tensions between the couple, where the mothers potentially misinterpreted the fathers' actions as demonstrating a lack of care for the baby.

8 DISCUSSION

Exploring the experience of decision-making by women and their partners following suspicion or diagnosis of a severe congenital anomaly has provided new insights into the difficulties encountered by the parents and clinicians alike. As with other sensitive research topics, there is often reluctance from clinicians, governance officials, and researchers to engage, due to a desire to protect participants at an already emotional time (Lee 1993). However, this has resulted in a restricted evidence base on which to develop recommendations for care for this group. This study was designed to explore the experiences of women and their partners following suspicion or diagnosis of a severe congenital anomaly. Particular emphasis was placed on the decision-making processes employed. Untangling the multi-layered contextual framework in which the decision-making process was enacted has provided insight into the tensions which parents and clinicians face, and the impact of these on perceived options and decisions. In addition, the impact of, or making sense of, the decision provided an important perspective on this under-investigated area, and will enable a more informed view to be established on how best to support women and their partners in these circumstances.

The volume of data generated within this study has provided a significant contribution to our understanding of the decision-making processes of women and their partners following diagnosis or suspicion of a severe congenital anomaly. Analysis of the 80 plus hours of recordings, including consultations and interviews with clinicians and parents, has provided the basis from which these findings have emerged. These will now be placed in their broader context throughout this discussion chapter.

The chapter is presented in seven sections. First, in a brief summary, the key findings are reiterated and contextualised within the current literature. Second, the strengths and limitations of this study are considered. Two over-arching themes, namely 'decision-making' and 'identity' are subsequently presented in third and fourth sections respectively. The fifth section returns to the starting

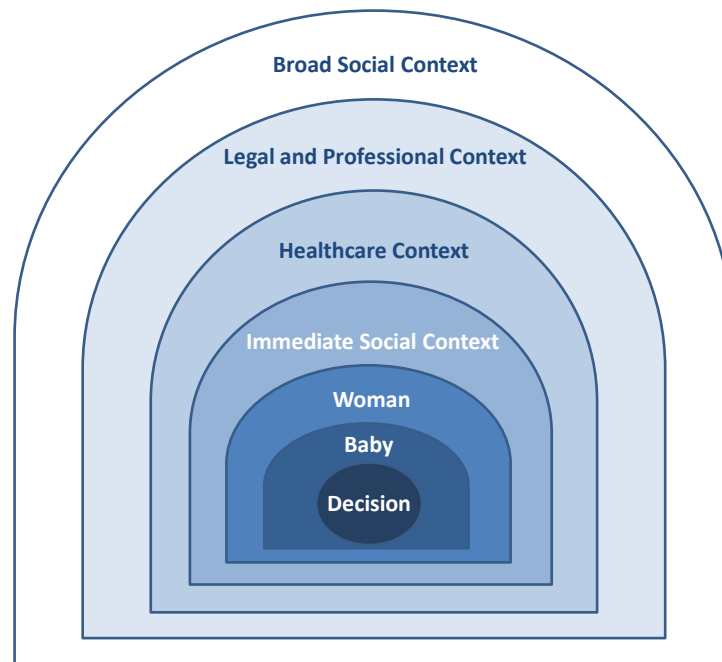
point of the thesis by examining the findings within the context of variations and inequalities. The issues highlighted are subsequently drawn out and presented as a number of key recommendations in the sixth section. Finally a summary and conclusion to the study are provided.

8.1 SUMMARY OF THE STUDY

Capturing and presenting the multiple complexities of the decision-making process has proved challenging. A temporal approach has provided an effective mechanism through which to examine these complexities. It is clear that decision-making and the processes involved do not occur in isolation, but within the constraints and pressures applied across a multilevel contextual framework. Identification of the commonalities and differences in the decision-making processes enacted by groups of parents enabled the development of a conceptual decision-making model. This proved a useful structure in which to explore the tensions that arose within the interactions between clinicians and parents. For the women who terminated their pregnancy, the aftermath and mechanisms employed to make sense of their decision became a strong theme.

Despite the uniqueness of each decision-making process, shared influences and factors were apparent. This is summarised within the contextual framework presented in Chapter 5 and reproduced in Figure 8-1.

Figure 8-1 Summary of the contextual framework in which decision-making occurs



Variables relating to the baby, including the type of anomaly identified, along with those pertaining to the mother, such as personal beliefs and values and previous experience or knowledge of the anomaly, constitute the first and second contextual layers. The influence of these variables is presented in Section 2.2, and was prominent in determining the sampling frame from which the study population was selected. Family, partner and immediate social support networks further influence the process and subsequent decision made. Although some of these variables were shared, their influence on the decision-making process of each woman differed.

What was common with regards to the decision-making processes was the influence of the external contextual layers; namely the healthcare, the professional and legal, and broader social contexts. Together these determined the context in which the decisions were enacted. Fear of scrutiny, generated within the broader social context, led clinicians to practise defensively despite the freedom applied by the legal context in England, where the law remains open to interpretation. The healthcare context, too, perhaps unintentionally, constrains and confuses the decision-making process through its representation of screening as an integral

part of antenatal care. In addition, the hybrid nature of scanning in particular impacts on the way the status of a baby is constructed while still in-utero.

The impact of the wider contextual layers on clinicians, in terms of their response to the constraints applied to them, was reflected in their interactions with the parents. This resulted in an expectation, on the part of clinicians, that an ideal decision-making process would be followed. Thus the process became more important than the decision itself. Failure to demonstrate adherence to this ideal process gave rise to tensions between parents and clinicians.

Whilst considering the contextual framework identified above, six themes inductively emerged from the data collated from parents relating to decision-making, namely: information seeking, namely the approach to and need for information, acceptance, referring to parents' ability to come to terms with the diagnosis, values and beliefs, in terms of belief systems including religion, imagined futures, relating to the consideration for future consequences, weighing up options, whether the decision was head or heart led, and engagement with healthcare professionals, pertaining to the dynamics of the parental relationship with healthcare professionals. A spectrum of responses within each theme was noted and used to categorise the decision-making processes of parents. These were grouped into four main categories that were labelled: Consequential, Absolute, Avoid/Delay and Assess/Reassess. The latter was subsequently further subdivided into Choice Removed and Choice Disturbed. The grouping was undertaken on the basis of a number of shared decision-making traits. After grouping, it was apparent that there were a number of similarities about the women, including their decision outcome and socioeconomic status within these groups.

The Consequential Group actively engaged in an 'ideal' decision-making process, thus reflecting the process sought by clinicians. Meanwhile, the Choice Removed Group experienced difficulties committing to a decision and were finally 'pushed' by clinicians into making a decision. Although all the women from both of these groups terminated their pregnancies, the process through which the decision was

made varied substantially. The groups also differed in socioeconomic status, with all the women in the Choice Removed Group of a lower socioeconomic status than those in the Consequential Group.

The women in the remaining groups all continued with the affected pregnancy, yet again their decision-making processes differed significantly. Whilst the fundamental belief system of the Absolute Group determined that there was essentially no decision to make, the Avoid/Delay Group continued more through default than active decision-making. In turn, a critical event resulted in the Choice Disturbed Group disengaging from clinicians. Breakdown in the relationship resulted in a loss of trust and as a result the parents continued with the pregnancy. All of the women who continued with their pregnancies were of low to medium socioeconomic status, apart from one of the couples in the Choice Disturbed Group who was of high socioeconomic status. Other attributes, including the type of anomaly, the association of a chromosomal disorder, and gestational age at diagnosis, were variably distributed within all the groups identified.

Although the sample consisted of an equal number of women who terminated and continued, the experiences of women who terminated their pregnancy constituted a larger proportion of the study. Over the duration of the study, the temporal dimension of the decision-making process became more apparent and the scope extended to include making sense of the decision ultimately reached.

‘Falling through the gap’ became the over-riding theme for the women who terminated their pregnancies. Care pathways were designed to meet their needs within the fetal medicine departments, but once the decision had been made to terminate, they no longer belonged. Care was then transferred to the maternity units, where the women felt out of place. A number of practical issues were highlighted, and tackling these could provide support to women and their partners during this life-changing event.

8.2 INFORMED BY THE LITERATURE; THE IMPORTANCE OF CONTEXT

Despite an ever-growing body of literature focussing on decision-making in relation to antenatal screening for fetal chromosomal anomalies, in particular Down's Syndrome (Reid *et al.* 2009), literature pertaining to decision-making in the antenatal period is sparse in relation to the process undertaken by women and their partners following suspicion or diagnosis of a severe congenital anomaly (Bijma *et al.* 2008, Pryde *et al.* 1993, Shaffer *et al.* 2006).

Findings from this study tentatively suggest a shift in the way screening is perceived by many women that contrasts with the findings of much of the existing literature. Screening appears to be commonly framed around Down's Syndrome. Although knowledge and acceptance of Down's Syndrome appears to have moved on, public perception of the FASP anomalies appears to be lagging behind.

The commonalities shared between this PhD study and the body of literature pertaining to screening of Down's Syndrome include the exploration of maternal decision-making processes (setting), along with the complexities arising from the dynamics of the maternal-fetal relationship when screening for a significant congenital anomaly, in this instance Down's Syndrome (anomaly). Assumptions could be made that findings are transferable between the two. However, despite the significant similarities, the findings of this study suggest that the literature pertaining to decision-making following diagnosis of a severe congenital anomaly, and that relating to the decision-making process in terms of Down's Syndrome screening, diverge.

In terms of the setting, much of the literature relates to the decision-making process pertaining to screening. An informed decision at this point would incorporate discussion or consideration on whether to continue or terminate a pregnancy if an anomaly was identified during screening. However, as discussed in section 2.1, in effect any decision made at this juncture regarding termination, would be made based on a hypothetical scenario (Williams 2005). Evidence suggests that attitudes towards screening and terminations vary considerably

(Markens *et al.* 2010, Hewison *et al.* 2007) and hypothetical and actual decision-making with regards to termination decisions, frequently differ (Erikson 2007, Sawyer *et al.* 2006).

In terms of anomaly, the significant difference between the anomalies included in this study and Down's Syndrome is the severity. Despite the very subjective nature of the word 'severe' Down's Syndrome is not necessarily a life limiting syndrome, unlike the FASP chromosomal anomalies and a number of the structural anomalies. The remaining FASP anomalies are screened for antenatally, in order to identify and plan for post-delivery surgery, and are essentially correctable.

Data collected within this study highlighted the fundamental role that Down's Syndrome appeared to play within the screening and fetal medicine environments. In many ways, this contradicts what was expected based on existing evidence. Literature suggests parents have limited preparation and understanding of the primary role of Down's Syndrome screening (Baillie *et al.* 1997, Garcia *et al.* 2002, Mitchell 2001, Williams 2005, Thomas 2014). However, the evidence gathered within this study suggests that this view has evolved to an extent that decisions made about screening are primarily framed around constructed understandings of Down's Syndrome. Of the women interviewed, all but one made some form of comparison to Down's Syndrome and this provided the framework in which they understood their own diagnosis, something that was often alien to them. It is known that framing techniques can be used to reduce the obscurity of an unknown subject by contextualising the information in a way that enables people to associate it with what they already know (Goffman 1986). In this case, perception of Down's Syndrome became the frame in which other anomalies were encountered. It also became apparent that work undertaken to increase public understanding of screening may have shifted the social framing of the screening process to incorporate Down's Syndrome. The early scan (nuchal translucency calculation) and associated serum tests provide a risk profile for chromosomal anomalies (including Down's Syndrome). Although soft markers noted on the anomaly scan may trigger referral for invasive testing for chromosomal anomalies (including Down's Syndrome), the social reframing of screening is perhaps more

evident in relation to the early scan and serum testing, with the impact on the anomaly scan less noticeable.

One possible explanation for this shift is that considerable research was undertaken about a decade after the introduction of Down's Syndrome screening into mainstream healthcare, with much of the early work completed in the early part of the 2000s. A decade later, we have seen the introduction and amalgamation of FASP with the Down's Syndrome programme (UK National Screening Committee 2009). The prognostic uncertainty, prevalence, and the public perception of Down's Syndrome remain compelling factors to explore, and this is reflected in the ever growing literature pertaining to Down's Syndrome (Thomas 2014). However, the impact and difficulties encountered by women following diagnosis of other congenital anomalies is no less significant. The lack of knowledge and understanding of the FASP anomalies, and subsequent shock at diagnosis in this study, reflect many of the criticisms of the system in the literature relating to Down's Syndrome 15 years ago. Although many parents interviewed had discussed Down's Syndrome screening and made informed decisions to participate, or in some cases decline, few had been aware of the implications of the tests in relation to the FASP anomalies.

Patterns reflecting a socioeconomic divide were noted, with women of high socioeconomic status more likely to suggest that they had prior knowledge of the FASP anomalies (before screening or diagnosis), than those from a low socioeconomic group. This was also demonstrated in the differing reactions of the Consequential (high socioeconomic status) and Choice Removed or Avoid/Delay Groups (low socioeconomic status) following diagnosis, with many of those in the Consequential Group demonstrating an existing awareness of the FASP anomalies. This reflects similar patterns to those presented in the older studies pertaining to Down's Syndrome (Dormandy *et al.* 2005, Khoshnood *et al.* 2004).

Evidence suggests that there are no inequalities in access to screening (Rowe, Garcia 2003). However, as the findings from this study highlight, the variation in knowledge and understanding of the anomalies themselves between women of

differing socioeconomic status resulted in an unequal starting point when the anomaly was identified. The time-lag between diagnosis and decision to terminate in the Choice Removed Group is perhaps one indication of this. Compared to the Consequential Group (all high socioeconomic status), who from diagnosis to termination averaged one week, the Choice Removed Group (all low socioeconomic status) took up to six weeks to decide. This cannot entirely be attributed to differences in uncertainty related to the anomalies themselves, as although there were a greater proportion of chromosomal anomalies in the Consequential Group, there was a spread of anomalies through both groups. What was noticeable was that whilst the Consequential Group suggested that this preliminary decision had already been made, or at least considered, the Choice Removed Group (all low socioeconomic status), along with the Delay/Avoid and Absolute Groups, appeared shocked at the identification, despite having consented to initial screening.

One of the difficulties resulting from this is achieving the balance between providing 'too much' information pertaining to the anomalies and 'not enough'. Information can generate fear, and where risk is low, it is possible this may do more harm than good. This study provides insights into the experiences of the small group of women who were diagnosed with a severe congenital anomaly. For many of them, added information may have accelerated their decision-making, thus removing the need to make additional decisions such as that relating to feticide. Whether there would have been a different outcome had the Delay/Avoid Group been better informed when they entered the process is debatable, as subsequent lack of engagement with clinicians resulted in no active decision being made. Perhaps greater information would have empowered them as a group, thus influencing the subsequent dynamics between clinicians and parents and creating opportunities for active decision-making. However, more realistically perhaps, this would have had little impact other than to create fear. By the nature of the Absolute Group, additional information would have been unlikely to change the course of their decision. Achieving a balance must take into consideration the stress caused to the much larger group of women who undergo screening and are deemed as being at low risk of an anomaly.

8.3 STRENGTHS AND LIMITATIONS

The restrictions of time, resources and scale imposed by the PhD research process is potentially the greatest limitation to this research project. The gap on decision-making processes within the literature is extensive and faced with the same aims again there would likely be a number of different choices and decisions made on how to address this gap. However, many of these would be another way, rather than necessarily a better way.

Reflecting on this, there are two sets of issues that require consideration. The first relates to the way the study was undertaken, and includes the impact of methodological decisions made, and the second is a reflection of the impact of 'me'/myself as a researcher on the study findings.

One of the hardest decisions made was to determine 'the period of interest': at what point is a severe congenital anomaly suspected? At the other end of the time scale, at what point has a decision been made? In many ways these questions were answered pragmatically. Therefore the question; "At what point is it appropriate to approach the parents?" was posed alongside; "What is it feasible to accomplish within the time scale and resources available?" On the first question the PPI group, clinicians and parents, stressed the sensitivity of the timing, and the need to stagger the information provided about the study. The acceptability of approaching parents at the point at which the anomaly was identified was perceived as acceptable (if done delicately) by the parents, with a precedent set within a small number of papers. However, the clinicians were initially more reticent. The inclusion of a clause stating that the initial approach would be made by them appeared to provide sufficient reassurance to the clinicians that they could veto the study if they felt that approaching parents about the study would add to their distress. In addition, the issue of recruiting large numbers of women, only to discover that risk was not converted to diagnosis, was ethically uncomfortable. The initial approach was planned to occur at the time of 'high suspicion', in other words, at the point at which usually the sonographer had

highlighted an abnormality that was likely to reflect a severe anomaly. The parents were then referred to one of the clinicians for further investigation.

