UNIVERSITY OF LEICESTER

Children of Parent's with Chronic Inflammatory Musculoskeletal Diseases: Experiences, Needs and Resources

by

Elizabeth Dorothy Pamela Hale

2017

A thesis submitted to the University of Leicester for the degree of

APPLIED PSYCHOLOGY: DOCTOR OF PSYCHOLOGY (PsyD)

Department of Neuroscience, Psychology and Behaviour

College of Medicine, Biological Sciences and Psychology

The Greenwood Institute of Child Health

The University of Leicester

April 2017

Declaration

I declare that this work is my own and has not been submitted for any other academic award.

Style Guide

This thesis follows the *Publication Manual of the American Psychological Association*, Sixth Edition (2010). American Psychological Association: Washington.

Thesis Word Count: 29, 269

Thesis Abstract

"Children of Parents with Chronic Inflammatory Musculoskeletal Diseases: Experiences, Needs and Resources"

Hale, E.D.[1, 2]

Department of Neuroscience, Psychology and Behaviour, University of Leicester, UK.[1]

Department of Rheumatology, Dudley Group of Hospitals, NHS Foundation Trust, Dudley, UK.[2]

Overview: Whilst each section of the thesis is required to stand alone, it is presented in the following order so that the thesis has a logical flow.

Literature review: Presents an overview of the rheumatic diseases and their impact upon parenting and families. We note the provision of patient education for parents, but not for their children. The comprehensive but selective narrative review focuses upon how adults and children conceptualise and understand illness, and explores the concept of normalisation as a potential family management strategy.

Service Evaluation: We sought to establish whether parents/grandparents would welcome the provision of appropriate patient education about rheumatic disease being made available to their children/grandchildren. Using a cross-sectional study design, a questionnaire was distributed to adult patients attending the local rheumatology service and members of four UK national rheumatology charities. Participants were strongly in favour of developmentally appropriate patient education for their children/grandchildren. Suggestions were made for content, format, timing and method of delivery.

Main Report: We sought to understand how the diagnosis and impact of parental rheumatic disease has been understood, talked about, and managed within families who have young children. Again, we asked for views about providing patient education for children. We were particularly keen to give children a voice in determining whether, and how, any resources designed for them should develop. Utilising a qualitative design informed by an interpretivist framework, we employed semi-structured interviews and visual data collection methods. Eleven families with children aged between seven and 11were recruited from the local rheumatology service. Interviews and visual data were analysed using thematic analysis. The results are discussed within the concept of 'normalization'. Implications for clinical practice and further research are highlighted.

Critical Appraisal: Contains a critique of both the research process and the methodology used.

Acknowledgements

As I write this, reflecting upon the last few years, there are many people I would like to thank for enabling me to carry out my research and produce this thesis. I will thank you all, but some deserve a particular mention.

Firstly, my thanks and gratitude go to Professor Panos Vostanis, Dr Michelle O'Reilly (University of Leicester, Department of Neuroscience, Psychology and Behaviour) and Professor George Kitas (The Dudley Group of Hospitals NHS Foundation Trust) who are the exceptionally talented supervisors who inspired me and showed me nothing but kindness, support, encouragement(and endless patience) throughout.

Thanks of course must go to Arthritis Research UK for funding part of the research project. My colleagues at the Research and Development Department, and the Rheumatology Department at the Dudley Group of Hospitals NHS Foundation Trust have provided all manner of administrative support and lively discussions during research meetings, that have helped to facilitate and shape the research. Particular thanks to Margaret Marriott, Helen Hollis, John Walters, Ben Watkins and Rebecca Storey for always having some idea of where something is stored both physically and electronically!

My research participants receive heartfelt thanks, for completing questionnaires and attending interviews with me. The support of the National Rheumatoid Arthritis Society; the National Ankylosing

Spondylitis Society and the Raynaud's and Scleroderma Society in hosting the questionnaire on their website was invaluable. Similar support was shown by Lupus UK who posted questionnaires to members on my behalf. It was a privilege working with the families who gave up their time to attend interviews with me at the hospital, and a delight to meet the children who were so invested in the project

Thanks also go to Clinical Nurse Specialist Sharon Petford and Senior Nurse Shirley O'Hare who were recruiters extraordinaire, giving information leaflets to their patients about the research study. Sharon particularly, deserves my gratitude for her help with thematic analysis discussions, and pushing me to explain "why"?.

The research group at the University of Leicester Department of Neuroscience, Psychology and Behaviour was a joy to be part of, and was a demonstration of how researchers from very different disciplines, epistemologies and countries can collaborate effectively. I always came away feeling inspired.

Although they may not be aware of it, Professor Alice Witney, Mary Hall (SpR Public Health), Dr Sue Peacock, Dr Sheila Bonas, Professor Sarah Hewlett, Dr Marianne Morris, Professor Karen Rodham, Dr Gail Parsons, Jane Taylor, Catherine Beasley, Reverend Mark Stobert, Dr Jet Veldhuijzen van Zanten and my dear friend Dr Gareth Treharne always seemed to have just the right thing to say when I needed it, and provided endless tea, biscuits, expertise, and words of encouragement.

Finally to my dearest husband Mark, my children David and Daisy, to Jana and beautiful baby Olivia thank-you. You are my everything.

This is for you.

Contents

Chapter Three: Research Report

"We do try and do everything that other families would do":
How families manage and talk about parental rheumatic disease.

Chapter Four: Critical Appraisal

List of Tables:

Chapter One

Chapter Two

Chapter Three

List of Figures:

Chapter Two

Chapter Three

Chapter One

Abstract for Literature review

"Providing information to the children of parent's with chronic rheumatic diseases: How do adults and children understand health and illness, and how do families attempt to 'normalise' this within the family? A narrative review"

Hale, E.D.[1, 2]

Department of Neuroscience, Psychology and Behaviour, University of Leicester, UK.[1]Department of Rheumatology, Dudley Group of Hospitals, NHS Foundation Trust, Dudley, UK.[2]

Objectives:- Parent's with chronic rheumatic diseases are signposted to patient education materials in order to help them understand and manage their condition. There are no similar resources for their children, nor any resources to help them talk to their children about their condition. With this in mind, our objective was firstly, understand how adults and children conceptualise health and illness, secondly, how they manage to live 'normal' lives within their family settings.

Design:- A comprehensive but necessarily selective narrative literature review, only focusing upon illness perception models for adults; cognitive-developmental models and perception models for children; and the concept of 'normalisation' within family management strategies of illness were reviewed.

Methods:- Electronic database searches were performed in PubMed (Medline), CINAHL, PsychINFO, (1979-2012); Google Scholar, Web of Science (including SCIE;CPCI-S; CPCI-SSH) (1970-2013) and periodically updated. Broad date ranges, search terms and no limits on study design ensured inclusivity. Studies were selected if they were English language, full text articles, and relevant to the research topics. Primary snowballing of reference lists were used to obtain additional relevant studies. Literature was evaluated using the Critical Skills Appraisal Network (CASP) framework. Editorials, Reviews, Commentaries, Concept/Position papers, patient and charity magazines, were discarded.

Results:- Reports of 20 studies were included. Studies were generally limited by cross-sectional designs and small sample sizes. Qualitative studies benefited from a range of designs. The Illness perception model demonstrates that the way adults and children think about illness may directly affect outcome measures relatively independently of coping. Cognitive developmental stage models, coupled with illness experience suggest an additional means of gauging where a child's understanding may be. Families' attempts to be 'normal' in the face of chronic illness are complex and shifting.

Conclusions:- The synthesis of evidence provides useful information for the development of patient education for the children of parent's with chronic rheumatic diseases.

Chapter One

1.0 Introduction

This chapter will provide firstly, some background information about the rheumatic diseases and their impact upon both individual and family life, in order to place the subsequent literature review in context. Secondly, a comprehensive and selective narrative review of the relevant literature will be presented, within the necessary confines of the space available. In conclusion, we will look at possibilities for future research.

Background information

1.1 The Rheumatic diseases:-

Musculoskeletal conditions including the rheumatic diseases affect over a billion people worldwide (Woolf, 2012). The rheumatic diseases are a group of over 200 conditions that affect the musculoskeletal system. Once diagnosed, these conditions will be life-long and range in severity from mild to life-threatening. Some of the rheumatic diseases are known as 'inflammatory' diseases. These are auto-immune systemic conditions that may affect not only the skeletal system, particularly the joints, but also the connective tissues and internal organs. The prevalence of the rheumatic diseases increases with age, and often appears in both sexes during young adulthood, when many are thinking about becoming parents, or may already be parents (Barlow, Cullen, Foster, Harrison & Wade, 1999; Jolly, 2005; Madsen, Jensen & Esbensen, 2015). However, there remains a common misperception

that rheumatic diseases only affect people of mature years (Primholdt, Primdahl & Hendricks, 2016).

The most common inflammatory joint disorders are Rheumatoid Arthritis (RA), Ankylosing Spondylitis (AS) and Psoriatic Arthritis (PsA), with Systemic Lupus Erythematosus (SLE) one of the most common inflammatory connective tissue disorders (Hill & Ryan, 2000).

1.1.1Rheumatoid arthritis:-

RA is the most common chronic inflammatory joint disease in the United Kingdom (UK), affecting approximately 1- 3% of the general population, with women affected more often than men in a ratio of about 3:1 (Symmons, 2005).

RA can have a sudden onset. Individuals may feel generally unwell with pronounced fatigue. Characteristic of the disease is morning joint stiffness, with symmetrical pain and swelling in the small joints of the hands and/or feet and wrists (Hameed & Akil, 2009). As it is a systemic disease, it has the ability to affect the heart, lungs, eyes and renal systems.

Treatment goals focus upon controlling the progression of the disease by reducing inflammation in order to prevent damage to the joints (Pullman - Mooar, 1999). A characteristic of the inflammatory rheumatic diseases is that they fluctuate over time, having periods where the disease is more active (a 'flare') followed by periods of quiescence.

1.1.2 Psoriatic arthritis:-

PsA is characterised by the co-existence of inflammation in and around the joints with the skin condition psoriasis. Prevalence has been estimated at between 0.3 and 1% of the population, affecting men and women equally (Gladman, Antoni, Mease, Clegg & Nash, 2005). Usually people will already have psoriasis before developing pain and stiffness in multiple joints. Dactylitis, enthesitis, and pain and stiffness in the neck or back are all common. Treatments for PsA may have a symbiotic effect in that treatment for joint inflammation may improve the skin and vice versa (Arthritis Research UK *Psoriatic arthritis*, 2011).

1.1.3 Ankylosing spondylitis:-

The spondyloarthropathies are a group of conditions that share similar features. Of these, AS is an inflammatory arthritis that affects the joints of the spine, often causing pain and stiffness in the neck and lower back area. Pain in the sacroiliac joints is common (Braun, et al. 2011).

AS predominantly affects men, with a ratio of approximately 3:1, and has an early age of onset, typically in the late teens to early twenties. One of the features of the condition is the growth of bone from the sides of the vertebrae in the spine and sacroiliac joint following periods of inflammation when the disease is active. This bony outgrowth can eventually cause the vertebrae to fuse together (ankylose), leading to difficulties with mobility and the risk of severe damage such as spinal fracture in the event of a fall or accident. The primary goals of treatment

are to reduce inflammation with drug therapy and preserve mobility through daily stretching and strengthening exercises.

1.1.4 Systemic Lupus Erythematosus:-

SLE is a complex multi-system autoimmune disorder. Joint pain is common (arthralgia) as are skin rashes and photosensitivity to sunlight. Individuals may develop a classic malar ('butterfly') rash across the cheeks of the face after exposure to sunlight, even in winter, and high factor suncreams (over factor 50) are a daily requirement. Hair thinning and hair loss (alopecia) may also occur (Al-Mossawi & Gunawardena, 2012).

SLE typically onsets during early to mid-adulthood, although 15 - 20% of all people with SLE develop the condition under the age of 17 (Ioannou, 2014). SLE predominantly affects women, with ratios estimated in the range of 9 to 12:1, and also disproportionately affects people of African-Caribbean, Chinese and Asian origin compared to Caucasians (Al-Mossawi & Gunawardena, 2012; Danchenko, Satia & Anthony, 2006; Hay & Snaith, 1995).

Although relatively uncommon, it is a serious condition and mortality rates are higher than the general population (Gabriel & Michaud, 2009; Lateef & Petri, 2012). Between 30 - 60% of patients will develop some degree of heart, lung and/or kidney involvement, and individuals may have disturbances of the haematologic, immunologic and neurologic systems (American College of Rheumatology, 1997).

2.0 Impact of rheumatic diseases:-

Perhaps not surprisingly, the psychosocial well-being of individuals and their families can be severely challenged by a rheumatic disease. People with SLE are reported to have worse health related quality of life (HQOL) when compared to people with other chronic diseases (such as diabetes), and this may be particularly so for people with poor body-image related HQOL (Jolly, 2005; Jolly et al., 2012).

Levels of depression and anxiety are reported to be higher than the general population in people with RA, and can persist over time and reduce treatment effectiveness and other health outcomes (Matcham, Norton, Scott, Steer & Hotopf, 2016). The ability to perform valued activities has been found to increase levels of depression, although this may be mediated by how it is viewed. Remaining satisfied with what you can do, although reduced, has been shown to have no effect on depression (Katz and Neugebauer, 2001). It is possible that positive support from family and friends can help to reduce feelings of depression (Riemsma et al., 2000).