Additionally, determining at what point the decision had been made required consideration. The complexity of the decision-making process did not become clear until data collection was underway. In planning the study, I had made a number of assumptions: that the women would make a decision; that the decision would be relatively linear; and that there was only one decision to make (i.e. whether to terminate or continue). However, as I interviewed the clinicians and the women, and collected large volumes of data, doubts arose as to the validity of these initial assumptions. It soon became clear that 'making sense of the decision' was an integral part of the decision-making process: however, time restraints meant that I made the decision to continue with the data available instead of revisiting the possibility of collecting additional data from the women who continued with their pregnancy after the birth. This meant that the data collected from the women who continued did not necessarily reflect how they later made sense of the decision (where one had been made). I had also underestimated the volume of data I would be managing and thus, in terms of managing the study, this decision proved the 'right' one. Nonetheless, lack of follow up data, particularly from those who continued with the pregnancy, has limited the coverage of the findings. If asked what I would do differently, incorporating a further interview with parents who continued, perhaps six to eight weeks after delivery, may have provided a more balanced picture of the experiences of all women. However, following the birth of a baby with a severe anomaly, it is likely that accessing the mothers to interview them again may have considerably increased the time required for data collection, thus making the study unfeasible in the time allowed for a full-time PhD study.

Whilst reviewing the literature available, one of the issues identified was the methodological divide, where the majority of studies used the construction of narratives as their primary data source, for example (Fisher, Lafarge 2015, France *et al.* 2013, Hunt *et al.* 2009, Lafarge *et al.* 2013, McCoyd 2007, Sandelowski, Jones 1996) with a limited use of consultation recordings where conversation analysis

was employed to analyse doctor-patient interactions (Pilnick 2008, Pilnick, Zayts 2012). This study has provided a different approach within this topic area, where consultation recordings and interviews have been combined to offer a 'real time' reflection of the decision-making process. Section 3.3.8 provides a discussion of the decision-making process behind this. In terms of data generation, there are a number of benefits and drawbacks to this approach. The sensitivity of the subject area means that recordings of the consultations are invasive, and development of trust between me and the clinicians over a period of time prior to data collection was essential in order for this to be feasible. The help of the PPI group in determining the acceptability of this approach, and the consent process to support it was indispensable. This process was particularly time consuming, and at times frustrating, in terms of ensuring the PPI group understood the legalities associated with consent (the practical limitations of any proposal) but particularly with regard to the time spent preparing. Around ten weeks were spent observing and shadowing the clinicians, without actively recruiting women. In addition, this preparatory work was only feasible in one trust as I was a single researcher. Although I was able to complete all the clinician interviews during this time, the sense that time was disappearing was very frustrating. However, the patience required was rewarded as the data subsequently collated has provided a unique insight into the parents' interpretation of the clinician-patient interaction, alongside an appreciation of how these interactions have impacted on the decision-making process and subsequent decision.

In hindsight, one of the strengths of my study has been my decision to examine parental decision-making from a naturalistic perspective (see section 2.5). The theoretical underpinning of this perspective highlights the importance of context, the decision-making process over the decision *per se*, while disregarding the concept of a 'rational' decision. Whilst the findings discussed in the preceding chapters have highlighted the importance of context and process, the argument over rationality has raised an interesting dichotomy. Whereas I proposed that decisions within this context could not be deemed 'rational' as this supports the concept of a decision being right or wrong, clinicians sought a 'rational' response from parents (see section 5.3.3), something that parents varied in their ability or

willingness to achieve. Whilst this thesis cannot address the dichotomy seen, it is clear that the search for rationality creates a number of tensions between clinicians and parents.

From a practical perspective there are a number of limitations to this study that must be acknowledged. Although four centres in two trusts (two in each) were involved in the study, the majority (18 out of 20) of cases recruited were identified in one trust, although the parents were referred between the Trusts for speciality care (neurological or cardiac); therefore there was a less pronounced difference in the number of consultations recorded in each centre. The variation in recruitment rate was likely to reflect the significant ground work undertaken in the first trust. In terms of clinicians, recruitment was equally divided between the four centres. The transferability of the findings to a wider context must be considered. The units recruited were all Tertiary Referral Centres; large centres with significant resources and specialised services. Many of the clinics held in smaller DGH's were run by clinicians from these centres, as outreach clinics. Nonetheless, the skills, knowledge and resources available within smaller DGH may vary and the care received may differ if women choose to deliver in a smaller hospital closer to home, with midwives being less used to caring for women who terminate, and private facilities more difficult to access. The locations of the centres recruited were similarly homogeneous; all based in ethnically diverse areas, with deprivation scales around the national average. Both trusts were large and cared for patients in multiple centres. This meant that the facilities available in a single trust could vary depending on to which centre the parents were admitted. This was perhaps of benefit to the study in terms of the transferability of findings.

I was reliant on clinicians' permission in order to access parents, and subsequently for parents to agree to participate. Although no parent who was invited to participate refused, I cannot be sure how many parents clinicians chose not to invite, nor their reasons for this. This was particularly difficult to gauge in the centres where I was not visibly present. However, in the centre that I based myself, suspicion or diagnosis of a severe anomaly quickly became 'general' knowledge amongst the clinical group to which I was privy. I was unaware of any parents at

this site who were not invited whilst I was present in the clinical area. As a single researcher, I was only able to be in one centre at a time. I chose to base myself predominantly in the centre closest to where I study, partly for pragmatic reasons (it was easier to get in and out) but also because they were running a number of additional clinics at the time. This provided a greater throughput of parents, and thus improved my chances of recruiting. However, it is likely that opportunities to recruit parents from other centres were missed, when I was unable to attend concurrent clinics across centres. This is likely to have been a major practical barrier: however, intrinsic barriers relating to clinicians' concerns over the impact on the doctor-patient relationship are also likely to have been influential (Ross-Langley *et al.* 1987). There is some evidence to suggest that clinicians are more likely to approach patients for research when they feel more confident in their relationship (Ross *et al.* 1999), perhaps because introducing research can be perceived as difficult and intrusive, particularly in a sensitive setting (Mason *et al.* 2007). In terms of this study, clinicians may not have met the patient at the time of approach. This is likely to have made approach more difficult. Again, the groundwork undertaken in establishing myself and the study as 'non-threatening' and trustworthy provided some reassurances. Identification of potential participants before the clinicians established a relationship with them is likely to have protected this study to an extent from a clinician led 'selection bias'. It is perhaps questionable whether the opportunity to recruit parents from the Choice Disturbed Group would have been possible had recruitment been reliant on an established patient-doctor relationship. Overcoming the potential barrier of approaching at an early stage in patient care may have inadvertently increased the representativeness of the data collated. Nonetheless, the potential limitation remains; that parents who were not invited (rather than missed) may have represented a group who were excluded by nature of some intrinsic characteristics.

A further group who were excluded from recruitment were those women who did not speak sufficient English to consent and subsequently participate. These were likely to belong to minority ethnic groups. The funding available for this PhD study did not extend to provide sufficient funds for a translator. Although I was able to

recruit a number of women from minority ethnic groups, those recruited did not reflect either the population makeup or prevalence of the anomalies within the wider population. To what extent inclusion of non-English speakers would have changed this is unclear. However, this does indicate that there remains a hidden group whose voice is not represented in these findings. The difficulties in recruiting minority ethnic participants into research is well documented (Yancey *et al.* 2006), as is the importance of recruitment in order to ensure applicability of findings (MacNeill *et al.* 2013). Both ethnicity and religion have been identified as predictor variables for decision-making following diagnosis of a severe congenital anomaly (Ahmed *et al.* 2012, Ahmed *et al.* 2006, Balkan *et al.* 2010, Davies *et al.* 2005), and are likely reflectors of unexplored cultural and contextual characteristics (Okazaki, Sue 1995). Exclusion of this group may have precluded the opportunity to identify specific or differing needs.

The size and heterogeneity of the sample recruited for this PhD study has provided a “*slice from the life world*” (Denzin 1983, pg.134) of a group of parents. The differing meanings attributed to their life worlds is a reflection of the diversity of the sample, and potentially demonstrates how I have attempted to avoid an “*elite bias*” (Miles, Huberman 1984, pg.230) where the most articulate and easily accessed groups become the spokespersons, thus resulting in only part of the whole story being heard. Despite the limitations reflected by the exclusion of women who could not speak English, the diversity of the sample, in terms of demographics and to a lesser extent ethnicity, is a strength and has provided the opportunity for recommendations to be made that will, I hope, improve care.

The second aspect of the study that requires some reflection relates to the way in which I, as a researcher, have engaged with the research. The dual role of nurse and researcher was reflected upon in Chapter 4, where the distinct contribution of the “*nursedness*” qualities have in this instance provided a unique perspective to the experience of these pregnant women (Leslie, McAllister 2002, pg.700).

The experience from this study has been overwhelmingly positive, not only in accessing participants, as reflected in the recruitment rates, but also in managing

situations of particular emotional significance. As I have previously mentioned, I was attracted to the research by both my personal and professional interest in fetal medicine and patient experiences. I spent much time in the centres developing relationships and trust with the healthcare professionals. The boundaries between researching and socialising sometimes became difficult to maintain. As such, I was often party to the clinicians' informal assessments and interpretations of events. I was able to retain some distance by presenting the teams with my professional self. Therefore, although I participated in the baking and tea-making aspects of the centres' social lives, I refrained from divulging too much detail about my personal identity. 'Too much' is difficult to quantify, and difficult to define. I relied heavily on well-honed professional skills that guided me on how much and when to disclose personal aspects to healthcare professionals in order to demonstrate empathy, understanding and authority, and when to remain silent. Developing a good relationship with the healthcare professionals and clinicians in particular, was the pivotal point to the success of this study, and something I believe I achieved particularly well. Skills developed when working in the clinical setting over many years were indispensable. Conversely, I risked becoming overly influenced by the views of the clinicians, to the detriment of the voices of the parents by potentially making assumptions and not asking 'the simple questions' because I had become blind to the routines of the clinical environment. As I had used my nursing background to access the clinical settings, I was unable to hide it. Acknowledging the potential to present a biased perspective served as a constant reminder for me to remain vigilant. The PPI group has been indispensable in providing advice and critique on my interpretation of events, and has provided a credibility and validation function by ensuring I retained as objective a stance as possible (Sandelowski 1986).

In terms of interviewing I was thrust into a steep learning curve. Although I believed I was well prepared, by performing practice interviews with the PPI group, I found the interviews challenging. I don't believe experience alleviated these difficulties either. One of the hardest interviews was with a woman in the Delay/Avoid Group. It became apparent early on in the interview that she did not understand what was happening to her, and the seriousness of her baby's

situation. I found it particularly difficult to focus and was preoccupied by my concerns for her. Whether my instincts as a nurse overtook those of the researcher is perhaps open to debate. In hindsight, I would like to have explored her relationship with the clinicians to a far greater extent and the factors that prevented her from engaging with them. Her relationship with her partner, like that of a number of the women in that group, was complex. Again, I feel that I perhaps missed the opportunity to explore this area in greater detail. These missed opportunities perhaps reflect themes that could have been further explored. Rather than framing this as a limitation, it perhaps reflects the reality of a scoping study such as this, and highlights the need for further research in this area.

8.4 DECISION-MAKING AND THE PARADOX OF CHOICE

Much of the experience of parents following diagnosis or suspicion of a severe congenital anomaly revolves around the subsequent decisions or choices they are faced with. What is apparent from the findings presented within this study is that there is no universal process through which these decisions are made. In addition, a multilevel contextual framework imposes varying restraints or constraints on individuals that gives rise to the enactment of the decision-making process. This section considers a number of the discourses surrounding decision-making and choice, including the discourse on non-directive counselling, issues involved around choice of method, and the fiercely debated area of termination time limits.

8.4.1 *NON-DIRECTIVE COUNSELLING AND INFORMED DECISION-MAKING*

The term 'informed choice' is widely used within the literature pertaining to counselling and clinical decision-making. However, during interviews, the use of the word 'choice' was criticised by parents due to the positive connotation associated with it that infers that positive options are available, when in fact this was not the case. For this reason, the word 'decision' has been used throughout this thesis when referring to enactment of one option over another by the parents. 'Choice' is used when referring to the existing literature, to reflect the term used there.

The spectre of eugenics remains a constant presence within the field of human reproduction. In a conventional definition, the key aspect of eugenics lies in the coercion of people's reproductive choices (Schwennesen, Koch 2012). In an attempt to distance the medical profession from this, the principles of non-directive counselling and informed choice have become an integral part of genetic and antenatal counselling, where non-directiveness is defined as the provision of completely unbiased information and a restraint from giving practical advice (Rehmann-Sutter 2009). The benefit for clinicians of supporting these practices is also perceived at an individual level, where Clarke suggests that this enables them to create an emotional distance, thus protecting the professional from over-involvement, and ensures that professionals will not be held legally accountable for decisions (Clarke 1997).

Literature exploring the application of these ideals within the environment of antenatal decision-making suggests that the reality is far more complex (Williams *et al.* 2002, Henwood *et al.* 2003) with Williams *et al.* stating that there are "*a variety of circumstances when nondirective counselling did not seem to be possible or to be the most appropriate response to the situation*" (Williams *et al.* 2002, p.345). On this basis, Williams and colleagues suggest viewing antenatal counselling as a process that forms a continuum between choice and coercion (Williams *et al.* 2002). Pilnick adds a different dimension as she talks of professionals entering a vicious circle, where, as professionals, they have the knowledge required to make a decision, while by virtue of being a clinician they cannot bring it to bear on decision making (Pilnick 2013).

The discourse over the difficulties encountered when striving for non-directive counselling, or even the attainability of non-directiveness as a principle, is well established within the literature (Williams *et al.* 2002, Pilnick 2008, Bosk 1992). Subsequent studies have given consideration to the manifestation of non-directiveness as a means of care (Schwennesen, Koch 2012, Mol 2008). This study similarly approaches the discourse from a practical angle, providing insight into the relationship between non-directiveness (and conversely directiveness) and the provision of good care (Schwennesen, Koch 2012, Mol 2008). Associated with this,

is the drive towards a shared-decision making model, which is reliant on non-directive counselling, 'informed choice' and 'informed consent' (Charles *et al.* 1999). What this study adds is an insight into how it may be possible to determine when a more directive care and decision-making approach may become appropriate, and the potential risks should attempts at directive counselling be rejected by parents.

Findings from this study highlight a number of tensions that arose for clinicians and parents alike. Despite the evidence suggesting that ideals of non-directiveness, informed choice and subsequent shared-decision making, laid out in policy and guidance, are not possible, clinicians still perceived this as attainable and the essence of good care. In order to enact this, an expectation was placed on parents to engage in an idealised decision-making process. From a theoretical perspective, this is perhaps representative of the relativist stance discussed in section 2.5. As argued, this stance is questionable in terms of its utility in the field of decision-making, with the varying responses of parents to these expectations further indicating the need for a paradigm shift to view decision-making through a naturalistic perspective. Some parents, predominantly those in the Consequential Group, had the ability and aspiration to manage the process in this way, and tensions between clinicians and parents in these cases were minimal. Furthermore, ownership of and responsibility for the decision was subsequently taken by parents in this group. The Assess-Reassess Group embarked on the process, but appeared to lack the skills required for its completion. Clinicians responded by providing more directive counselling. For the majority of parents in this situation (those in the Choice Removed Group) this form of care was accepted and appreciated. Unlike the Consequential Group, they perceived their involvement as consenting to a recommended option, with responsibility for the decision being externalised onto the clinician. However, for a small minority (Choice Disturbed), a breakdown in the relationship with the clinician resulted in withdrawal from the process, and the parents appeared to isolate themselves. This did not appear necessarily to be as a result of a rejection of a more directive approach *per se*; rather, a seemingly small inconsistency or misunderstanding resulted in loss of trust between parent and clinician. This in turn precluded

acceptance of any intervention from the clinicians. Reinforcement of information through a second opinion was viewed as a conspiracy, rather than collaboration, as the relationship deteriorated. Disengagement from the clinical team meant that termination became a non-option. Potentially, early intervention by a mediator may have helped prevent further deterioration of the relationship. However, this would require clinicians to identify the breakdown in communication early, as time to resolve issues is limited. What was noted in some of these consultations was that additional senior team members were brought in to the counselling sessions. Although the rationale for this is not clear, it may have been a strategic attempt to protect clinicians from scrutiny or litigation in the future. However, from the parents' perspective this could be construed as a further attempt to pressure or intimidate them. The difficulties faced on both sides are clear, and no simple solution is apparent. Quick resolution of the miscommunication is likely to be the only feasible mechanism for managing the situation. However, with the added emotional aspect of the circumstances, that is far easier said than done. Perhaps the role of the midwives could be explored in terms of acting as mediators at this point.