Studies have indicated that children within families where a parent has a rheumatic disease may also experience poorer psychological well-being than children who have 'healthy' parents, (Hirsch, Moos & Reischl, 1985). A review by Armistead and colleagues suggested that 'disrupted parenting' (p.418) is the factor that influences how well children within a family fare, and involve things like changed family routines and unavailability of the parent due to symptoms (Armistead, Klein & Forehand, 1995).

Feeling guilty about not being the parent you intended to be, or worries about children becoming young carers are common (Hewlett et al., 2012; Meade, Sharpe, Hallab, Aspanell & Manolios, 2013). For fathers, inability to play with their children due to pain or fatigue can impact on their identity of what a man could or should be expected to do (Madsen et al., 2015), and has been described as 'I let you down again syndrome' (p.145. Barlow et al., 1999).

A conceptual model developed by Rolland (1999), suggests that families struggle with the impact of a chronic health condition as it impacts upon the personal identity of the adult, and subsequent identity of the family. Often families respond to this by trying to be 'normal'; redefining what this means as new challenges arise and time moves on. In other chronic health conditions, parents are often advised to keep family routines as normal as possible, and integrate communication with their children along the way when changes occur (see Breast Cancer Care, 'Talking with your children about breast cancer' (2007, revised 2014).

Talking to children about a parent or grandparent's rheumatic disease appears to be highly valued, but one that has been described as difficult (Grant, Foster, Wright, Barlow & Cullen, 2004). Parents may feel at a disadvantage as they often do not know how to do this, and may not understand the condition well enough themselves to convey complicated information appropriately (Madsen et al., 2015).

Adults diagnosed with a rheumatic condition are given verbal information and advice by their multidisciplinary specialist healthcare

team, and should be given or signposted to written patient education leaflets and relevant websites. In the United Kingdom (UK), these are produced by charities such as Arthritis Research UK (ARUK), (for example, "Rheumatoid Arthritis" ARUK, 2011) and other disease-specific charities such as The National Rheumatoid Arthritis Society (NRAS) and Lupus UK. These leaflets give advice about specific diseases, treatments and some psychosocial advice.

However, there are no age-appropriate information or educational materials available specifically for children within these families, in order to help them to understand or talk to their parent about their rheumatic condition. There are also no similar educational materials to help parents (or grandparents) to talk to their children appropriately. Electronic searches did not reveal any peer reviewed literature that has focused upon providing education resources or support for children who have a parent/grandparent with a rheumatic disease, whilst conversely, there is a wealth of information focusing on providing information and support to children with juvenile rheumatic disease, and their families. The only sources of information currently available are accessed via specific charities or other organisations, and it is difficult to assess the quality of the evidence-base that they are built upon, or who has been involved in their development. For example, a company called *Medikidz* has produced a series of 'comic books' for children about a range of health conditions and health-related procedures. In the rheumatic diseases there are currently books on RA, PsA and AS. These are written by doctors and professional medical writers, and peer reviewed

by clinicians (Chilman-Blair & DeLoache, 2012). However, their target age is unclear, and are often based upon a single “case study”. Children themselves appear to have had little input into their formation.

Review of Literature

3.0 Purpose:-

It is unclear how the diagnosis and impact of parental rheumatic disease has been understood, talked about, and managed within families who have young children, or whether providing information and /or support for children about parental rheumatic disease would be useful. In order to consider research in this area, the aim of this short review was to focus upon two related areas. Firstly, how do adults and children understand health and illness? (illness perceptions) and secondly, what do we know about the impact of ill-health and the attempts to 'normalise' this within the family?

4.0 Method:-

A comprehensive but necessarily selective literature review was performed in PubMed (Medline), CINAHL, PsychINFO, (1979-2012); Google Scholar, Web of Science (including SCIE;CPCI-S; CPCI-SSH) (1970-2013) and periodically updated. Broad date ranges and search terms were used to be as inclusive as possible. No limits were set on study design. Key words were used, informed by the extant literature, for example: 'illness perceptions';' illness representations'; 'self-regulatory'; 'common-sense model'; 'normalisation'. Studies were selected if they

were English language, full text articles, and relevant to the research topics. Older, seminal studies were included.

Primary snowballing of reference lists (see Cramp et al., 2013, p.240) of identified studies were used to obtain additional relevant studies. Journal alert feeds, for example, *British Journal of Health Psychology; Musculoskeletal Care; Rheumatology; Health Psychology Update;* provided current early content viewing. Additionally, personal searches at the local NHS Trust gave some access to relevant journals and books. The Critical Skills Appraisal Network (CASP) provided useful checklists to help evaluate studies (www.casp-uk.net/casp-tools-checklists). Editorials, Reviews, Commentaries, Concept/Position papers, Patient and Charity magazines, were discarded.

5.0 Results:-

The literature review presented below has been selected for relevance to the areas of interest, and includes seminal studies. Sixty-three studies were identified relevant to section 5.1; 53 studies for section 5.2 and 25 for section 5.3. Results focusing on young carer literature were discarded. Where possible, work has been selected that focuses upon the rheumatic diseases. In the interests of brevity, studies included are shown in Tables 1-3 which show notable limitations and key/notable results.

5.1 How do adults understand illness?

The illness representations model (also known as the illness perceptions model) grew out of the health-related compliance research

of the 1960's (Leventhal, Jones & Trembly, 1966). How people responded to a health threat was not always straightforward, as fearful messages sometimes produced avoidance of the recommended action. In later work looking at how people understood and engaged with treatment for hypertension, using structured interviews coded by students (to avoid interview bias) into *a priori* hypothesized domains, researchers suggested that individuals will act to reduce health risks dependent upon their subjective 'common-sense' ideas (representations) of the health threat (Meyer, Leventhal & Gutmann, 1985). Leventhal and colleagues suggested individuals form representations about their illness from sources like the media, family, friends and medics, and integrate these with past experience of illness and their current symptoms (Leventhal, Meyer & Nerenz 1980). They proposed five dimensions to the model: *identity* - the labels and symptoms of the illness; perceived *causes* - including what might exacerbate or remit it; perceived *consequences* - the impact on a person's life such as loss of work; beliefs about *controllability* or *curability* of the condition and *time line* - beliefs about whether the illness is acute/episodic/chronic (Meyer et al., 1985; Pimm & Weinman, 1998).

A key proposition of the model is that it is self-regulatory (S-R), that is, how you think about your illness (what you understand or believe) influences how you cope with it, which in turn influences various outcomes. People self-regulate by appraising outcome and coping efforts and respond accordingly. This casts the individual as an active problem-solver, in a dynamic model that can respond to change over

time (Nerenz & Leventhal, 1983). The work that Leventhal and his colleagues undertook often included structured interviews in order to access participants perceptions (Leventhal et al., 1966; Meyer et al., 1985). Whilst the reporting detail of these is deficient by today's standards (no or little methodological detail) considerable research has ensued investigating the various components of the model and their impact on coping and outcome; therefore these studies must be seen as the seminal studies in their field. Indeed, later work by Moss-Morris, Weinman, Petrie, Horne, Cameron & Buick, (2002), returned to the original qualitative concept work, which had proposed that a cyclical timeline was important, and realised its usefulness.