For a third group, the Avoid/Delay Group, a poor clinician/patient relationship and lack of engagement with the process from the outset resulted in clinicians stepping back and disassociating themselves. Fear of litigation or perhaps avoidance of potential confrontation resulted in a passive response from the clinicians, where non-directive information was passed onto the parents in a one-way transaction. Lack of desire and perhaps ability to engage resulted in a potential limit on options available to the parents. The final group, the Absolute Group, provided an alternative perspective. They entered the consultation with an outcome in mind; the influence of their moral or religious beliefs meant that they were not prepared to consider terminating the pregnancy. The constraint was applied to the options prior to engagement with clinicians. Clinicians appeared to find this difficult to manage, and responded by frequently repeating the offer of termination. This in turn caused distress to the parents.

In their quest for the enactment of an ideal decision-making process, clinicians anticipated that, first, parents had (or should have) the desire to engage in active decision-making, and second, they had the ability to do so. Both these suppositions have been questioned in the literature. For example, a study of the general population, designed to explore participant preference for active or passive decision-making, concluded that a significant proportion of patients did not wish to engage in active decision-making (Levinson *et al.* 2005). Whereas 96% of the respondents wanted to be offered choices and be involved, over half preferred to leave the final decision to the clinician and over 40% preferred to rely on clinicians for medical knowledge rather than seeking out information themselves. These findings are also borne out within the antenatal setting, with evidence suggesting that women hold a variety of views on the nature of informed choice, and that, contrary to policies of autonomous informed choice, many women seek and value the advice of health professionals (Ahmed *et al.* 2012, Marteau *et al.* 1994). This has been further explored in relation to socioeconomic status, where low socioeconomic status has been associated with a preference to delegate authority and responsibility of decision-making to clinicians (Arora, McHorney 2000).

The second assumption is reliant on the ability of parents to engage with the decision-making process. Engagement places demands on a patient's literacy and numeracy skills, in order for complex health information to be understood (Smith *et al.* 2009). Whilst health literacy has been defined as the “*degree to which individuals have the capacity to obtain, process, and understand basic health information and services needed to make appropriate health decisions*” (Institute of Medicine 2004), some experts argue that this definition is too narrow and should extend to the ability to interact with a health professional (Nutbeam 2000), as well as awareness of public health issues, scientific processes and cultural differences (Zarcadoolas *et al.* 2005). An association between socioeconomic status and health literacy has been acknowledged, thus raising concerns that patients from lower socioeconomic groups may have difficulties in participating in the process (Smith *et al.* 2009, Perzynski *et al.* 2013).

Trust between parent and clinician was paramount, and appeared to be the cornerstone of the decision-making process. For some parents the decision-making process was uncomplicated and they possessed the resources to navigate the complexities of the decision, with the support of the clinicians. For many, this was not the case and the parents sought 'care' from the clinicians by means of the provision of some direction. Due to external legal and professional pressures, where engagement between parents and clinicians was poor, clinicians avoided a more directive approach, thus leaving parents without the support they potentially needed in order to make an informed decision (Beck-Gernsheim 1995). For others again, directive care was dismissed as the parents believed there was no decision to make. This resulted in tensions and a perceived lack of support from clinicians. Finally a small minority disengaged with the process once trust had been lost. For these parents a more directive approach was perceived as eugenic.

This mix of reactions leads to the question of how clinicians should behave. It is clear from these findings that the drive against directive counselling is not only unrealistic but potentially detrimental to clinicians and parents. However, trust is required on both sides before a more directive approach can be successfully implemented. In addition, identifying the small group of parents who have essentially no decision to make (the Absolute Group) is necessary to avoid adding to the parents' distress. There is a particularly fine line between offering choice and applying pressure. Identifying the attributes of the parents in terms of the model proposed may perhaps provide some guidance for clinicians.

8.4.2 *RESPONSIBILITY AND REGRET*

The literature pertaining to the psychological impact of a diagnosis of a severe congenital anomaly generally approaches the topic from either the perspective of the women that terminate or those that continue, or a comparison between the two. This suggests that the outcome of the pregnancy is perceived as influential on grief and regret. Loss following diagnosis of a congenital anomaly is well documented as a traumatic life event with potentially significant and lasting emotional impact (Davies *et al.* 2005, Fisher, Lafarge 2015, Hunfeld *et al.* 1993, Iles 1989, Iles, Gath 1993, Korenromp *et al.* 2007, Korenromp *et al.* 2009, Lafarge *et al.*

2013, Lalor *et al.* 2009, McCoyd 2007, McCoyd 2003, Statham *et al.* 1999, Statham *et al.* 2001, Salvesen *et al.* 1997, Wool 2011, Zolese, Blacker 1992, Williams *et al.* 2008).

This study was not designed to explore the long-term psychological impact of the decision on the parents. However, placing the findings from this study within the context of literature correlating levels of grief and regret with parental characteristics as well as decision-making attributes, highlights the potential vulnerability of women in some of the decision-making categories to higher than anticipated levels of distress, and suggests that the outcome of the pregnancy has less impact on grief and regret than the process through which the decision-making was enacted.

As discussed in section 2.3, parental characteristics, including low socioeconomic status (Statham *et al.* 2001), young age (Zolese, Blacker 1992, Statham *et al.* 2000), strong religious beliefs and advanced gestational age (Davies *et al.* 2005, Korenromp *et al.* 2007) have been shown to correlate with a high likelihood of a severe psychological response, particularly when the decision to terminate an affected pregnancy was made. Whilst active decision making appears to protect from high levels of regret and grief (Smith *et al.* 2009), conversely high levels of doubt during decision-making (Korenromp *et al.* 2007) and women who internalised responsibility for decision-making were hypothesised to be at greater psychological risk (Sandelowski, Jones 1996).

Applying these findings to this study, the women in the Consequential Group would appear the least likely to encounter complications of grief and regret. As a group, they were marginally older than the other women, were all of high socioeconomic status and were active in the decision making process. However, they did internalise responsibility for the decision. For many in this group, internalising the responsibility in this way was perceived to be an effective mechanism to alleviate future regret. However, these women had the ability and desire to process the information available, and thus arrive at an informed decision. Conversely, based on the available evidence, the combination of

socioeconomic status and the decision to terminate would place women in the Choice Removed Group at high risk of a complicated grief reaction. Nonetheless, this group frequently externalised responsibility for the decision onto the clinicians, which hypothetically would reduce the psychological risk (Sandelowski, Jones 1996). Their narratives further distanced themselves from the decision by suggesting that there was no decision to make due to the severity of the anomaly. The apparent contradiction between the two sets of findings highlights a tension between the longer and shorter term responses to a more directive counselling style, and raises the issue of the impact of the way in which the counselling was enacted. Great importance was placed by the Choice Removed Group on confirming that the decision had been 'right' and based on correct information. This meant they either needed the reassurance of seeing the anomaly after birth, or to receive the post-mortem results. Whereas a visible anomaly provided some reassurance, 'invisible' structural anomalies resulted in these women questioning the decision. Delays in accessing post-mortem results resulted in a heightened sense of anxiety. For this group, closure was not achieved through the 'taking of the decision' as happened with the Consequential Group. Instead they required additional reassurance after the birth, thus enabling them to 'move on'.

There were also apparent differences in the pathway of care and resources available to women who continued and those who terminated the affected pregnancy. Whereas processes appeared embedded in the service to support those who continued, access to the same services were more haphazard following a termination. This held true at healthcare and local organisational level facilities, but also wider societal services. Access to regular counselling and bereavement support, alongside legal rights to time off and maternity payments, differed between the groups, leading to some women who terminated suggesting their grief was a private rather than a shared experience. It is difficult to envisage a solution to these sentiments. Formal after-care pathways for those who terminate may ensure availability and equality in accessing counselling services as well as more tangible help in the form of financial contributions towards headstones or memorials. Many of these 'extras' are available through charitable funds, but the stigma attached to termination perhaps creates difficulties in the approaching and

requesting of help as well as the desire of some charities to provide assistance. There are groups across the UK who offer guidance and support to women and their partners following termination of a pregnancy. However, public support is often limited and raising funds difficult as termination remains an intensely emotive and highly stigmatised issue (Fisher 2013). Perhaps one option is to engage the public with the real stories of the desperate decisions made by these parents, in an attempt to overcome some of the stereotypical characteristics engendered by society.

The psychological impact on the women who continued in the Absolute, Choice Disturbed and Delay/Avoidance Groups are more difficult to evaluate as data post-delivery were not collated. The indescribable grief following any neonatal or infant death is well documented within the literature (Cacciatore 2013, Campbell-Jackson *et al.* 2014, Moore 2013, Sturrock, Louw 2013, O'Leary, Warland 2013, Boyle *et al.* 1996), although the impact of a diagnosis of a congenital anomaly is unclear. As discussed, the decision-making process engaged in by women who terminated their affected pregnancies impacted on their subsequent experience. Similarly those who continued with the affected pregnancy are likely to have varied experiences depending on the decision-making process in which they engaged. Whilst the Absolute Group actively made a decision to continue, neither the Choice Disturbed nor the Delay/Avoid Group had reached a sufficient level of acceptance or understanding to make an active decision. Where the resolution of grief has been associated with an increasing acceptance of loss (Prigerson, Maciejewski 2008), the differing impact of the decision-making process on the psychological recovery of the parents is likely to be visible between the groups, where the Choice Disturbed and Delay/Avoid Groups are theoretically at higher risk of a complicated grief reaction at the time of delivery and the subsequent loss of their baby. In order to meet the differing needs of these women and their partners, a greater understanding of the impact of the pregnancy is required.

8.4.3 *DECISION-MAKING AND METHOD OF TERMINATION*

The journey from suspicion of an anomaly to termination involves numerous points at which decisions must be made, from whether to undergo screening,

consent to further invasive testing, whether to terminate or continue the pregnancy, and whether to have a feticide or not. However, for those who do decide to terminate, one practical decision appears to be lacking: method of termination. Whilst changes in policy have the potential to improve care outcomes for women terminating an affected pregnancy, other national policies have complicated and further restricted choice for women. In this instance, one such political initiative has been the transfer of commissioning of termination services from the public (NHS) sector, to the private sector, namely the British Pregnancy Advisory Service (BPAS) or Marie Stopes. This is now commonplace, with 93% of the women undergoing a termination in the private sector accessing this through NHS commissioning (BPAS 2014).

Within the private sector, the majority (around 80%) of terminations are performed surgically (Fisher 2013). However, a large disparity is visible compared to the NHS sector, where fewer than 20% are performed surgically (Lysus 2014).

It is clear from the evidence available that there are no clinical reasons why medical terminations should be preferred over surgical. Although the literature available pertains specifically to Section C terminations, the clinical process and complications of the procedure will not differ for those performed under Section E. Complication rates following surgical terminations are comparable to medical methods, with randomised controlled trials showing lower complication rates, cost and time requirements, along with improved psychological outcomes for the women undergoing the surgical process in the first and second trimesters (Kelly *et al.* 2010, Bryant *et al.* 2014). In addition, a number of randomised controlled trials conducted with women undergoing Section C terminations have concluded that surgical terminations are more acceptable to women than medical inductions, with one study unable to proceed as insufficient numbers of women were prepared to be randomised to the medical arm of the trial (Grimes *et al.* 2004, Kelly *et al.* 2010). No literature pertaining specifically to women undergoing termination under Section E was identified, and generalising the findings relating to the psychological impact must be made with caution. However, at comparable gestations, over 75% of Section C terminations are performed surgically,

compared to 16% of terminations for a congenital anomaly (Lysus *et al.* 2013). This large variation is likely to reflect choice, or lack thereof, where, following diagnosis of a congenital anomaly, parents tend to remain in the care of the NHS hospital (Thomas *et al.* 2003), whilst those terminating under Section C are referred to a private clinic. This raises the question of whether the lack of choice pertaining to method is a reflection of something intrinsic to the NHS or to the nature of the congenital anomaly. In terms of the anomaly, the main clinical benefit of a medical termination over a surgical procedure is the ability to perform a post-mortem. However, where a chromosomal anomaly has been identified, no post-mortem would be required, as the diagnosis would have been obtained through invasive chromosomal testing. Therefore, irrespective of rationale for the lack of choice, it can be argued that the decision should be made by the parent, not assumed by the clinician.

The lack of choice is likely to be as a result of the policy changes that have directed termination procedures into the private setting. In essence, removal of a large volume of procedures from within the NHS has resulted in a deskilling of clinicians (Fisher 2013). Trainees are no longer seeing many surgical terminations, and are arriving as consultants without the competence to perform these procedures. Therefore, the option for a surgical termination has gradually disappeared, with few centres offering such terminations after 12 weeks (Fisher 2013). Within this study, none of the women were offered the option of a surgical termination.

Although the post-termination narratives of the parents reflect positively on the legitimising of the fetus through childbirth, it is unclear whether this would have remained the case had the option for a surgical termination been made available. Lack of choice of termination method for women following diagnosis of a severe anomaly is a national issue. Policy has driven changes that have restricted the choices available to this group of parents. It is unlikely that the policy direction will change: better working between the NHS and private sectors will be required to ensure that the current variations do not remain as a result of a systematic failure to provide best care to all. This can potentially be achieved through a

rigorous examination and planning of clinical training as well as ensuring access of all women to the private services available, if wanted.

8.4.4 *DECISION-MAKING AND THE LAW*

The emotive nature and subsequent intense interest in termination practices by large groups of the public has resulted in a number of high profile criminal prosecutions being brought against clinicians. As a result there is a growing fear of scrutiny, particularly in relation to late terminations (after 20 weeks), and consequently a risk that the rights versus morality debate championed by pro-choice and pro-life social movements will inadvertently shift, thus restricting the choices available to some groups of women.

In order to protect women's choice, three options are available: first, continue as present; second, clarify and strengthen the law; or third, remove all limits and create a system where termination is no longer a legal but a medical process. In order to explore these potential options, examples have been sought from the international context that may provide some insight into the impact of each option on parental 'choice'.

Maintaining the status quo would in many ways be the easiest option. Termination is a highly emotive issue with the strength of feeling acutely apparent in the power and fear generated by the pro-choice and pro-life factions. Lobbying by both sides undoubtedly influences the formulation of new laws and legislation. Self-preservation would perhaps lead politicians to avoid this debate. Historically, concerns have been raised repeatedly by clinicians over the lack of clarity of definitions of terms such as 'severe' or 'significant' (Lilford, Thornton 1993) and, along with the impact of scrutiny, these difficulties were widely discussed by the clinicians interviewed. Processes including the collation of data on terminations undertaken after 24 weeks added to these concerns. Many talked openly about the impact of recent high profile cases, suggesting that their practice had subsequently become more defensive. Although informal care pathways have been developed through the offer of referral for second opinion to more 'termination-minded' centres, this corporate approach is likely to result in variations across

geographical areas, with the impact most noticeable in areas of low socioeconomic status where the cost, emotionally and financially, will restrict access to terminations in certain circumstances. Without clarification, it remains the role of the clinician to enact the law and, although in theory, this should enable a case-by-case application of the law, findings from this study suggest a more complex situation where the lack of clarity in fact reduces 'choice' through the practice of defensive medicine.

The second option is to strengthen and clarify the law as has occurred in the US. Attitudes to termination are generally more restrictive there than those in England. As a result of lobbying by the pro-life movement, state-wide legislation has been introduced to support and tighten the existing laws. These changes have resulted in a significant drop in the number of terminations performed (New 2014). Although the legislation did not alter the termination law as such, it impacted on the way that clinicians practise within the law. For example through legislating for the tightening of 'informed consent' procedures (New 2014), state legislation in the US has essentially been devised to control clinical practice (Bitler, Zavodny 2001). Whilst this has potentially provided some legal protection to clinicians through clearer direction on what is and is not permissible, the reduction in the number of terminations performed overall (New 2014) suggests considerable restriction on the options open to women.

The third option relates to the complete overhaul of the law, thus removing all limits and decriminalising the act of termination. This model has been applied in other countries, for instance Canada. In 1988, the Supreme Court of Canada removed the legal limit in its entirety for all women (Rahman *et al.* 1998). This was based on the right to freedom where; "*The decision whether or not to terminate a pregnancy is essentially a moral decision and in a free and democratic society, the conscience of the individual must be paramount to that of the state*" (Tatalovich 1997 pg.77). Arguments that this would result in an increase in the number of late terminations appear to have been disproved (Cook *et al.* 2014).

In theory, this directive would remove termination from the political agenda, placing it into the healthcare context. Professional guidelines provide the framework in which terminations can be accessed and clinicians make a case-by-case judgement, based on these guidelines, without fear of criminalising either themselves or the woman. However, literature from Canada suggests that the debate is far from over. For instance, one of the primary drugs used to induce a medical termination has not been approved in Canada (Glendon 1988). The rationale for this is unclear, and there has appeared to be some avoidance by politicians to address this issue as highlighted in the minutes from a meeting of the Health Committee (Parliament of Canada 2013). Alternatives can be used, but these take longer to act (Pymar, Creinin 2000) thus increasing pain and subsequent distress for the woman. In addition, funding for terminations is only granted where the procedure is carried out in a large state hospital. Smaller private hospitals or clinics are not funded (Glendon 1988). This is likely to result in variations in access to termination services, particularly in light of the size and geography of the country. These factors highlight the powerful influence of policy and broader social attitudes, and how they come to bear on the enactment of the legal system itself (Johnston 2012).

Greater consideration to the issue of 'stigma' will be given later in this chapter, but the interaction between decriminalisation and stigma deserves some consideration here. No literature was identified on the impact of decriminalising termination in Canada and subsequent attitudes and stigma. However, in similarly stigmatised areas such as prostitution and obesity, a number of articles highlighted powerful associations. The concept of "structural stigma" is used in the context of sex workers, where law defines attitudes (Bruckert, Hannem 2013 pg.299). Further, in the case of obesity legislation, in areas where the law has been used to enforce anti-obesity policy stigma attached to obesity is significantly higher than in areas where it has not (Yeh 2012). The law reflects and directs attitudes and public perception. An overhaul of the termination law may make stigmatisation less likely, by medicalising rather than criminalising the procedure. As Clare Murphy one of the directors of BPAS stated; "*no woman in any part of the*

world should have to live in fear because of the way other people have legislated over her body.” (Murphy 2014)

8.5 THE NEED FOR AN IDENTITY

‘Identity’ emerged as a prominent theme throughout the study. A number of common identities were portrayed by the parents within the sample and perhaps reflected how participants sought to make sense of their situation. Whilst the participants universally sought to humanise the fetus, enactment of this need was simultaneously supported and constrained within the different layers of the contextual framework surrounding the decision-making process. Assigning the identity of a baby to the ‘unborn’ appeared to offer parents some comfort and provided a rite of passage to engage in the social rituals of grief. However, by assigning this identity, those who terminated the affected pregnancy faced the stigma arising from a “*spoiled identity*” (Goffman 1963 pg.130). Findings from this study suggest that this was experienced and managed in different ways, and was often expressed through the decision to disclose or not to disclose their actions to others. Alongside their pre-existing views on termination, this appeared to be an indicator of subsequent feelings of regret and blame, thus adding to the psychological impact of the process.

For those who continued the affected pregnancy, the identities of ‘parent’ and ‘baby’ were equally strongly enacted and similarly experienced in different ways. For some, the identity of an ‘imperfect’ baby was never fully accepted. The paradox of hope created by uncertainty, and further confounded by a degree of misunderstanding, contributed to the difficulties encountered in accepting the identity.

Universally, the identity of mother and protector of the unborn was paramount, although this could be enacted either through continuing the pregnancy and giving the baby every chance at life, or terminating the pregnancy to ensure the baby did not suffer. This became their first decision as a parent.

The following section extends the discussion on identities and explores the ways in which they are constructed or negotiated within the contextual framework surrounding the decision-making process.

8.5.1 *CREATING A SOCIAL IDENTITY*

Social identity has been defined as “*that part of an individual’s self-concept which derives from his (sic) knowledge of his (sic) membership of a social group (or groups) together with the value and emotional significance attached to that membership*” (Tajfel 1972 pg.292). Therefore, an identity arises from the distinguishing and labelling of a set of characteristics to which an individual claims membership.

The terms ‘parent’ and ‘baby’ have intentionally been used throughout this thesis as they reflect the language adopted by the participants. The use of terminology in this way highlighted the universal desire to assume these identities, irrespective of the decision to continue or terminate the affected pregnancy.

Within the different contextual layers, mechanisms of constraint or enablement towards the attainment of these identities were observed. The use of technology, for example, reinforced the identity of the fetus as one of a baby through visualisation and subsequent humanisation. Conversely, the law creates an impenetrable divide between fetal and baby status. This divide is reflected in what is provided in terms of support such as maternity leave and benefits.

Since the invention of ultrasound in 1956, the way in which women experience pregnancy has changed dramatically (Sedgmen *et al.* 2006). This is due in part to the increasing drive to medicalise pregnancy and childbirth, but also down to the changing status of the fetus (Lupton 2013). Ultrasound has provided the tools to create a visible entity, and as such contributed to its humanisation before birth (Lupton 2012). New 3D and 4D imaging techniques that serve little medical purpose have enabled parents to meet and bond with their baby long before birth. Thus women have become custodians of the fetus (Lupton 2012) that has gained a social presence that previously did not exist until birth. Despite the spectrum of reactions expressed by participants in relation to their understanding of the

primary function of the ultrasound scan within the antenatal setting, the social nature of the scan was appreciated by all, with many of the parents retaining scan pictures as evidence of their subsequent loss and as evidence of their parenthood identity. Their choice of terminology further contributed to this changing status. Use of key terms such as 'baby' versus 'fetus', and 'mother' versus 'woman' imply certain expectations, whilst further humanising the fetus (Evans, O'Brien 2014).

The identities of 'parent' and 'baby' are routinely reinforced within the healthcare setting. Primarily this occurs through the shared location of antenatal, screening and fetal medicine clinics. In addition, routine practices are enacted within the setting that further emphasise these identities. For example, the use of Bounty bags¹⁷ was highlighted by a number of parents as a mechanism which reinforced the social construct of a 'perfect' mother and baby identity. For some parents, albeit a small percentage, their reality will differ following identification of a congenital anomaly. Allowing these social expectations to proliferate within the antenatal setting reinforces the social perceptions of the 'routine nature' of antenatal screening, and the 'deviance' in cases where anomalies are identified. For the parents affected, this further added to their distress (Chitty *et al.* 1996) and subsequently contributed to the stigma associated with termination and disability.

Within the legal context, the humanisation of the fetus is restricted by laws governing the demarcation of the gestation at which a fetus has 'rights'. Current law defines this artificial divide at 24 weeks, unless the fetus shows any signs of life at birth. For those terminating a pregnancy after 22 weeks' gestation, guidelines recommending the use of feticide reduce the chance of a live birth, thus ensuring that the fetal status is retained by law.

¹⁷ Bounty bags (bags of pregnancy related goodies) are distributed in antenatal clinics. They contain information on maternity benefits, pregnancy health, as well as a selection of product samples and are financed through advertising.

Although terminations performed under Section E should not be affected by the 24 week legal cut-off, findings from this study suggest that the lack of clarity of the law and subsequent defensive practices may impact on parents' ability to access a termination after 24 weeks. This may result in some parents being unable to access social rituals associated with loss of a baby. These include registering the birth and death, and funeral. A 'birth certificate' has been created by SANDS¹⁸ and can be provided to parents in many hospitals, irrespective of the gestation. This can be retained as a memento although it has no legal status. However, the need for such a document suggests a widespread desire to humanise the fetus. Many of the participants showed me their memory boxes, which contained the certificate along with pictures, foot or hand prints, and other mementos of their baby.

When allocating resources, there is a need to create boundaries that clearly define who is entitled to support. Organisation of the English system has a consistent 24 week limit applied to the legal and benefit systems. This means that if the baby is born after 24 weeks (whether live or stillborn) women can access maternity benefits in terms of funding and time off as it has gained 'human' status as defined by the law. This raises two issues: the first pertains to the impact of removing the legal definition as discussed in section 8.4.4, and the second to the care available to those women whose baby was delivered before 24 weeks. As discussed, decriminalising the legal system would not necessarily impact on the time limits set by the benefit system. However, conversely, reduction of the benefit limit would likely provide additional ammunition to pro-life groups who could argue that provision of benefits at an earlier gestation is indicative of a change in fetal identity and viability to that of 'baby' at the earlier gestation. Any change in benefit limits could impact on legal limits.

¹⁸ SANDS stands for Stillbirth and Neonatal Death Charity, and offers support to parents following the loss of their baby.

Perhaps the solution lies in ensuring support is available in other forms for those women who have terminated a pregnancy for a severe congenital anomaly. This study has highlighted a desperate need; however, consideration of the risk of increasing stigmatisation of women undergoing termination under Section C must be given. By differentiating between the rationale for terminating a pregnancy, a 'them' and 'us' is reinforced. By decriminalising termination in the way described above, the need for different sections is perhaps overcome, thus going some way to breaking down the barriers. By managing the over-riding cause of stigmatisation of termination, and the problems inherent with it, the desire to separate themselves from those terminating under Section C, as demonstrated by some parents, might no longer be relevant. There is clearly no simple solution. However, as a society it is a debate that needs to be heard.

8.5.2 *PARENTAL IDENTITY AND STIGMA*

As the identity and status of a baby is endowed on the fetus, the identity of a parent emerges. This identity comes with perceived responsibilities, including that of protector of the fetus or baby (Lupton 2011). Acceptance of the baby and parent identities risks reinforcing public beliefs and attitudes towards termination, and potentially feeds into the stigma associated with it (Evans, O'Brien 2014).

Stigmatisation can occur where negative stereotypes are assigned to either the identity as a whole or certain characteristics associated with it, resulting in discrediting of the person holding that identity (Norris *et al.* 2011). The role of the mother as protector of the unborn is strongly embedded in our social and cultural context (Lupton 2011). Termination violates this norm, and those who take on this "spoilt identity" (Goffman 1963 pg.130) are at risk of encountering stigmatisation in some form. In turn, this can result in feelings of shame and guilt, thus complicating the grief reactions (Bleek 1981, Lithur 2004).

There are a number of ways of categorising social stigma, including the 'external', 'deviation in trait' and 'tribal' stigmas defined by Goffman in his seminal work (Goffman 1963). As a hidden stigma, in other words not external, the use of this categorisation within this scenario does not provide any added value. More

recently stigma has been conceptualised from a different perspective and three domains identified, namely; perceived stigma (extent to which the individual believes others will devalue them), experienced stigma (actual experiences of discrimination), and internalised stigma (incorporating negative perceptions to themselves) (Link *et al.* 1997). The benefit of breaking it down in this way is that it enables a more nuanced analysis of the impact of specific aspects of stigma on the parents that perhaps goes some way to explain some of the differences observed between the decision-making groups. Applied to termination, the three domains represent: the way in which women feel others will react, thus influencing their decision to disclose or not; how others react; and the potential manifestation of feelings of guilt or shame (Cook *et al.* 2014).

‘Abortion stigma’ is generally poorly understood, with little research available to indicate what negative consequences it may have on women’s lives (Kumar *et al.* 2009). During the planning stage, initial reticence from many people (friends and healthcare professionals alike) about my engaging with and interviewing women who terminated their affected pregnancies illustrated how ‘these women’ are labelled and stereotypes created around the labels. Amongst the participants themselves, the different responses to stigma highlighted the lack of homogeneity in its impact. Notably, the women in the Consequential Group appeared less affected by stigma at any level or domain. Although they acknowledged the emotiveness of undergoing a termination, support from their social network was generally strong, and many in this group discussed disclosure of details about the termination to family, friends and colleagues. Termination is a concealable stigmatised identity, thus disclosure crosses the boundary “*between the safe confines of concealment and the vulnerability of visibility*” (Chaudoir, Fisher 2010 pg. 22). The decision to disclose or not provides some insight into the parents’ perception, experience and particularly internalisation of stigma (Cook *et al.* 2014). Although the benefits of disclosure are widely discussed in the literature, including individual psychological (Major, Gramzow 1999, Zea *et al.* 2005), behavioural (Broman-Fulks *et al.* 2007, Quinn *et al.* 2004), and health benefits (Ullrich *et al.* 2003), removing the protection of their concealed identity risked the experiencing of negative outcomes such as rejection and discrimination (Chaudoir,

Fisher 2010), which in turn has been linked to social isolation and lack of social support (Quinn, Earnshaw 2011, Shellenberg *et al.* 2011, Lepore *et al.* 1996, Major, Gramzow 1999).

Conversely, many of the women in the Choice Removed Group did not disclose or only partially disclosed. Due to the gestation at termination, their pregnancy status would have been visible. Saying nothing would therefore not have been an option. Instead phrases such as “we lost the baby”, or “the baby died” were employed to describe the termination. A number of these parents attached negative stereotypes to termination in general, and in particular to the identity of women who ‘chose’ to terminate their pregnancy for non-medical reasons. Thus, following the termination of their affected pregnancy, they gained an identity of which they heavily disapproved (Quinn, Earnshaw 2011). Two mechanisms for managing the resulting emotions were employed. First, they typically distanced themselves from these others by applying more stringent labels including ‘unwanted’ pregnancy versus their ‘wanted’ pregnancy, or ‘choice’ versus ‘decision’. In addition, they externalised responsibility for the decision onto the clinicians, thus rejecting the identity of ‘decision-maker’ and relieving themselves of some of the burden of guilt (McCoyd 2007). Also, they typically internalised the stigma, thus generating further negative emotions such as shame, regret and guilt (Bleek 1981, Lithur 2004). The quandary between disclosure and the risk of stigmatisation was widely expressed within this group, with the added burden of guilt for ‘lying’ to friends and family about the decision.

Of those parents interviewed who reported having experienced stigmatisation, the episode recalled most frequently arose from healthcare professionals encountered during delivery. This included forced disclosure in order to access the delivery suite, as well as the apparent judgment of midwives and clinicians who cared for them during the procedure. During these interactions with healthcare professionals parents will be at their most vulnerable, not only due to the emotiveness of the procedure that they are undergoing, but also because their ‘concealed identity’ was laid bare for all to see. Experience of stigmatisation in this way may result in fear of further discrimination and thus lead to future avoidance

of disclosure (Wolitski *et al.* 1998). This in turn has been linked to social isolation and lack of social support (Quinn, Earnshaw 2011, Shellenberg *et al.* 2011, Lepore *et al.* 1996, Major, Gramzow 1999, Pennebaker 2000). Subsequently, women who benefit from social networks and support may experience less grief and anxiety than those who were unsupported by their communities or wider environment (Goodwin, Ogden 2007). Conversely, concealing abortion becomes part of a vicious cycle that reinforces the perpetuation of stigma (Kumar *et al.* 2009).

Interactions between healthcare professionals and parents they care for may have a much greater impact on the long-term psychological wellbeing of the parents than is frequently perceived. The need for specialist counselling training for all involved in the care of this group of women is paramount. This is revisited in section 8.7.

8.5.3 *IDENTITY, STIGMA AND GRIEF*

Although the issue of grief was explored in section 7.4 in relation to the existing literature and the characteristics revealed by the participants, the unique impact of 'abortion stigma' on the grief processes of the parents is deserving of some examination. The combination of stigma and the constraints applied by law perhaps reinforces some parents' beliefs that society is either overtly judgemental or negates the impact that a termination can have on a woman (Goodwin, Ogden 2007).

However, the consequence for some of those who terminated was a "*disenfranchised grief*", a grief that "*persons experience when they incur a loss that is not or cannot be openly acknowledged, socially sanctioned or publicly mourned*" (Doka 1989 pg. 4). Examples of disenfranchised grief include death following drug overdose or suicide, as well as fetal loss through termination, stillbirth or miscarriage (Feigelman *et al.* 2009). Lack of social validation of the loss frequently translates into a lack of support systems, traditions, or institutions (Feigelman *et al.* 2011); in this instance compassionate leave, social rituals such as funerals, or registration of the birth and death. Added to this are assumptions that the women have made a choice and therefore their grief must be less (Norris *et al.* 2011, Chitty

et al. 1996), despite little empirical evidence to support this (Davies *et al.* 2005). In addition, parents referred to termination as a private grief not shared by others. Subsequently humanising the fetus to take on the identity of a baby enables the loss and grief to be shared.