However, research in the field of illness representations has grown enormously since the development of the illness perceptions questionnaire (IPQ); designed to give a quantitative assessment of the five domains of the model (Weinman, Petrie, Moss-Morris & Horne, 1996a). The IPQ has been revised (IPQ-R) and new items have been added, including *emotional representations* - how people respond emotionally to illness and *illness coherence* - how people understand and make sense of their illness (Moss-Morris, et al., 2002). We will specifically review some of the work using the IPQ and IPQ-R, see Table 1.

The relationship between illness perceptions, coping and outcome appears to be complex. In a cross-sectional study looking at illness perceptions, coping and adjustment in chronic fatigue syndrome (CFS) Moss-Morris, Petrie & Weinman (1996) suggested that illness

perceptions and coping were related logically as expected from the S-R aspect of the model. People who believed they had some control coped more positively with active coping efforts, planning and reinterpretation, and less behavioural disengagement. A strong illness identity, beliefs in a chronic time line and believing CFS has serious consequences were related to emotion-focused coping (venting emotion, seeking emotional social support and disengagement from stress). However, the study showed that illness perceptions were more strongly associated with adjustment and well-being than the coping subscales. This means illness perceptions affected outcome *independently* of coping.

Confirmation of this was seen in a study by Heijman (1999) that aimed to describe the illness perceptions of people with Addison's disease, and investigate how illness perceptions and coping were related to adaptive functioning. As in the Moss-Morris et al., (1996) study above, illness perceptions were better predictors of adaptive outcome than were the coping scores. An alternative explanation is that the coping measure used might have been too generalised, and that coping may be more effectively measured if conceptualised in behavioural terms.

Studies have also shown that illness perceptions are not related to disease activity or severity. Carlisle, John, Fife-Schaw & Lloyd (2005) found that illness perceptions seemed independent of current disease status, in a study looking at the relationship between illness perceptions, coping strategies and outcomes in RA. Similar results were found by Graves Scott, Lempp & Weinman (2009) who found that illness

perceptions in RA were associated with disability and quality of life, but not explained by disease status. Patients with active RA did not necessarily view their disease as worse than other people's, but if they believed their RA would have negative consequences they had worse physical, social and mental functioning.

As coping does not necessarily seem to mediate outcome, contrary to the S-R model's original proposition, researchers have moved to investigate how illness perceptions impact on many different types of outcomes directly, independently of coping. A study comparing illness perceptions and psychological distress in patients with either primary Sjögren's syndrome (pSS), RA or SLE, and whether their health related quality of life (HRQoL) was similar, found that patients with pSS attributed more symptoms to their disease and also had little understanding of their disease compared to the other groups. HRQoL and depression scores were similar however across groups. Additionally, pSS patients had stronger beliefs in the consequences of their condition (more serious), which were more strongly correlated with physical HRQoL than pain. It may be possible therefore, that certain disease groups require information targeted in a different way than others. Better information provision by healthcare practitioners could help to achieve a more coherent understanding of the condition and better HRQoL (Kotsis, Voulgari, Tsifetaki, Drosos, Carvalho & Hyphantis, 2014).

High consequences scores have been shown to have predictive power in other outcome measures. In a study looking at illness

perceptions in multiple sclerosis (MS) and their relationship to outcomes, participants who also had beliefs that the consequences of MS were serious, had higher illness intrusiveness scores, greater impairment in physical functioning, higher depression and anxiety and lower self-esteem (Vaughn, Morrison & Miller, 2003). The sample in this study were generally low in psychological distress however, and this may have been a function of them being recruited from a health psychology service. The effect of psychological interventions may have affected the results as it focused upon education about MS and addressed adjustment issues to a diagnosis of MS.

5.2 How do children understand illness?

Research into children's understanding of health and illness has broadly followed three theoretical approaches. Early work focused upon cognitive-developmental approaches derived from the work of Piaget (see Shaffer, 1996), which propose children's understanding of illness follows predictable sequences according to their developmental stage. Researchers in this area began to realise however, that children's cumulative experiences of illness also played a major role in shaping their understanding, often outside the boundary of the cognitive-developmental 'stage' they were expected to be at (for example, Hergenrather & Rabinowitz, 1991), and have taken a more 'experiential' approach. Current work integrates both approaches to a degree, and has focused upon the illness perceptions approach, broadly following the domains proposed in work with adults, as above (Paterson, Moss-Morris & Butler, 1999), see Table 2.

One of the first, and still often cited, studies investigating children's understanding of the causes of illness was published by Nagy (1951). Conducted in early post-war Europe, Nagy compared the beliefs of children aged 3-12 in Hungary with children aged 8-11 in the UK. The study is poor methodologically and attempts to make some comparisons that are not delineated nor discussed, for example, children in the UK study were drawn from two schools, one with 'good average social and cultural background' and the other with 'poor social and cultural background' (pp. 8-9). Nonetheless, the study suggests that children in both countries followed expected developmental stages, and that all children between the ages of six and eleven believe that all illness comes from infection. Children could relate cause and effect, and with maturity develop more sophisticated explanations of infection.

Picking up this theme, other studies have looked at whether children's concepts of illness change with development and also whether they are influenced by the views of their parents, becoming more like theirs over time (Campbell, 1975). To ensure illness was 'apparent', children aged between 6-12 years currently experiencing a short stay in hospital were interviewed and asked what the difference was between well and sick, and how they knew when they were unwell. The nature of their hospitalization is unknown, and details of their health history, although apparently collected from mothers, is not reported. Using thematic content analysis, Campbell proposed that children's concepts of illness fall into five distinct categories; Somatic states;

objective states; specific named states; psychosocial understandings and restrictive concepts i.e. 'If I just have a cold, I'm not sick'.

As children got older, they used more themes to describe illness, their explanations becoming more sophisticated. Older children could give more explanations of specific diseases and diagnoses whereas younger children were much vaguer and often described feelings instead. When specific mother-child pairs were compared, there was no indication that children's illness concepts were similar to those of their parent, however, group comparisons indicated that children's concepts did grow to correspond with their mother's over time. Campbell suggests that this evidences a developmental trend in concept content. Of note, however, was the contribution of illness experience and age to a child's illness concept. Children who were younger (under 9) whose health was judged poorer, had the least sophistication in their concepts, whilst the reverse was true for older children with poor health, as their illness concepts were much more sophisticated. This means that experience of ill health plays a role, but is dependent upon the child's age.