Paradoxically, humanising the fetus and seeking a parental identity increases the risk of stigma; a technique frequently employed by the pro-life movement to discourage termination, and which contributes significantly to the feeling of guilt and regret associated with the termination process (Goodwin, Ogden 2007). Contrary to the findings from this PhD study, evidence pertaining to the antenatal detection of Down's Syndrome suggests that interactions between healthcare professionals and parents during consultations seek to de-humanise the fetus once high risk status has been identified (Thomas 2014). One possible explanation for this apparent inconsistency may be the gestation at identification. Where Down's Syndrome risk is calculated at the nuchal translucency scan (between 11 and 14 weeks), the majority of the structural anomalies were identified at the 18 to 21 week anomaly scan. This contradiction may simply reflect the difference in gestation of the fetuses. Literature linking the decision to terminate with specific variables has demonstrated the relationship between early gestation and likelihood to terminate (Marteau *et al.* 2002, Jeon *et al.* 2012). Furthermore, literature examining coping mechanisms post termination for a congenital anomaly has highlighted the need for parents to acknowledge and provide the baby with an identity (Lafarge *et al.* 2013).

The need for a specialised bereavement care pathway or package for the parents terminating a pregnancy following diagnosis of a severe congenital anomaly is pressing. Research indicates that, several years post termination following diagnosis of a severe congenital anomaly, women continue to display symptoms of grief (Green, Statham 2007). The impact of stigmatisation, although not universally experienced in the same way by all parents, can increase the risk of bereavement difficulties. Subsequent failure to access support within their community further adds to their suffering. Identification of those who feel unable to disclose to friends

or families may be a useful indicator to identify those at greatest risk of complicated grief reactions.

8.5.4 *IDENTITY AND FATHERS*

Fifteen men were interviewed for this study alongside their wives or partners. Although I met the majority of these in the fetal medicine unit during their consultations, most expressed surprise when invited to be interviewed, citing a perception that the clinical process revolved around the woman, not them. This perception is supported by the legal position in England, where the woman has the ultimate legal right to make the decision. The need to maintain this legal position is not disputed. However, this stance does create a tension, where the importance of the immediate social context in which the decision is made is not supported. This section explores the impact of prioritising the maternal over paternal or couple identities and seeks to identify mechanisms through which one can be supported without disempowering the other.

For some, diagnosis of the anomaly proved too great a strain on the relationship and the parents separated before the birth. Within this study, two couples separated during the course of the pregnancy, with one couple separating soon after diagnosis. In both cases the women continued with the pregnancy. It is unclear what impact the loss of the partner had on the decision-making process, however, the added grief over a lost relationship is likely to have compounded the emotions experienced by these women. In turn, it could be hypothesised that the baby was perceived as the last tangible part of the relationship and was therefore 'retained'. Clinicians provided much anecdotal evidence suggesting that unsupported mothers were unlikely to terminate an affected pregnancy. However, no literature pertaining specifically to this group following diagnosis of a severe anomaly has been identified. The differing needs and coping mechanisms employed by men and women have been highlighted as significant factors in the way parents manage their grief (Mourik *et al.* 1992), with a major contributor to relationship problems attributed to a lack of synchrony in the grieving process (Robson 2002) and poor communication (Mourik *et al.* 1992). This raises the important issue of legitimating the father in the process, not only to avoid yet

further grief should the relationship fail as a result of it, but also to optimise the support the parents are able to provide for each other.

Little research into the experiences of men whose partners are undergoing fetal screening and diagnosis has been undertaken (Green *et al.* 2004). Yet previous studies have found evidence that the grieving process following termination in particular is dependent on the perceived support of a partner (Black 1989, Statham *et al.* 1999, Korenromp *et al.* 2009), thus highlighting the importance of informing and caring for men in their own right, and in order that they can be supportive to their partner (Statham *et al.* 2001). Reactions by the men to the interview invitation suggested that this is still not the case. Data collated from the interviews highlighted the sense of being a 'bystander', a role that resonates with studies of men's experience of pregnancy and childbirth more broadly (Locock, Alexander 2006).

The concept of disenfranchised grief was examined in section 8.5.3 in relation to termination. However, the identity of a father is another version of disenfranchised grief and, reflected in the social expectations of stoicism and hidden grief (where men don't cry), can further complicate and intensify the grieving process (Robson 2002). Added to this, the frequent assumption that there is a homogenous grief experience raises concerns that as a society and as healthcare professionals we are failing to provide the individualised support required.

Within this study, the delivery process was frequently raised as being the most difficult period for the fathers because they were supporting their partner through a physically and emotionally draining process, whilst grieving themselves. At this time attention was usually directed towards the needs of the women, with the men reporting feeling powerless, a bystander. Lack of engagement from clinical staff was widely reported; a phenomenon commonly highlighted in the literature (Mourik *et al.* 1992, Murphy 1998). Whether this was related to low staffing numbers and thus time, individual views on termination, or simply that staff were unsure or nervous about communicating or engaging with the fathers, is unclear.

Nonetheless, as has been previously raised, healthcare professionals need to consider the father as more than just a supporter for the woman, and acknowledge and meet his needs as an individual and as a father (Locock, Alexander 2006).

We need to find ways to ensure that men are engaged and informed from the outset: this does not currently appear to be the case. Throughout the process the maternal identity is reinforced. From the physical changes in her body to the legal requirements of her signature on a consent form, her role is confirmed by society. Conversely, the paternal identity exists only when validated by others. Yet the grieving process for both is inextricably linked one with the other, and needs to be given consideration when caring for the couple. Symbolic procedures can be implemented to support the couple as an entity without disempowering the mother. Examples such as providing the opportunity for both parents to sign the consent form when a termination has been decided upon, engagement of the father by documenting his name on the front of the fetal medicine notes so healthcare professionals can refer to him by name, and practical aspects such as ensuring both parents have access to basic facilities such as bedding and food whilst in hospital. As the 'patient', these items will automatically be provided for the mother, but as a 'visitor' the father, as revealed in many of the experiences recalled in this study, was frequently overlooked.

8.6 INEQUALITIES OR VARIATIONS

The relationship between deprivation and mortality rates of neonates and infants affected by congenital anomalies (Neasham *et al.* 2001, Oakley *et al.* 2009, Smith *et al.* 2010, Olesen *et al.* 2009), has been partly attributed to the variation in rates of termination of the pregnancy (Smith *et al.* 2011). Examining the processes of parents as they decided whether or not to continue with the affected pregnancy provided some insight into how decisions are reached. By categorising the decision-making groups it was possible to identify and explore possible variations in the processes employed. This highlighted the importance of the decision-making process, rather than the decision *per se*, in creating variations and potential inequalities.

The discourse pertaining to inequalities in health is immense and well established (Carlisle 2001, Pierret 1993, Blair 1993, Kawachi *et al.* 2005). Rather than engaging further in this wider debate this section focuses instead on the impact of socioeconomic status on the doctor-patient interaction and the influence of this on the decision-making processes enacted by the groups identified within this study.

The ways through which socioeconomic status affects healthcare are complex and include the wider social contexts and factors as well as the more immediate social environments, individual psychological and behavioural factors, and biological predispositions and processes (Adler, Ostrove 1999, Van De Mheen *et al.* 1998, Lynch *et al.* 1997, Pickett, Pearl 2001). The influence of these factors on the interaction between doctor and patient, and subsequently the decision-making process itself, provides an important insight into whether the variation in the number of terminations performed across the spectrum of deprivation relates to systematic inequality or parental choice.

Available literature suggests that patient demographics, in particular socioeconomic status and ethnicity, create a lens through which the consultation interactions are encountered, with evidence highlighting the influence of these factors on the way clinicians approach the patient (van Ryn, Burke 2000, Pilnick, Zayts 2012), and the way in which patients perceive the clinician (Doescher *et al.* 2000). Furthermore, these two elements have a cumulative effect on doctor-patient interaction. Patients from more deprived areas are potentially disadvantaged due to the risk that doctors misperceive their lack of interaction as a lack of desire and need for information. Subsequently their ability and opportunity to take part in the care process is diminished (Willems *et al.* 2005).

Clinicians within this study highlighted their desire for parents to engage in an idealised decision-making process. In Chapter 6, tensions arising from failure to engage in components of this process were identified. These pertained to the desire for a 'rational' decision-making process where the parents were actively engaged and options weighed up. Cross-referencing these with the inequalities literature, described above, shows the skills required to adhere to the desired

process are often reported as being less common in more deprived communities. Literature demonstrating the direct influence of personal and social attributes, such as educational level, on patients' communicative behaviour with clinicians is well established (Street 1992, Street Jr. 1991). Whereas patients from higher socioeconomic groups demonstrate more active communication, along with a greater ability to express their needs (Stewart 1995), those from lower groups are more likely to experience difficulties during the interaction (Willems *et al.* 2005). This may in part be due to the smaller "*cultural distance*" due to similarities in background between high socioeconomic status patients and the clinician (Street Jr. 1991, pg. 546). Clinician responses to patients of low socioeconomic status may include a more directive and less participatory consulting style, as well as being associated with less information giving and openness (Willems *et al.* 2005). However, as demonstrated in this study, this was dependent on the establishment of a 'good' doctor-patient relationship.

The rational decision-making exhibited by the Consequential Group (all high socioeconomic status), and the largely uncomplicated interactions between this group and the clinicians, reflects the experiences reported within the literature. The lack of tensions between this group and the clinicians caring for them is also suggestive of a shared understanding, and social capital (Webb *et al.* 2008).

The directive approach employed by clinicians when caring for the Choice Removed Group (all low socioeconomic status) also reflects the consultation style between clinicians and patients of low socioeconomic status, reported in the literature. Within the context of this study, the benefits of the directive counselling for this group, highlighted in previous chapters, suggest that the clinicians' interactions with the group appeared to be a calculated attempt to respond to their needs, rather than an enactment of an inequality.

Interactions between the Absolute Group (all low socioeconomic status) and clinicians were more strained. The difficulties and tensions highlighted between this group and the clinicians suggest a lack of insight into each other's perspectives. Although these parents all continued with the affected pregnancies,

and thus theoretically contributed to the 'inequalities' in outcome observed, an active decision, based on a fundamental belief system, was made.

The Avoid/Delay Group portrayed many of the attributes applied to patients of low socioeconomic status within the literature. The influence of the doctor-patient relationship, and the perceptions of clinicians and of women, on which the relationship was built, were reflected in the subsequent difficulties experienced in communicating. These ongoing issues resulted in parents in this group continuing the pregnancy by default, rather than through choice. The parents in this group and clinicians appeared to enter a vicious cycle that was fuelled by the lack of skills available to the women, and the potential misinterpretation, by the clinicians, that lack of engagement and subsequent withdrawal equated to lack of interest on the part of the women. This perhaps reinforced clinicians' beliefs about the women's cognitive ability and information needs (Street 1992). In turn, the women were unable to identify with the clinicians due to the large cultural distance and were perhaps dissuaded from asking questions (van Ryn, Burke 2000). As with other forms of interpersonal communication, medical consultations are "*processes of personal and mutual influence that unfold according to the characteristics of the individuals and to interactive processes related to how interactants adapt their communication to one another*" (Street 1992, pg.1155). Failure to adapt, risks the development of inequalities. Whether a different approach with this group of women would have resulted in a different outcome is uncertain. However, an inequality is evident within this group that has resulted from the lack of opportunity to make an informed decision. This may have arisen partially as a result of the intrinsic cultural differences between the clinicians and the parents within the Avoid/Delay Group.

The decision to terminate or continue an affected pregnancy is largely representative of the different attitudes and expectations of the parents involved. Therefore, associating a decision to terminate or continue a pregnancy to an inequality does not necessarily reflect the underlying complexities involved. Examining the different decision-making processes employed highlights some inequalities in the way in which the decision-making processes are supported.

However, more effective communication could be established by clinicians and parents through clinicians' awareness of the inherent differences in perspective (Willems *et al.* 2005).

8.7 RECOMMENDATIONS AND IMPLICATIONS

Findings from the study have enabled a total of twenty recommendations to be made. These have been drawn from across the contextual framework and include those relating to care processes and pathways, staff training and development, infrastructure in terms of buildings and materials, as well as local and national policy recommendations.

8.7.1 CARE PROCESSES AND PATHWAYS

A better understanding of the various processes employed by parents to make a decision is required by clinicians. The drive against directive counselling needs to be reconsidered. This is not an attempt to reclaim paternalism, but rather to highlight the differing needs of parents and permit clinicians to respond to these needs in an appropriate and compassionate way. Use of the model developed to highlight attributes of particular parents may aid clinicians in determining when directive care is appropriate. It is unlikely that clinician education alone will be effective in this area. Policies and guidelines designed to demonise directive counselling need to be reconsidered.

In addition, the current informed consent process in relation to screening for FASP anomalies is clearly ineffectual. Although a balance must be reached where undue fear is not created through the overemphasis of unlikely events, the difficulties encountered by women when faced with a decision following an unexpected diagnosis should be addressed. The current process is simply a perceived exoneration of responsibility from healthcare professional to parent. By providing information on which the parents can make an 'informed choice', the onus moves from the healthcare professional (to provide the information), to the parent (to make the decision). This process enables the healthcare professional to distance themselves from the decision and subsequent outcome.

The care pathways and support available to women following stillbirth or neonatal death are well documented. Although women who terminated frequently accessed aspects of this care, the support offered was generally more haphazard and lacked the same structure. This held true both in the healthcare and wider social contexts. Delineating the gestation at which women are entitled to claim financial support, in terms of benefits and time, meant that women risked being forced back to work before they were ready, out of necessity. Although provision of financial support cannot be universal, alternative ways of supporting this group need to be given consideration. This could potentially involve a new form of benefit, or funding available through clinicians. The mechanism of accessing funding has not been explored: however, the need, in some cases, is clearly apparent.

At the time of undertaking this study, a joint project was underway between the fetal medicine clinicians in one of the centres and coroners to investigate the possibility of creating a fast-track post-mortem process following termination for a severe congenital anomaly. This is an example of good practice that could be replicated. The distress caused to some parents in particular due to the delays in receiving post-mortem results was extensive where confirmation of the antenatal diagnosis was required for reassurance that the 'right' decision had been made.

Consideration also needs to be given to balancing the legal responsibilities of the mother with legitimising the role of the father within the process. The ultimate responsibility for the decision remains with the mother. However, providing opportunities for fathers to actively engage in the process, with the consent of their partner, should be considered. Adoption of practices such as a joint consent form for termination, although not a legal requirement, would provide the opportunity for parents to demonstrate their joint responsibilities whilst also caring for the parents as a unit. However, the final decision would rest with the woman, thus ensuring that a joint consent did not enable a partner to prevent a woman terminating their pregnancy. The long-term psychological needs of the fathers frequently differed from those of the mothers. Provision of specific counselling services tailored to meet their needs requires assessment and subsequent investment.

The use of Bounty bags and similar promotional material is well established within the healthcare setting. Arguments relating to the negative impact of the distribution of these items, particularly on vulnerable groups such as the parents in this study, are equally well established (Chitty *et al.* 1996, Donnelly *et al.* 2000). Nonetheless, a cultural acceptance of their presence appears to have dampened any resistance, with healthcare professionals allowing this practice to continue unchecked. This requires a new debate to be opened, and consideration to be given to the impact on vulnerable groups who do not necessarily follow the 'normal' pathway.

Box 8-1 **Summary of recommendations - care processes and pathways**

Immediate Social Context

- *Meet the psychological needs of **both** parents during and after the decision-making process*

Healthcare Context

- *Create structured care pathways and automatic access to counselling through the NHS*
- *Speed up post-mortems*
- *Legitimise the role of the father through use of joint consent forms, where appropriate*
- *Place constraints on commercial services such as Bounty within the clinical environment - patients can ask for a pack, but they are not automatically distributed*

Legal and Professional Context

- *Re-examine and discuss of the 'informed' consent processes for screening to determine an appropriate way forward*
- *Examine and discuss issues surrounding non-directive care – policies and guidelines should be re-examined*
- *Greater access and provision of information is required, so clinicians can gain a greater insight into the way parents enact the decision-making process*

Broad Social Context

- *The option of some financial support for parents following diagnosis of a severe congenital anomaly is required*

8.7.2 STAFF TRAINING AND DEVELOPMENT

Whilst midwives in the fetal medicine unit had chosen by virtue of their job to care for women who choose to terminate, discussion with senior midwives highlighted that local policies did not allow labour ward midwives to opt out of caring for women undergoing a termination, an issue which has been reported more widely (Lloyd 2014). Within medicine, healthcare professionals are expected to care for any patient requiring their attention. However, the field of reproductive health, and particularly termination, is the only area in which freedom of conscience is accepted as an argument to refuse care to women (Fiala, Arthur 2014). The Abortion Act 1967 allows healthcare professionals to refuse to participate in direct involvement in 'abortion' care, provided it is not an emergency case (Abortion Act 1967). A ruling following a court case taken by a Scottish Trust against two midwives who refused to be involved is currently awaited at the time of writing (Greater Glasgow Health Board v Doogan and another 2014). The result of the appeal by the Scottish midwives is likely to have far wider consequences than the

immediate rights of the midwives. There is either the issue of inequality of access, where rural areas supported by a small number of midwives may be unable to offer a termination service, or of midwives who do not wish to care for women terminating a pregnancy being obliged to do so. The latter was reflected in some of the comments made about their care by women and their partners in this study. One of the most harrowing aspects of this study has been hearing the narratives of poor care arising from the perceived judgement of healthcare professionals on the decision of parents to terminate an affected pregnancy.

The sentiments of staff working on the maternity units have not been explored; care must be taken in attributing causality for these events. In addition, there is little literature exploring their perspective to provide additional insight (Vinggaard Christensen *et al.* 2013). Furthermore, a recent parliamentary report into midwifery services in England described a workforce that was overstretched (Public Accounts Committee, 2014). Margaret Hodge, who chairs the committee, said: "*There is evidence that many maternity services are running at a loss, or at best breaking even, and that the available funding may be insufficient for trusts to employ enough midwives and consultants to provide high quality, safe care*" (Public Accounts Committee, 2014). Staffing levels are likely to have played an important part in the care received. However, seeking a solution is essential, and a number of approaches could be given consideration. New standards into minimum staffing levels currently being considered may assist but will not resolve issues pertaining to attitudes of permanent staff. Trusts should seek to better appreciate their midwives' understanding and acceptance of termination practices. Extending the role of fetal medicine midwives, who actively choose to work in an environment where women's 'choice' is supported, is perhaps one solution.

Although most fetal medicine units across the UK provide care through a model similar to that presented at the centres studied, one centre offers an alternative, bridging the gap between fetal medicine and maternity services through sharing and rotation of midwifery staff (Fisher 2013). In addition, clinicians retain responsibility for the care of the women terminating and of delivering an infant with a severe congenital anomaly. Anecdotal evidence from national charities

supporting women following the diagnosis of a severe anomaly suggests that many of the care concerns highlighted within this study appear to have lessened with the implementation of this novel model of care (Fisher 2013). Recommendations pertaining to clinician training and recruitment also require consideration. First, the gradual dilution of skills in terms of late surgical termination techniques requires addressing. As described in Section 8.4.3, national policy changes have moved a large proportion of terminations from the NHS to the private sector. This has resulted in a de-skilling of NHS clinicians, particularly in late termination practices. The case for choice in relation to method is well documented. A number of options are available. The most effective is likely to be development of better working relationships between NHS and private termination services. This would enable easier access to late surgical terminations through the private sector, with ongoing care provided through the NHS organisation. This is perhaps a short term solution, with a more effective long term solution resulting from the use of private services as a training placement for trainees. The long term goal would be the reintroduction of late surgical termination skills to the NHS. There are likely to be a number of obstacles to achieving this, including funding issues. Exploring these are beyond the scope of this thesis; however, the argument for choice of method is strong and careful consideration must be given to overcoming these obstacles in order to provide best care to parents.

Policy is unlikely to have been the only barrier to the availability of late surgical terminations, with some evidence suggesting that clinicians find the “*dirty work*” associated with the procedure distasteful (Harris 2008, pg. 79). This is unsurprising, as the procedure is undertaken by removing sections of the fetus individually. This is a more difficult issue to overcome, and perhaps clinician choice to perform these procedures remains the only option. However, this may require consideration when recruiting clinicians to work within this specialised field. In addition, consideration should be given to the availability of a non-judgemental support for these healthcare professionals. ARC has recently instigated a membership forum for healthcare professionals. The impact of this is unclear, although responses following the study interviews suggest that this is something that could meet a growing need.

A further recommendation pertains to making bereavement counselling training obligatory for all healthcare professionals caring for women undergoing a termination. At present these courses remain optional, and are often difficult to access in terms of time and funding requirements. A number of national courses are provided by charities including ARC and SANDS. However, support from the trust would be required in order to access these.

Box 8-2 Summary of recommendations - staff training and development

Healthcare Context

- *Trusts need to gain a greater insight into the views and beliefs of midwives around caring for a woman who is terminating a pregnancy in order to effectively plan and provide services*
- *Counselling training should be mandatory for all clinical staff involved in the care of women and their partners following suspicion and diagnosis of an anomaly*
- *An increase in staffing levels is required in order to provide one-to-one care for those terminating a pregnancy – moving between a live birth and a termination creates intense difficulties for both midwife and parents*
- *Consideration to alternative care models where parents remain in the care of fetal medicine throughout their journey, including delivery*

Legal and Professional Context

- *Supporting choice of method through closer working with private sector, including BPAS*
- *Re-examination of training opportunities for surgical terminations*
- *Clinician recruitment – both nationally and locally, recruitment should ensure that there is access to clinicians trained to provide all necessary procedures (including surgical termination)*

8.7.3 INFRASTRUCTURE

A number of practical improvements could be considered when renewing or building centres providing antenatal care. First, provision of separate waiting areas for fetal medicine and antenatal care should be considered. This would potentially work well where consulting rooms were shared, but dual access was available. This would enable parents to avoid facing other pregnant women in the event that an anomaly was suspected, but prevent the demarcation of rooms as ‘bad news’ rooms. In addition, for those that decide to terminate the pregnancy, avoidance of other women following ingestion of the tablets to induce labour, or following a feticide, would be possible. Conversely, for those that subsequently continued with the pregnancy, the option to remain integrated with other women in the antenatal setting would be feasible.

Significant investment in areas where women can deliver following loss of their baby is required. Investment to date in all the centres studied was predominantly made through charitable means, frequently driven by individuals who had been through the experience. Although all the centres were working towards having a separate bereavement room, policy in all the centres involved dictated that parents had to choose between the private room and some forms of pain control. Consideration needs to be given to the complete separation of bereavement

delivery, whether stillbirth or termination, and other deliveries. A separation in location would also ensure a separation in staffing.

Box 8-3 Summary of recommendations - infrastructure

Healthcare Context

- *Two doors to the consultation room, enabling parents to leave without facing pregnant women*
- *National level investment in facilities for delivery – choice between privacy and pain control is not acceptable – separate facilities required*
- *Provision of facilities for partners during the delivery – bedding, food etc.*

8.7.4 LOCAL AND NATIONAL POLICY RECOMMENDATIONS

Public perception and understanding of the FASP anomalies is likely to change over time, as reflected in the experience with Down's Syndrome. However, the low incidence, lethal nature and high termination rates of the FASP anomalies may result in an ongoing poor understanding of these anomalies within the wider community where babies with these anomalies are unlikely to be seen. This in turn impacts not only on the support available to parents facing a decision, but may heighten the stigma attached to termination for these anomalies where public understanding is poor.

An over-riding recommendation for change stems from the current legal position in England. As discussed in section 1.4, termination is currently a criminal procedure, although exceptions are made within certain circumstances. Consideration should perhaps be given to reversing this, and instead provide clear professional guidance on which to determine when the offer of a termination is morally acceptable. By decriminalising the procedure, the current fear of scrutiny experienced by clinicians should be resolved. In addition, removal of the criminal

label applied to termination may help reduce the stigma attached to the procedure, and subsequently to those who undergo it.

Box 8-4 Summary of recommendations – local and national policy

Legal and Professional Context

- *High level discussion is required into the feasibility and potential to reduce the stigma associated with termination, by decriminalising termination*

Broad Social Context

- *Increasing public awareness of anomalies*

8.7.5 FUTURE RESEARCH

Despite the contributions of this study, this remains an under-investigated field. Future work is required in this area, to give consideration to the medium and longer term outcomes of the parents represented here. The role of the father, in the decision-making process and subsequent support of the decision, is lacking within the literature, as is the impact of the process on his psychological wellbeing. As identified in section 8.3 there are a number of limitations to this study, including the applicability of the findings to non-English speaking minority ethnic groups, and to other centres around England and Wales. Undertaking a form of audit to explore and highlight different practices across settings may provide evidence of other ways of doing things. In addition, extending this study to include non-English speakers will provide new insights.

More widely, a greater understanding of parental attitudes to the different anomalies is required in order to gain some insight into the impact of the variables identified as influential in decision-making. In particular public perception of the ‘treatability’ of some cardiac conditions such as hypoplastic left heart syndrome may impact on the decision to opt for palliative or comfort care when termination

is not an option for whatever reason. Overall the option for comfort care appeared to often be overlooked. Clinicians' attitudes to the offer and uptake of this option require exploring to better understand the difficulties faced when caring for unborn babies with lethal conditions.

In relation to the impact of socioeconomic status on decision-making following suspicion or diagnosis of a severe fetal anomaly, whilst this study provides some insights, it lacks the nuanced investigation required to provide wide ranging recommendations. Application of the model to identify interventions that could aid decision-making is essential to improve care offered to the women.

Box 8-5 Summary of recommendations - Future research

Future Research

- *Medium and longer term outcome studies*
- *Role and impact of the father*
- *Extend to non-English speakers*
- *Explore different practices across trusts*
- *Clinician and parental attitudes to comfort care*
- *Public understanding of specific congenital anomalies*
- *Application of the model to identify potential interventions*

8.7.6 MY FUTURE PLANS

Following submission of this thesis, I plan on publishing a number of articles derived from my work. At the time of writing, I have a partially drafted article documenting the experiences of women as they make sense of their decision. This reflects closely the findings presented in Chapter 7. I will also be presenting these findings at a national conference arranged by ARC (September 2015) and

subsequently speaking at a SHINE¹⁹ conference (September 2015). Publications reflecting Chapters 4 (combining the role of nurse and researcher), 5 (the influence of context on decision-making) and 6 (review of the decision-making model) will follow. My aim is to identify funds to continue the research into the experiences of this group of patients. Primarily, I would like the opportunity to explore public and parental understanding of specific anomalies. Perception of cardiac anomalies is of particular interest, and relates closely to my clinical expertise. The difference between antenatal perception and subsequent beliefs of parents following birth appears to diverge. The term ‘heart warriors’ is widely used to describe the babies who survive cardiac surgery, and expectations of parents appear to be limitless in terms of what is feasible through surgery or intervention. How clinicians feed into these expectations is also unclear. Nonetheless, these expectations are likely to impact on the decision to terminate or continue an affected pregnancy and may subsequently result in poor psychological outcomes where reality and expectations do not meet. Applying the model developed within this study to a group of parents may provide additional insight into potential interventions to support them.

8.8 CONCLUSION

This study set out to gain an insight into the decision-making processes of women and their partners following diagnosis or suspicion of a severe congenital anomaly. Subsequently, a greater awareness of the variations in the pathways and processes followed by them emerged, thereby providing an understanding of how variations in termination rates arise.

It has taken much strength and tenacity to pursue this study. At many points it would have been simpler to avoid the multitude of barriers by searching out a less emotive topic. However, this study has provided a platform for parents to tell their story. With patients’ experiences described as *“the final arbiter in everything the*

¹⁹ SHINE – National Charity for: Spina Bifida, Hydrocephalus, Information, Networking, Equality.

NHS does" (Department of Health 2011, pg.25), the importance of this cannot be underestimated. Furthermore, the gradual acceptance and slow developing appreciation of the value of the study by the clinicians may, in the future, lead to a change in their practice. This in turn may result in the opening up of opportunities for further sensitive research with the ultimate aim of improving outcomes for patients.

Despite the challenges posed, the combination of consultation recordings and interviews has proved an effective way of illuminating the complexity of the decision-making processes. As the complexity of the process became apparent, initial assumptions about the nature of the decision-making process were challenged. In particular, the extension of the definition applied to decision-making to include aspects relating to making sense of the process was fundamental to the insights gained.

The diagnosis of a severe congenital anomaly during pregnancy is a devastating experience. The unexpected nature of the events that unfolded left many parents feeling unprepared for the subsequent dilemmas they faced. Decision-making is a complex process, navigating ill-structured problems, fuelled by fluctuating uncertainty, high stakes and the competing goals of multiple players, all within a multitude of interwoven contextual and temporal factors. An assumption is often made that more information will lead to a more informed decision. However, as demonstrated within this study, this assumption fails to account for the complexities of the decision-making processes enacted by the parents. Added to this, the stigma associated with termination and disability accentuated the difficulties encountered by parents, whether they terminated or continued the affected pregnancy.

Clinicians, too, are not immune to the impact of the complexities involved as they negotiate the vagueness of the law and manage the intense scrutiny placed on their practice. The rhetoric of non-directive counselling creates an artificial barrier between themselves and the decision by devolving responsibility for the decision onto the parents. An idealised 'rational' decision-making process was sought by

clinicians. For some parents this proved unproblematic as their willingness and ability to engage with the situation reflected that espoused by the clinicians. For others tensions arose between the clinicians and parents, where parents would not or could not synthesise the information provided in order to engage in the sought after 'rational' decision-making process. Decisions were made as a result of either a reaction to or interaction with the clinician.

There is an intricate relationship between the variation in termination rates following diagnosis of a severe congenital anomaly and the different decision-making processes observed. Each process brings its own intrinsic problems and reflects the impact that the combined contextual framework, in which the decision is enacted, has on the decision-making process. 'Softening' of the wider contextual layers, in order to enable the system to be more reactive and responsive to individual parent's needs, may reduce some of the variation seen. Although the findings from this study have provided some insight into how the variations occurred, care must be taken not to conclude that systematic inequalities exist, but see the variations in outcome as a reflection of differences in attitudes and expectations of parents. What is required is a system that supports the decision-making process of all parents, rather than a fixation on the outcome itself.

Literature pertaining to the decision-making processes of women and their partners following a diagnosis of a severe congenital anomaly is sparse. Thus the findings from this thesis provide a valuable insight into the lived experiences of parents coping with this traumatic event. It is hoped that these will provide a much needed contribution to the understanding of those who determine the policy and those who practise within the field of fetal medicine.

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APPENDIX A - SEARCH TERMS AND STRUCTURE

Population / problem	Intervention	Comparison	Outcome 1	Outcome 2
fetal anomaly	decision-making	socioeconomic status	prenatal testing	abortion
Alternative wording				
fetal /foetal	decision	socioeconomic	prenatal	termination
congenital	making	status	pre-natal	eugenic
anomalies/	choice/s	inequalities	pre natal	abortion
abnormalities	decision/s	social class	antenatal	
anomaly/		educational	ante-natal	
abnormality		level	screening	
			testing	
			diagnosis	

Once a series of concepts that reflect the PICOS elements had been compiled they are then combined using Boolean logic (AND, OR, NOT) to create a set of results that contained articles relating to the topic in question. As can be seen from the table overleaf, use of Boolean logic reduced the searches identified significantly.

EXP – explode, results using the MeSH term and selected subheadings, demarcated by square brackets. Where there are no brackets all subheadings were used.

MeSH terms following a / indicate *additional* MeSH terms identified from the main tree headings adjunct to the original MeSH term searched for.

Subheadings used to restrict the search

Searches such as #2 and #3 (decision-making and socioeconomic status) were omitted as they were too broad, returning nearly 200, 000 articles.

#	Searches	
#1	"eugenic abortion"	
#2	"Socioeconomic factor?" OR "social class" OR "educational status"	
#3	"decision?making" / OR "choice behavio?r" / OR "uncertainty"	
#4	EXP "congenital abnormalities"/ OR "genetic diseases, inborn"/	
#5	EXP "congenital abnormalities" / [Classification, Complications, Congenital, Diagnosis, Epidemiology, Mortality, Nursing, Psychology, Surgery]	
#6	"Prenatal Diagnosis" / OR "amniocentesis" / OR "chorionic villi sampling" / OR "fetoscopy" / OR "maternal serum screening tests" / OR "nuchal translucency measurement" /	
#7	"Prenatal Diagnosis" / [Classification, Ethics, Mortality, Nursing, Psychology, Trends, Utilization]	
#8	#1 and #2	142
#9	#1 and #3	251
#10	#1 and #2 and #3 and #5	16
#11	#2 and #5	1531
#12	#2 and #7	351
#13	#2 and #5 and #6	136
#14	#2 and #5 and #7	9
#15	#3 and #5	200
#16	#5 and #7	166

MeSH codes in inverted commas; wildcard operators represented by a question mark

APPENDIX B - PATIENT AND PUBLIC INVOLVEMENT

CLINICIANS

Following informal discussions with clinicians and parents, the recruitment process originally envisaged was radically overhauled, as theoretical understanding of the patient journey failed to reflect the complexity of the real clinical situation.