Similar results have been found with hospitalized children aged between four and 14, experiencing either acute self-limiting illness such as pneumonia or cellulitis, or chronic major illness such as cancer or cystic fibrosis. Both age and experience of illness contributed to understanding of illness, and the researchers caution against assuming that children's understanding is boundaried by cognitive-developmental stages and age (Crisp, Ungerer & Goodnow, 1996).

Research that has used a cognitive-developmental approach has tried to determine whether children's understanding of illness broadly falls within the stages known as prelogical (two to seven years); concrete logical (seven -11 years) or formal operational (11-12 years). One of the most influential studies in this area was able to expand this theoretical framework, and proposes additional categories within each stage: *phenomenism* or *contagion* (prelogical); *contamination* or *internalization* (concrete-logical); *physiologic* or *psychophysiologic* (formal-logical) (Bibace & Walsh, 1980).

Children aged between four and 11 years were interviewed about what they knew about common illnesses, explanations about their own personal illnesses, or illnesses of friends or family. Using this information the researchers gave examples of how children gradually distinguish the self and other, and provide less mature/more mature examples for each category. Of interest is the clear relation of this work to providing education to children in a way that they will understand about their health. The authors note that children's books about illness, hospitalization and medical personnel are written by adults and assume an adult view of what children might be expected to know, rather than what empirical research suggests they do actually know. They further suggest that information could be more effectively presented about causes of illness and personal control of health if given within the correct stage of cognitive-development. This study has remained one of the most durable in its categorization of illness understanding related to age,

and may be a useful 'guide' in the formulation of patient education for children.

When providing information intended to help children understand illnesses or health conditions that *other* people might have, the situation may be more complex than assumed however. The more observable an illness is, the less attractive it is seen to be (Potter & Roberts, 1984). Providing a description and explanation of an observable illness may not achieve much improvement in children's attitudes to the person experiencing it, with explanatory information *decreasing* perceived attractiveness rather than improving it (although the findings were marginal and must be treated with some caution).

One of the criticisms of both the cognitive-developmental and experiential work is that it often focuses on the causes of illness and does not go far beyond this. As reviewed above, adult perceptions of illness have been shown to encompass five main and two additional dimensions in their explanations. Research has moved to determine whether children also use broader domains of thinking about health and illness, that are not conceived as being boundaried according to age, and which can account for experience and change over time. This is interesting, as the seminal studies by Leventhal et al., (1980) had originally proposed that adult understanding of illness was partly founded upon past experience, and yet this had been ignored in the early child-focused literature.

In a study looking at children's illness perceptions of colds and asthma, researchers interviewed children aged between seven and 14 years along the domains of *identity, cause*, *consequences*, *control/cure* and *timeline*, with the additional dimension *prevention* (Paterson, Moss-Morris & Butler, 1999). Children did indeed show thinking that could be attributed to these domains. Furthermore, children with experience of asthma had more sophisticated thinking within the domains of cause, chronicity (timeline), and controllability but not within the consequences and prevention dimensions. This means that children may not be aware of how to prevent an attack of asthma, nor concern themselves with actual or possible consequences. Arguably, this is important as information for children could be organized along these domains, and emphasize areas that might be of specific interest or worthy of intervention.

Similar work has been carried out from another health promotion perspective, using semi-structured interviews with 'normal' weight and 'overweight' children aged between seven and 12, in order to understand their constructions of obesity (Babooram, Mullan & Sharpe, 2011). Although perhaps skewed by the fact that the overweight children were sampled from a group actively seeking 'appetite awareness' training (arguably a sub-group in themselves), some results are notable. Firstly, the children were all able to conceptualise obesity within the domains of the illness perceptions model and all endorsed the idea that exercise was the most useful 'cure' for obesity. The authors note that weight management requires other changes however. The overweight

children who were waiting for appetite awareness training, might not appreciate that a number of changes would be necessary.

Whilst these studies have focused upon children's understanding of illness, or experiences of personal illness or conditions likely to influence personal health, some researchers are now extending this to children's understanding of parental illness. Content analysis of a small semi-structured interview study carried out with children aged between seven and 14, who had a parent with MS, suggested that children believed their own, or other people's behaviour affected the course of their parent's illness (Cross & Rintell, 1999). Whilst the analysis is extremely limited, with no age related information or quotations from the data, the analysis easily fits the domains of the illness perceptions model, (although not analysed in this way). Children spontaneously talked about parental mood changes and distress, showing an awareness of the emotional representations aspect of the model. The authors suggest accurate information is necessary to correct a number of misperceptions that children held. Additionally, they recommend that parents talk to their children openly about their condition and use information which may need updating as the child develops.

5.3 Managing parental illness within the family

One of the ways in which researchers have sought to understand how families manage chronic illness within the family is through the concept of 'normalisation'. Normalisation is the process by which families seek to live their lives as 'normally' as possible within the experience of chronic illness. Having a family member with a chronic

illness, particularly one that may be disabling, has implications for the whole family unit. Hilbourne (1973) suggested that to be disabled is to be judged 'different' by society, and people will move to mitigate this as far as possible by living as normally as possible. Furthermore, the family of the disabled lies open to 'courtesy stigma' (p.502), simply by being the family, and may in themselves become disabled by the effects of caring for the sick family member.

Much of the research into normalisation initially focused upon how families cope when a child has chronic illness, although more recent work has investigated adult chronic illness. We shall focus upon five studies using a variety of qualitative methodologies, four of which included or were focused on rheumatic diseases.

One study whose sample combined both parents managing a child's chronic illness, and adult's managing their own, spouses, parents or siblings chronic illness, investigated how families constructed a story of life 'as normal' (Robinson, 1993). One important means of preserving normality, was to continue to do 'normal things'. This might include work where possible, or maintaining hobbies, clubs and shopping. Keeping to routines was a key aspect of normal life.

Over a period of time, families minimized the significance of the chronic illness in their lives and chose instead to highlight aspects of their life that illuminated a view of 'life as normal' rather than aspects of life that indicated otherwise. Robinson refers to this as a 'normalisation lens' or frame. For example, treatments carried out at home may not be

the norm for most people, particularly if carried out by a child. However, if acknowledged but minimized, they do not upset the overall frame of normality. Families can identify what *is* different, but not be overwhelmed by it (Knafl & Deatrick, 2002). Although normalising may be difficult, Robinson suggests that her participants show that trying to be a normal family is a positive coping strategy (Robinson, 1993).

Other families have also suggested that keeping normal routines and membership of social groups is important to preserving normal life. This may be particularly so for families with stigmatising illnesses like HIV/AIDS (Rehm & Franck, 2000) where disease status disclosure is guarded carefully. Careful monitoring of health and taking care of yourself is also a means of preserving a normal life, where health can be foregrounded and illness left in the background (Rehm et al., 2000). However, in other instances the reverse may be true, where monitoring simply serves as a reminder that normality will never be truly restored such as with post-lung transplant patients and rheumatology patients requiring infusions and blood test monitoring (Dabbs et al., 2004).