In addition, the use of a diagnosis as an inclusion criterion had to be abandoned following the realisation that this would exclude a number of women for whom a diagnosis is not made antenatally as the women choose not to proceed with further testing. Although the clinician may suspect severe anomalies, based on the soft markers, no formal diagnosis can be made. This group of patients is particularly important as they may contribute significantly to the infant mortality deaths, where the anomaly is not compatible with life.

The initial meeting with local clinicians resulted in an invitation to observe the centre at work, with the aim of developing some insight into patient care pathways and potential problems in recruitment. Other issues highlighted by the clinicians were the need to reconsider inclusion and exclusion criteria, the timing of the interview and the method of recording. Prior to commencement of the data collection, and as part of the preparatory stage, a number of interviews were conducted with clinicians. The initial aim of the interviews was to familiarise and sensitise myself to issues that may arise within the interviews with parents. However, the data arising from these provided a much greater insight than initially anticipated.

OBSERVATION

Through the clinicians who took part in the PPI aspect of the study, I was able to secure an honorary contract that enabled me to observe the day to day workings of the centre before starting the research. This was immensely beneficial in

providing time for relationships and trust to develop between myself and the healthcare professionals before engaging in the research. It also helped in familiarising myself with processes and procedures that allowed me to gain insight into the workings of the centre.

CHARITIES

A number of local and national parents' groups and charities were approached, with a representative providing some insight into the needs of the parents. Little Hearts Matter, a national charity supporting parents following diagnosis of a severe cardiac anomaly, was particularly supportive of the research aims. In addition, they felt that the use of recordings of consultations is to be applauded as it will provide much help to parents. Although this does not constitute normal practice, the data arising from these consultations will be very rich. Assistance was offered by a number of the charities in the dissemination of the findings at the end of the study.

Charities approached included ARC (Antenatal Results and Choices), Shine (National Spina Bifida charity), LHM (Little Heart Matters), local cardiac charities (Heartlink and Keepthebeat). In addition, a number of parents contacted me through social media, having read comments on Facebook made about the project by other parents. The spokesperson from Shine discussed the influence of social media on support networks between parents. She felt that the geographical barriers (particularly with rare disorders) and deprivation barriers are removed through the use of Facebook in particular. In light of the mechanism through which a number of individual parents contacted me, this was of particular interest and may warrant further investigation in the future.

PARENTS

All the parents were antenatally diagnosed at varying gestations. They came from a cross-section of deprivation, as defined and calculated above. Different anomalies and decisions, in terms of continuing or terminating the affected pregnancies, had been made. Although the parents were predominantly female, one father was included.

Parent number	Gender	Pregnancy terminated or continued	Anomaly
1	Male	Terminated	Structural anomaly
2	Female	Continued	Structural anomaly
3	Female	Continued	Structural with underlying chromosomal anomaly
4	Female	Terminated	Chromosomal and severe cardiac
5	Female	Continued but opted for palliative care	Structural anomaly

The demographic spread of the parents who contributed to the project design is documented above. It is generally representative of the population that was recruited for the project. The parents were spoken to individually, and it was made clear that this aspect of the research development would not constitute any part of the actual research, but was preliminary work to investigate the acceptability and importance of the research from the parents' perspective. Each of the parents was asked three questions:

- *What are your thoughts about the research question?*
- *Who gave you your diagnosis and which doctors did you see?*
- *How would you have felt if someone asked you to take part in some research at that time?*

All the parents were very supportive of the research aims. There appeared to be a general consensus that research in this area was lacking and an increase in awareness of issues would be beneficial to families and staff.

No negative comments were made with regards to interviewing parents, with some of the positive comments listed above. There was an over-riding feeling that talking about their experience validated their choices and gave meaning to the child. For those that terminated the affected pregnancy, difficulties in overcoming the stigma attached to termination were mentioned, along with the desire to talk in an environment away from health care professionals. There was a sense of needing to validate the fetus and again to give it some meaning.

Another issue raised by parents was the number of people in the consultation room. Although many of them were there in an observational capacity, the addition of a researcher in the room would not have been readily accepted. One woman stated that there were eight members of staff present at her initial consultation with the cardiac specialist. This needs to be taken into consideration with regards to consent for recording consultations, as well as the potential to make transcribing more difficult.

AUTHOR CORRESPONDENCE

A number of academics who have published on related subjects were contacted. A number of issues were highlighted and ideas implemented. These are discussed in section 4.2.

APPENDIX C - INTERVIEW SCHEDULES

Interview schedule: women and partners	Theme
Have you read the information sheet? Do you understand that you can change your mind about being involved with the project at any time? Are you OK to sign this form to say that you've understood what is going to happen?	CONSENT
Is it ok if I record the interview? Have you got any questions about the project? If you want to stop the interview, please let me know and we will stop straight away The reason for talking to you today is to find out about your experiences of your pregnancy	BEFORE WE START
Can you tell me a bit about yourself? • <i>Demographics</i> • <i>Relationship</i> • <i>Number of children</i> • <i>Any problems with previous pregnancies</i> • <i>Same father – ask the father how many children he has</i>	TELL ME A BIT ABOUT YOURSELF
Can you tell me a little bit about the pregnancy? Was it planned? What kind of care? (Consultant, GP, midwife-only).	PREGNANCY BEFORE
When did you first think there was a problem? Can you tell me about what happened? How many weeks pregnant were you? Did you have any tests? What kind of tests did you have? Can you talk me through what happened in order?	PREGNANCY - SUSPICION OF ANOMALY
Where applicable Some of the tests you had gave you a 'risk' of a disorder. How did the healthcare professional explain the risk to you?	APPROACH TO RISK

How easy did you find that to understand? Can you tell me about what happened after you were given the results? How did your partner feel about the decision? Did the clinicians help you make a decision on what to do about further testing? Is there anything they could have done to make it easier for you to understand?	
I would like to talk a little bit about how you were told that there was/ might be a problem with your pregnancy. How many weeks pregnant were you at this point? Who told you and how were you told? If you had to tell someone else what would you have said or done differently?	BREAKING THE NEWS
When it came to deciding what to do next, how much support did you feel that you had? If you made the decision with a partner - Did you discuss it together - before seeing a professional/afterwards? Did either of you discuss it with anyone else? (friends or family, another professional) Did you have enough time to make a decision? Did you discuss the diagnosis with anyone else? Where did you get your information from? Did you get any advice from family and friends? What sort of advice? When you spoke to the hcp's: Did they all give you the same advice, or did different people tell you different things? Did they support your decision? Why do you feel that? Did you want to talk things through further with the hcp? How did the consultations with the hcp feel? How would you like things to have been? Did you approach or speak to any organisations or other parents?	MAKING THE DECISION

Going back to the problem you were told was affecting your pregnancy: Had you heard of it before? Can you tell me about it? Do you know anyone else who had experienced the same thing? Did you try and find anyone who had had the same problem? If so, can you tell me how that made you feel? If not, was there any reason why not? Do you think it would have helped? Did any friends or family know anything about it? If yes, did they talk to you about it? Could you tell me some more about that? Where did you get most of your information about the problem from?	UNDERSTANDING THE ANOMALY
How do you think our society sees people with disabilities?	APPROACH TO DISABILITY
Is there anything else you would like to tell me about your experience? Do you have any messages for other women in a similar position? What would you like to say to hcp's working in this speciality?	ENDINGS
Other people have said..... Is that your experience? I'd now like to move on and talk about your experiences You just mentioned and that brings me onto another one of my questions Can you give me an example of Some people I've interviewed said why do you think that might be? I just wanted to clarify You mentioned that could you explain why?	ADDITIONAL PROMPTS

Interview schedule: clinicians	Theme
Have you read the information sheet? Do you understand that you can change your mind about being involved with the project at any time? Would you sign the consent documentation?	CONSENT
Is it ok if I record the interview? Have you got any questions about the project? I'd really like to cover much of the information that we discussed when we originally spoke about the project. It would be really helpful if we could cover the same aspects again.	BEFORE WE START
Gender Ethnic Origin How long have you worked in your current position?	TELL ME A BIT ABOUT YOURSELF
How do you determine what a severe anomaly is and what it isn't with regards to offering a termination under Section E? In the case of a parent who wanted a termination and you felt there was insufficient evidence to suggest that the fetus had a severe anomaly, what would you do? Has this caused any problems with parents in the past? Can you tell me about it? Is there any aspect of your relationship with some women that makes your job harder? And easier? Can you tell me a bit more about it? Some of the national charities I have spoken to suggested that some women from smaller hospitals do not get as far as fetal medicine when a structural anomaly is suspected and are offered a termination by their obstetrician. Is this something you have encountered or have any views on?	PREGNANCY SUSPICION OF ANOMALY
Do you find parents justify their decisions to you? If so, what sorts of issues are raised?	APPROACH TO RISK
In your experience, what is it that parents want? Do you find that you can predict the outcome after meeting the parents?	BREAKING THE NEWS

Why do you think that?	
Do you think that there are differences in the way parents from different socioeconomic groups make the decision to continue or terminate an affected pregnancy? What sort of issues have you seen? How do you deal with that? Do you find any particular groups of parents more difficult to deal with than others? If so, why do you think that could be?	MAKING THE DECISION
In your experience do you find parents look for their own information? What are the biggest problems that you encounter with regards to women identifying misinformation? How do you determine how much information to give to parents? How do you present the different anomalies to parents?	UNDERSTANDING THE ANOMALY
Do you see many stereotypical perceptions of disability when you talk to parents?	APPROACH TO DISABILITY
In an ideal world, how would you change the service that you provide? What do you think is missing from the current service? Is there anything else you would like to tell me about your experience?	ENDINGS

APPENDIX D - DEPRIVATION

In 2004, the Office for National Statistics (ONS) introduced a new national geographical breakdown of area, called Super Output Areas (SOA). The aim of these was to define areas for the purpose of collecting, aggregating and reporting statistics. Previous geographical demarcations such as electoral wards had a number of disadvantages including the huge variation in size and changing of boundaries that resulted in significant difficulties in analysing and presenting statistics over time periods.

The SOA's boundaries are intended to be durable, thereby enabling ready comparison over time. These boundaries were used for the first time in 2004 in the Indices of Deprivation and represent the smallest areas for which deprivation data is available. The Indices of Deprivation are an attempt to measure deprivation in a consistent way for small areas across England. They were first developed in the mid 1990's using data from the 1991 Census. Since then the Indices have been revised several times using updated data and new indicators to reflect change.

There are 32482 SOA in England, with just under 400 in the area of study. Each SOA comprises of a maximum of 1500 residents which has the effect of limiting heterogeneity in each area. There are a number of indices of deprivation collated. These are combined into the Index of Multiple Deprivation that is an inclusive measurement of seven domains, thus ensuring a broad measurement of deprivation. The domains relate to income deprivation, employment deprivation, health deprivation and disability, education, skills and training deprivation, barriers to housing and services and living environment deprivation and crime.

For each SOA, an index of deprivation has been calculated. Although the deprivation indices for individual postcodes are not recorded, the SOA for each postcode is documented and can thus be cross-referenced. This process is demonstrated in Appendix E.

The study by Smith et al (2011), from which this study developed, created a deprivation scale across England (Smith *et al.* 2011). All the SOA's were ranked by deprivation scale and the scores divided into 10 groups to create deciles of deprivation. A similar structure was used to grade deprivation in this study. Postcodes were cross referenced with their corresponding SOA, and each participant's decile of deprivation identified. Once recruited, a proforma was completed to enable additional details to be collated directly from the women. Whilst the decile of deprivation provides an objective measure of deprivation, this study is more concerned with ensuring a spread across socioeconomic status. In order to ensure a cross-sectional sampling of participants, deciles of deprivation were equally grouped, and labelled as low, medium or high socioeconomic status. The additional information gained from the patient proforma was then checked against the label applied to ensure consistency.

APPENDIX E– CALCULATING SUPER OUTPUT AREA TO DEPRIVATION

Conversion of a postcode to a deprivation decile score was a two-step process. Initially a postcode was attributed to a super output area (SOA) code, then the SOA was converted to a deprivation decile figure.

POSTCODE TO SUPER OUTPUT AREA CODE

Conversion of postcodes to super output area codes was very simple. A free postcode searcher software was identified that enabled me to search for up to eight codes per day - <http://www.afd.co.uk>. Once registered, a postcode and house number can be typed into the search boxes as shown below.

AFD SOFTWARE DEMOS

Logged in as: **Robyn Lotto**
1 of 8 searches performed



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You must not at any time copy, reproduce, publish, sell, let, lend, extract, reutilise or otherwise part with possession or control of or relay or disseminate any part of this information or use it for any purpose other than your own private or internal use. You shall only be entitled to submit a maximum of eight (8) enquiries to this look up service per day.

Postcode Lookup - Finding an Address

Postcode	LE2 3HE	Hints
Property details		Hints
Lookup		

This produced a large amount of data on the property, including the super output area code, as illustrated below.

Geographical:	
Grid East:	460422
Grid North:	302210
Miles:	
Km:	
Latitude:	52.6143
Longitude:	-1.1090
SOA Lower:	E01013711

SUPER OUTPUT AREA TO DECILE OF DEPRIVATION SCORE

There are a total of 32482 super output areas in England. These are ranked using the Index of Multiple Deprivation (IMD) Score and published at: <http://data.gov.uk/dataset/index-of-multiple-deprivation>.

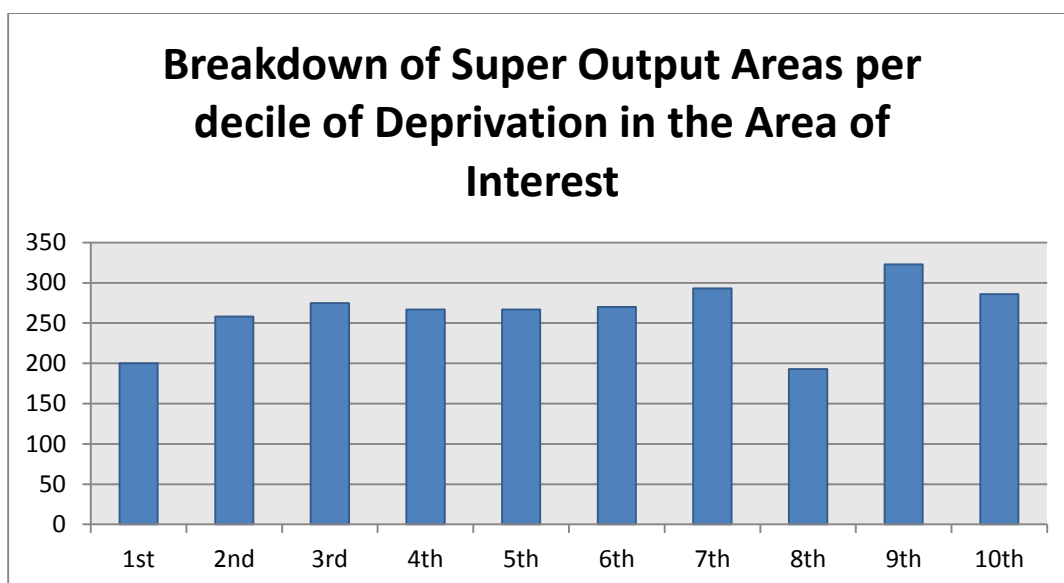
For the purpose of this study, these ranks were then divided into tenths as follows:

Rank of IMD Score			Decile of Deprivation Score
0	To	3248.2	1
3248.3	To	6496.4	2
6496.5	TO	9744.6	3
9744.7	TO	12992.8	4
12992.9	TO	16241	5
16241.1	TO	19489.2	6
19489.3	TO	22737.4	7
22737.5	TO	25985.6	8
25985.7	TO	29233.8	9
29233.9	TO	32482	10

A decile of deprivation score was then applied to each rank of IMD score, based on the bands calculated above. One is most deprived in terms of rank of IMD score and decile of deprivation.