Adults with RA have also indicted that 're-normalization' may be necessary to adapt to lower expectations of what is now possible due to deteriorating health. This may be a means of preserving psychological well-being. If an individual can 'reconstruct' normality to accept change, they may be able then to find satisfaction in achievements within the new normality, that are far below the old (Locker, 1983, p.30).

In the last ten years, with the advent of new treatments in rheumatology, individuals and their families have a much improved

prospect for disease control and reduced disability compared to the participants in the above study (Locker, 1983). However, treatments often fail after some time, or are withdrawn due to side effects. This means fluctuations in disease states are still common. Sanderson, Calnan, Morris, Richards & Hewlett, (2011) suggest that people actually hold multiple normalities (typologies) which fluctuate and dominate as treatment and health changes. Broadly these fall into the non-normal states of *disrupted normality*; *struggling for normality*; and *fluctuating normality* and the normal states of *resetting normality*; *returning to normality* and *continuing with normality* (the latter being reserved for people only minimally affected by their disease). The individual and family have to respond to the situation at the time and constantly reframe where their normality lies. Not surprisingly, some families will feel that any kind of normalcy is unattainable by pre-disease onset standards (Knafl & Deatrick, 2002).

For young adults with rheumatic disease, treatment decisions in themselves have been shown to depend upon the desire to have a normal life. It is important that healthcare professionals understand the particular issues for young adults who will be aspiring to be like their peer group, and do as they do. Parenthood and managing a family with a rheumatic condition will also be part of this (Hart, et al., 2016). Healthcare professionals have been cautioned to listen carefully to their patients attempts to live a normal life, and assist them to weigh up the costs and benefits of any coping strategies they employ (Robinson, 1993).

5.4 Conclusion

In summary, this review enables us to understand some of the ways in which adults and children may understand illness and one of the ways in which they may respond to, and manage illness within the family. To our knowledge there has been no research in this area within the rheumatic diseases that pulls these threads together with a view to providing patient education materials for the children within these families. Future research should establish firstly, whether such resources would be welcomed, and secondly how families talk about parental rheumatic disease. The involvement of young children would enable them to have a voice in developing resources.

Table 1. Summary of studies investigating illness perceptions in adults

First author	Year published	Sample (location, size, recruited from)	Assessment of illness perceptions	Other measures	Investigated coping	Limitations	Key result
Moss-Morris	1996	New Zealand N=233 Chronic Fatigue organization	IPQ Postal questionnaire	-COPE scale -MHI-5 mental health scale -Vitality scale -SIP sickness impact profile scale	Y	- Cross-sectional - No comparison to non-responders - Self-reports of CFS	Key study proposing IP affects outcome independently of coping
Heijman	1999	Netherlands N= 63 National Addison's disease society	IPQ Postal questionnaire& comprehensive interview	-Utrecht coping questionnaire -SF36 items as outcome=physical and social functioning; mental health; vitality	Y	- Cross sectional - Small sample - No comparison to non-responders - Motivated & well-informed sample - Society emphasizes biological explanations for illness	IP stronger predictors of outcome than coping
Carlisle	2005	UK N=106 Hospital out-patient clinics. RA patients	IPQ Observational; Questionnaire	GHQ-12 mental health HAQ physical functioning Pain VAS	Y London coping with RA scale	- Cross sectional - Small sample - Coping measure possibly too broad and not specific	Illness perceptions contribute to outcomes

Graves	2009	UK N=125 Hospital out-patient clinics. RA patients	IPQ-R Questionnaire	SF-36 for QoL HAQ=physical functioning DAS= disease activity in RA	N	- Cross sectional - small sample	Confirms illness perceptions cannot be explained by disease severity (Carlisle et al., above)
Kotsis	2014	Greece N=pSS 57 N=SLE 75 N=RA 199 Hospital out-patient clinics. Patients with primary Sjögren's syndrome; RA; SLE	B-IPQ (short form version of IPQ)	HRQoL SCL90R symptom distress checklist PHQ-9 Pain VAS DAS28 for RA disease activity score SLEDAI for SLE disease activity score	N	- Cross sectional - pSS sample small	pSS patients understand their disease less well than other rheumatic disease groups
Vaughn	2003	UK N=99 Past referrals to a health psychology dept. Patients with Multiple Sclerosis	IPQ	Illness intrusiveness rating scale ADL - activities of daily living HADS - anxiety & depression scale Rosenberg self-esteem scale	N	- Cross sectional - Small sample - Participants had received input from psychology service - psychoeducational or individual basis	Illness perceptions contribute to outcomes

Table 2. Summary of studies investigating children's understanding of illness

First author	Year published	Sample (location; age of child(ren); size; recruited from)	Method of assessment	Perspective/Focus of study	Limitations	Key/notable result
Nagy	1951	Hungary N=100 Age 3-12 School x3 UK N=250 Age 8-11 Schoolx2	Interviews Drawing Essay (20 mins)	Causal understanding of illness	- Cross sectional - Early post-war - Methodologically flawed - Consent? - Leading questions - Judgemental: 'poor' vs 'good' ' similar social & cultural background'	- Explanations fit with cognitive developmental theory - With maturity children can relate cause & effect
Campbell	1975	US N=264 Age 6-12 (plus mothers) Stratified age: Age 6-9.5 Age 9.5-12.5 During short- term hospitalization (median 5days)	Interview x 2 Hospitalization ensures health & illness of concern	Cognitive-developmental - Developmental change in illness concepts - Do illness concepts become more like adults with age	- Cross-sectional - Sample bias=higher SES & education of father - 'intact white families'	- Children's descriptions of illness become more sophisticated with age - Concepts become more like adults with maturity - No evidence concepts come from mother's views except social-psychological 'grouchy'; 'irritable' etc - Illness experience has an effect, but limited by age of child - Age + experience influence illness concept development

Biba ce	1980	US N=72 Age 4-11 Stratified: Age 4x24 (12M x12F) *Assumed pre-logical* Age 7x24 (12M x12F) *Assumed concrete logical* Age 11x24 (12M x12F) *Assumed formal operational* School recruitment	Structured Interviews 12 sets of questions	Cognitive-developmental - Children's understandings of illness; cause; cure - Ho = Will be consistent with cognitive-developmental approach	- Cross sectional - Small sample - 'Normal' children	- Thorough pilot work - Consent & assent - Expands cognitive-developmental theoretical framework - Child's sense of personal control increases with development - Relates to clinical usefulness - Asked about common illnesses and about personal experience of illness/friends/family experiences
Potter	1984	US N=112 Age 5-9 Stratified: Ages 5-6 x56 *Assessed preoperational* Ages 7-9 x56 *Assessed concrete operational* Recruited from x2 schools	Description vignette Explanation vignette Experimenter completed questionnaire (open & closed questions)	Cognitive-developmental - How/what to tell children about illness - What age can children benefit from an explanation - Would explanations increase/decrease acceptance of disabled peer	- Cross sectional - Some results are marginal	- Specific illnesses used (diabetes & epilepsy) - All children had improved comprehension of illnesses - More observable illness seen as less attractive - Information about observable illness decreased attractiveness