G1		RANK OF IMD SCORE (where 1 is most deprived)							
	A	B	C	D	E	F	G	H	I
	LSOA CODE	LA CODE	LA NAME	GOR CODE	GOR NAME	IMD SCORE	RANK OF IMD SCORE (where 1 is most deprived)	Decile of Deprivation (where 1 is most deprived)	
1	E01028276	37UF				77.12	50	1	
2	E01013818	00FY				74.35	97	1	
3	E01013726	00FN				74.15	100	1	
4	E01013877	00FY				73.42	119	1	
5	E01026173	32UD				73.03	132	1	
6	E01013691	00FN				72.35	157	1	
7	E01028263	37UF				71.72	177	1	
8	E01028066	37UC				71.44	185	1	
9	E01028065	37UC				71.41	186	1	
10	E01013632	00FN				70.87	203	1	
11	E01013948	00FY				69.75	246	1	
12	E01013692	00FN				69.61	255	1	
13	E01013620	00FN				69.33	268	1	
14	E01013755	00FN				69.11	276	1	
15	E01026968	34UB				68.42	302	1	
16	E01013638	00FN				67.71	329	1	
17	E01013754	00FN				67.70	332	1	
18	E01013640	00FN				67.25	351	1	
19	E01026383	32UH				66.83	373	1	
20	E01028265	37UF				65.51	437	1	
21	E01026152	32UD				65.35	448	1	
22	E01013482	00FK				65.20	458	1	
23	E01013590	00FK				65.14	463	1	
24									

LA name, GOR code and name have been removed in an attempt to maintain anonymity. The result is that each Super Output Area (SOA) code has a corresponding decile of deprivation. Within the area of interest there is a total of 2732 Super Output Areas (SOA). The deprivation deciles for the purposes of this study have been based on the deprivation scale for England, not just the area of interest. Calculation of deciles of deprivation for England involves the division of SOA into ten equally distributed groups of 3248.2. Comparison of the deprivation deciles of the area of interest against that of England involves comparison of a constant against the number of SOA's falling within each decile. As can be seen from the table below, the deprivation profile of the area of interest does not reflect that of England overall, in that it is marginally less deprived. The decision to use England data as the denominator was made in order to ensure that each patient's decile of deprivation reflects their position, in terms of deprivation, in the deprivation demographic of the whole of England. This will allow for greater transferability.



In addition this would have enabled me, if necessary, to include or exclude additional postcodes at a later date, without changing the deprivation decile. It was envisaged that this may have been necessary had unusual referral patterns been identified. For example, one patient was recruited from outside the area as the mother had been referred to the one of the specialised services which was not provided by her local area. Therefore, her postcode did not fall within the area of interest, but use of the England adjusted scale meant that her postcode remained comparable.

WORKED EXAMPLE

STAGE ONE:

Postcode chosen at random and not associated with any participant: LE2 4DP

The correlating SOA is identified on the AFD website as shown below.

Geographical:	
Grid East:	462171
Grid North:	300930
Miles:	
Km:	
Latitude:	52.6026
Longitude:	-1.0835
SOA Lower:	E01025974

STAGE TWO

The SOA code is then inputted into the index of multiple deprivation webpage.

	LSOA CODE	LA CODE	LA NAME	GOR CODE	GOR NAME	IMD SCORE	RANK OF IMD SCORE (where 1 is most deprived)
1							
2	E01000001	00AA	City of London	H	London	6.16	28814
3	E01000002	00AA	City of London	H	London	5.59	29450
4	E01000003	00AA	City of London	H	London	13.29	20047
5	E01000004	00AA	City of London	H	London	11.17	22503

	A	B	C	D	E	F	G
25969	E01025968	31UH				6.18	28793
25970	E01025969	31UH				14.74	18490
25971	E01025970	31UJ				7.20	27556
25972	E01025971	31UJ				7.04	27758
25973	E01025972	31UJ				5.97	29021
25974	E01025973	31UJ				5.77	29233
25975	E01025974	31UJ				4.95	30111
25976	E01025975	31UJ				4.31	30736
25977	E01025976	31UJ				24.11	11206
25978	E01025977	31UJ				9.97	24064
25979	E01025978	31UJ				11.89	21657
25980	E01025979	31UJ				6.12	28861
25981	E01025980	31UJ				5.72	29304
25982	E01025981	31UJ				6.97	27861
25983	E01025982	31UJ				3.51	31435

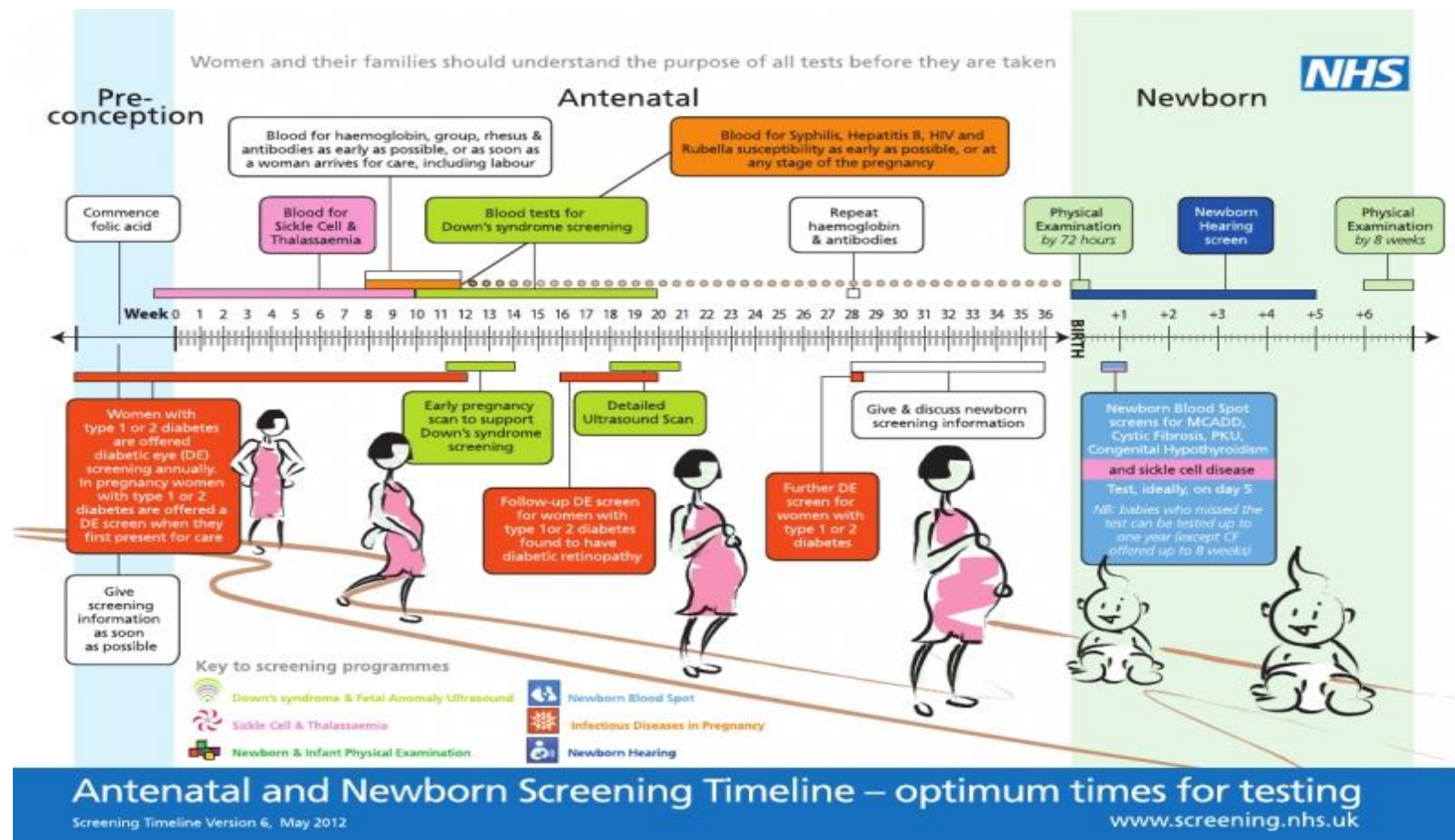
STAGE THREE

The correlating Rank IMD score in this instance is 30111 (row 25975, column G)

Rank of IMD Score			Decile of Deprivation Score
0	To	3248.2	1
3248.3	TO	6496.4	2
6496.5	TO	9744.6	3
9744.7	TO	12992.8	4
12992.9	TO	16241	5
16241.1	TO	19489.2	6
19489.3	TO	22737.4	7
22737.5	TO	25985.6	8
25985.7	TO	29233.8	9
29233.9	TO	32482	10

In this instance the rank IMD score lies within the 10th decile of deprivation.

APPENDIX F – SCREENING TIMELINE



APPENDIX G – PARTICIPANT INFORMATION SHEETS



Experiences of Pregnancy Permission to Contact and Record

We are currently looking for pregnant women and their partners who are willing to take part in a research study.

It will involve recording your consultations with your doctor and potentially an interview with a researcher at a later date. Nothing else will be required from you if you decide to take part. We are interested in your experiences of pregnancy and are running a study to try and improve the support available for families.

This is not a consent form. We are just asking for your permission to contact you in order to give you some more information about the study. If you are happy to be contacted, please fill in the form attached, place it in the envelope provided and return it to one of the researchers or members of staff.

*We would like
to contact you
about a
research study*

*We would like to
record your
consultation
with your doctor.*

We would also like ask your permission to voice record your consultation with the doctor today. We will not use your recording unless you give us permission to do so as part of the study. You will need time to read the information about the study and we don't want to rush your decision. As your appointment is today, we are asking for your consent to record the consultation, then you can take as long as you like to think about taking part.

If you wish to speak to us, or you wish to let us know whether or not you want to take part, you can contact the researcher :

Robyn Lotto, Department of Health Sciences, Princess Road West,
University of Leicester, Leicester LE1 6TP,

Tel: 0116 2525402, email: rrl6@le.ac.uk



PARENT INFORMATION SHEET



Experiences of Pregnancy

A study into the experiences of women and their partners managing risk and uncertainty in pregnancy

Pregnancy can be an emotional time and so we appreciate you taking the time to read this information. You are being invited to take part in a study looking at your experience of dealing with risk and uncertainty during your pregnancy.

Before you decide whether you would like to take part in the study it is important that you understand why the research is being done and what it would involve for you and your family. Read this information sheet carefully and discuss it with others if that would be helpful. Please ask us if there is anything that is not clear or you would like more information.

What is the purpose of the study?

The aim of this study is to understand how pregnant women and their partners deal with problems they face during pregnancy. If we can understand this better, we can try and improve the support that is available in the future.



What does the study involve?

With your permission we will voice record your consultations with the fetal medicine doctor. If you feel able to, we would also like to interview you when you feel ready. A researcher would be able to come out to your home and talk to you about your experience. If you prefer, we could talk to you in the hospital. We would like to voice record the interview. Everything you say will be confidential.

Please take time
to decide
whether you
wish to take part

Do I have to take part?

No. We will tell you about the study and go through this information sheet with you. It is up to you to decide whether or not to take part. You do not need to give a reason if you decide not to take part.

You do not
have to take
part in this
study.

You will not be identified in any report or publication.

Why have I been chosen?

You have been chosen because you have been referred to the fetal medicine unit. You may have had a test or a scan and the doctor or sonographer would like to have a second opinion.

Will my taking part in the study be kept confidential?

Yes. Any information that you give us will be kept strictly confidential.

With your permission your consultation and interview will be recorded. Recordings will be marked with a number only (never your name). These recordings will then be transcribed and any details such as names and places will be removed to make sure nobody can identify you from what you say. The transcripts are then studied by the research team. We sometimes like to quote things that have been said in the reports we write. If we do this, we always change details such as names and places to make sure nobody can be identified.

What are the possible risks and benefits of taking part?

We do not expect you to benefit from taking part but the information we get from this study will help improve the experience of families in the future. It will also help doctors and nurses to understand how you feel and what support you need. Some people like taking part in this sort of study, because it can give them an opportunity to talk about their experiences. If you became upset during the interview, we will stop straight away. We can also provide you with the names and contacts of a number of groups who can give you more support. All interviews will be handled with tact and sensitivity.

This study may not benefit you but could help families in the future

What will happen if I am interested in taking part?

If you decide to take part, a member of the research team will go through this form with you and give you the chance to ask questions. If you still want to continue, we will ask you to sign a form giving us your permission to listen to your recorded consultations. Some people may consent to doing an interview with the researchers as well. If this is the case, we will make an appointment to come out and see you at home or if you prefer, arrange to meet you at the hospital.

What happens during the interview?

If you consent to doing an interview we will ask to voice record it. The interview will be as long or as short as you like, but probably no more than 1 hour. You can stop the interview at any time.

What will happen to the results of the research study?

The findings from this study will be published as a report and may be used in professional journals and at conferences. They will also be fed back to a number of charities who support pregnant women and their partners. This research will also be used as part of Robyn Lotto's doctoral studies. The findings will be published on the University of Leicester website

<http://tinyurl.com/LeicUni-ExPres>

Information on how the study is going will be put on the website and a lay summary of the findings will be added at the end. If you prefer, you can ask Robyn Lotto for a copy.

Who is organising and funding the research?

The research is being funded by the University of Leicester. It is also organised and sponsored by the University of Leicester.

A lay summary of the results will be published on the website:

<http://tinyurl.com/LeicUni-ExPres>

Who has reviewed the study?

This study has been reviewed by the Nottingham Research Ethics Committee as well as the research departments of the University Hospital Trusts in Leicester and Nottingham

What if there is a problem?

If you have any concerns about any part of this research project please contact any member of the project team below. We will do our best to answer your questions. If you remain unhappy and wish to complain formally, you can do this through the NHS Complaints procedure.

Robyn Lotto, Department of Health Sciences, Princess Road West, University of Leicester, Leicester LE1 6TP, Tel: 0116 2525402, email: rl6@le.ac.uk

Dr Natalie Armstrong, Department of Health Sciences, Princess Road West, University of Leicester, Leicester LE1 6TP, Tel: 0116 2525402, email: na144@le.ac.uk

Dr Lucy Smith, Department of Health Sciences, Princess Road West, University of Leicester, Leicester LE1 6TP, Tel: 0116 2525402, email: lls1@le.ac.uk

Who can I contact for further information?

If you would like any further information at any time please contact Robyn Lotto at the address above.



Robyn Lotto is an experienced researcher who is currently undertaking a doctoral study with the Department of Health Sciences at Leicester University

Thank you very much for taking the time to read this information

CLINICIANS' INFORMATION SHEET

Experiences of Pregnancy Information for Clinicians

A study into the experiences of women and their partners when a severe fetal anomaly is suspected.

You are being invited to take part in a study examining how women and their partners make decisions in pregnancies where a severe fetal anomaly is suspected. Thank you for taking the time to read this information.

Please ask us if there is anything that is not clear or you would like further details.

What is the purpose of the study?

The aim of this study is to explore the decision making processes of pregnant women, from different socioeconomic groups, where a diagnosis of a severe congenital anomaly has been made or suspected. Recent research has demonstrated that rates of termination for congenital anomalies are much lower in more deprived areas. Although there is a significant body of quantitative evidence looking at factors that may predict women's decisions, there is no published research examining the reasons for this phenomenon.



We would like to record consultations and interview you.

What does the study involve?

With the consent of both you and your patient, we would like to record some of your consultations. We are also interested in your views about how women make decisions and would like the opportunity to interview you. The interviews will relate to your opinions and experience, not to any individual patient or consultation recorded.

Will my taking part in the study be kept confidential?

Every effort will be made to ensure that you are not identifiable from any data published. Your name, the unit you work in and any demographic details which may identify you will be withheld from any publication. There will be a limited number of interviews carried out with clinicians however, so if you wish to see your interview transcript prior to analysis, this will be made available to you.

Every effort will be made to protect your identity in every report or publication.

What will happen to the results of the research study?

The findings from this study will be published as a report and may be used in professional journals and at conferences. They will also be fed back to a number of charities who support pregnant women and their partners. This research will also be used as part of Robyn Lotto's doctoral studies. A copy of the findings will be made available to you once analysis is completed.

Who has reviewed the study?

This study has been reviewed by Nottingham Research Ethics Committee as well as the research departments of the University Hospital Trusts in Leicester and Nottingham.

Who is organising and funding the research?

The research is being sponsored by the University of Leicester. Researchers from the TIMMS and SAPHIRE groups are responsible for the study.

A copy of the findings will be made available to you once analysis is complete.

What if there is a problem or I would like further information?

If you would like any additional information, or have any concerns about any part of this research project please contact any member of the project team below.

Robyn Lotto, Department of Health Sciences, Princess Road West, University of Leicester, Leicester LE1 6TP, Tel: 0116 2525402, email: rrl6@le.ac.uk

Dr Natalie Armstrong, Department of Health Sciences, Princess Road West, University of Leicester, Leicester LE1 6TP, Tel: 0116 2525402, email: na144@le.ac.uk

Dr Lucy Smith, Department of Health Sciences, Princess Road West, University of Leicester, Leicester LE1 6TP, Tel: 0116 2525402, email: lks1@le.ac.uk