Hergenrather	1991	US N=60 Age 6-14 Stratified: Age 6-7 x20 Age 9-10 x20 Age 13-14 x20 Recruited from psychology department staff or local Church	Picture sorting task Multiple choice questions x14 Multiple choice questions x12	Development *and* experience Identifying cause; symptoms; treatment of illness - How child decides s/he is sick - Identifying the cause of an illness	- Cross sectional - Small sample - Sample unrepresentative: all from psychology department staff or local Church	- All children used behaviour cues (e.g., being sent to bed) to know they were ill, not change in physiological state. - Shift towards physiological cues with age - Younger children have a more accurate knowledge about cause; consequences; treatment than previously thought
Crisp	1996	Australia Studies x2 Total N= 111 Age 4-14 Stratified: Study 1: Age 4-10 N= 40 Study 2: Age 7-14 N= 71 Recruited during hospitalization	Study 1: Interview Conservation of amount & volume Study 2: Interview PPVT-R	Cognitive-developmental *and* experience - Relationship between extent of experience of illness and level of understanding about causes of illness	- Cross sectional - Study1 small sample - Measures and interview may not have been sensitive enough	- Both age & experience contribute to children's understanding of illness - Cognitive-developmental stages too boundaried - Raises question about different illnesses and their effects on experience

Paterson	1999	New Zealand N=182 Age 7-14 44 children had asthma Recruited from Auckland schools	Structured interview including illness representation questionnaire on colds & asthma 2 sub-scales of the WISC-III Performance & Verbal IQ	Illness representations (perceptions) Does experience; age; intelligence; socioeconomic status affect sophistication of illness perceptions of colds & asthma	- Cross sectional - Interview questions might have influenced replies	- Large sample - Ethnically diverse - Education about asthma should focus on consequences and prevention
Cross	1999	US N=21 Age 7-14 Recruited from Multiple Sclerosis Society	Semi-structured interviews Content analysis	Illness perceptions Children's understanding of MS; Observations; Cause and timeline	- Simple analysis = percentages with no linking to data - No analysis by age group	- Children worry about the impact of their own behaviour - Children are unsure about heredity and contagion
Babooram	2011	Australia N=33 Age 7-12 24 'normal' weight 9 'overweight' Recruited from: - Catholic schools system - Requested appetite awareness training	Semi-structured interviews based on CSM Drawing task to obtain 'identity' *A priori* Content analysis	Illness perceptions -Children's understandings of obesity	- From affluent area of Sydney - Overweight children were already seeking help so had an awareness of issue	- Children generally have fairly accurate understanding of the causes of obesity - Children underestimate the influence of sedentary behaviour

Table 3. Summary of studies investigating concepts of family normalcy or normalization

First author	Year published	Sample (location; size; recruited from)	Method of assessment/analysis	Perspective/Focus of study	Limitations	Key/notable result
Robinson	1993	Canada N=40 (62 accounts) Study 1: Parents of hospitalised children N=9 Study 2: Adults managing own/spouses/parents/siblings chronic conditions (includes 'arthritis') N=31	Qualitative Interviews Grounded theory Analysis is of both studies	Exploration of living life 'as normal' 'normalisation' Range of chronic illnesses	- No demographic information	- Proposes trying to live life as constructed as 'normal' (good and bad) is more beneficial than detrimental - People choose to minimize difficulties - 'normalisation lens' focuses only on what is considered normal - Doing 'normal' things - Routine is important - Healthcare practitioners do not understand the value of normalising
Rehm	2000	US N=8 families (24 accounts) Recruited from Paediatric medical centre	Qualitative Ethnography-interviews and observation Symbolic interactionist	Long-term goals and normalisation strategies in families affected by HIV/AIDS	- Analytical method unclear - Small sample - Further cultural exploration might be useful	- Includes child accounts - Being normal means staying healthy - Managing stigma - minimizing disclosure - -Facilitating social activities - Reluctance to manage treatment preserves belief in normality

Knafl	2002	US N=59 Families with childhood chronic illness x2 Interviews, 1 year apart n=24 subsample analysed	Qualitative interviews Composite Case examples: Four 'families' x3 Diabetes x1RA 'Concept analysis'	Families who find normalisation a challenge	- method of analysis not discussed	- Highlights families who experience barriers to normalisation - Normalisation happens on a shifting continuum - Healthcare practitioners need to be aware of the non-normalising family
Sanderson	2011	UK N=23 Adult RA patients aged 27-79 Recruited from hospital	Qualitative Interviews Framework analysis	Changing conceptions of normal life in RA. Normalisation of symptoms	- Cross sectional - Cultural factors may vary results (all White British)	- Proposes temporal 'shifting normalities' - Six typologies outlined to describe shifts
Hart	2016	UK N=68 Young people aged 16-25 Young people n = 37 Trusted others n = 15 Health professionals n = 16 Interviews n=44 Recorded consultations n=4 Focus groups n=4 Recruited from x3 hospitals	Qualitative Interviews & Focus groups Grounded theory techniques	How young people evaluate the risks of treatment for IA (JIA; AS; PsA; RA)	- Few recorded consultations, opportunity sample	- Sample size - Attempts at data triangulation - Focuses on younger age group - important as they contemplate parenthood

References

Al-Mossawi, H., &Gunawardena, H. (2012). Understanding and recognising systemic lupus erythematosus. *Lupus UK News & Views, 98,* 5-7.

American College of Rheumatology. (1997). *1997 Update of the 1982 American College of Rheumatology revised criteria for classification of systemic lupus erythematosus*. Retrieved from: http://www.rheumatology.org/ACR/practice/clinical/classification/SLE/1997_update_of_the_1982_acr_revised_criteria_for_classification_of_sle.pdf#toolbar=1

Armistead, L., Klein, K., & Forehand, R. (1995). Parental physical illness and child functioning. *Clinical Psychology Review, 15(5),* 409-422. doi:10.1016/0272-7358(95)00023-1

Arthritis Research UK. (2011). *Psoriatic arthritis* [Pamphlett]. Chesterfield: Author.

Arthritis Research UK. (2011). *Rheumatoid arthritis* [Pamphlett]. Chesterfield: Author.

Babooram, M., Mullan, B.A., & Sharpe, L. (2011). Children's perceptions of obesity as explained by the common sense model of illness representation. *British Food Journal, 113(2),* 234-247. doi:10.1108/00070701111105321

Barlow, J.H., Cullen, L.A., Foster, N.E., Harrison, K., &Wade, M. (1999). Does arthritis influence perceived ability to fulfill a parenting role? Perceptions of mothers, fathers and grandparents. *Patient Educatiion and Counseling, 37(2),* 141-151. doi:10.1016/50738-3991(98)00136-0

Bibace, R., & Walsh, M.E. (1980). Development of children's concepts of illness. *Pediatrics, 66(6),* 912-917. doi: 10.1002/cd.23219811404

Braun, J., van den Berg, R., Baraliakos, X., Boehm, H., Burgos-Vargas, R., Collantes-Estevez, E.,&.....van der Heijde, D. (2011). 2010 update of the ASAS/EULAR recommendations for the management of ankylosing spondylitis. *Annals of the Rheumatic Diseases, 70,* 896-904. doi:10.1136/ard.2011.151027

Breast Cancer Care. (2007). *Talking with your Children about Breast Cancer.* Breast Cancer Care: London. Accessed 27th July 2013 at www.breastcancercare.org.uk

Campbell, J.D. (1975). Illness is a point of view: The development of children's concepts of illness. *Child Development, 46,* 92-100. doi: 10.2307/1128837

Carlisle, A.C.S., John, A.M.H., Fife-Schaw, C., & Lloyd, M. (2005). The self-regulatory model in women with rheumatoid arthritis: Relationships between illness representations, coping strategies, and illness outcome. *British Journal of Health Psychology, 10(4),* 571-587. doi:10.1348/135910705X52309

Chilman-Blair, K., & DeLoache, S. (2012). *Medikidz Explain Rheumatoid Arthritis.* London: Medikidz Publishing.

Cramp, F., Berry, J., Gardiner, M., Smith, F., Stephens, D. (2013). Health behaviour change interventions for the promotion of physical activity in rheumatoid arthritis: A systematic review. *Musculoskeletal Care, 11,* 238-247. doi: 10.1002/msc.1049

Crisp, J., Ungerer, J.A., & Goodnow, J.J. (1996). The impact of experience on children's understanding of illness. *Journal of Pediatric Psychology, 21(1),* 57-72. doi: 10.1093/jpepsy/21.1.57

Cross, T., Rintell, D. (1999). Children's perceptions of parental multiple sclerosis. *Psychology, Health & Medicine, 4(4),* 355-360. doi: 10.1080/135485099106090

Dabbs, A.D.V., Hoffman, L.A., Swigart, V., Happ, M.B., Dauber, J.H., McCurry, K.R. & Iacono, A. (2004). Striving for normalcy: Symptoms and the threat of rejection after lung transplantation. *Social Science and Medicine, 59,* 1473-1484. doi: 10.1016/j.socscimed.2004.01.013

Danchenko, N., Satia, J. A., & Anthony, M. S. (2006). Epidemiology of systemic lupus erythematosus: A comparison of worldwide disease burden. *Lupus, 15,* 308-318. doi:10.1191/0961203306lu2305xx

Gabriel, S.E., Michaud, K. (2009). Epidemiological studies in incidence, prevalence, mortality, and comorbidity of the rheumatic diseases. *Arthritis Research & Therapy, 11(3),* 229. doi:10.1186/ar2669. Epub 2009 May 19.

Gladman, D. D., Antoni, C., Mease, P., Clegg, D. O., & Nash, P. (2005). Psoriatic arthritis: Epidemiology, clinical features, course, and outcome. *Annals of the Rheumatic Diseases*, *64,* ii14-ii17. doi:10.1136/ard.2004.032482

Grant, M.I., Foster, N.E., Wright, C.C., Barlow, J.H., &Cullen, L.A. (1999). Being a parent or grandparent with back pain, ankylosing spondylitis or rheumatoid arthritis: A descriptive postal survey. *Musculoskeletal Care, 2(1),* 17-28. doi: 10.1002/msc.53

Graves, H., Scott, D.L., Lempp, H., & Weinman, J. (2009). Illness beliefs predict disability in rheumatoid arthritis. *Journal of Psychosomatic Research, 67,* 417-423. doi: 10.1016/j.jpsychores.2009.01.006

Hameed, K., & Akil, M. (2009). Rheumatoid arthritis: Clinical features and diagnosis. In A. Adebajo (Ed.), *ABC of Rheumatology* (4th Ed., pp. 71-75). Chichester: Wiley-Blackwell.

Hart, R.I., McDonagh, J.E., Thompson, B., Foster, H.E., Kay, L., Myers, A., & Rapley, T. (2016). Being as normal as possible: How young people ages 16-25 years evaluate the risks and benefits of treatment for inflammatory arthritis. *Arthritis Care & Research, 68(9),* 1288-1294. doi:10.1002/acr.22832

Hay, E. M., & Snaith, M. L. (1995). ABC of Rheumatology: Systemic lupus erythematosus and lupus-like syndromes. *British Medical Journal, 310,* 1257. Retrieved from http://dx.doi.org/10.1136/bmj.310.6989.1257

Heijmans, M. (1999). The role of patients' illness representations in coping and functioning with Addison's disease. *British Journal of Health Psychology, 4(2),* 137-149. doi: 10.1348/135910703322370860

Hergenrather, J.R. & Rabinowitz, M. (1991). Age-related differences in the organization of children's knowledge of illness. *Developmental Psychology, 27(6),* 952-959. doi: 10.1037/0012-1649.27.6.952

Hewlett, S., Sanderson, T., May, J., Alten, R., Bingham, C.O., Cross, M., &...Bartlett, S.J. (2012). 'I'm hurting, I want to kill myself': Rheumatoid arthritis flare is more than a high joint count - an international patient perspective on flare where medical help is sought. *Rheumatology, 51,* 69-76. doi: 10.1093/rheumatology/keq455

Hilbourne, J. (1973). On disabling the normal. *British Journal of Social Work, 3(4),* 494-504. doi:10.1093/oxfordjournals.bjsw.a056444

Hill, J,. & Ryan, S. (2000). *Rheumatology: A handbook for community nurses*. London: Whurr.

Hirsch, B.J., Moos, R.H., & Reischl, T.M. (1985). Psychosocial adjustment of adolescent children of a depressed, arthritic, or normal parent. *Journal of Abnormal Psychology, 94(2),* 154-164. doi:10.10037/0021-843X.94.2.154

Ioannu, J. (2014). Lupus in the young person. *Lupus UK News & Views, 104,* 8-9.

Jolly, M. (2005). How does quality of life of patients with systemic lupus erythematosus compare with that of other common chronic illnesses? *The Journal of Rheumatology*, 32(9), 1706-1708.

Jolly, M., Pickard, A.S., Mikolaitis, R.A., Cornejo, J., Sequeira, W., Cash, T.F., & Block, J.A. (2012). Body image in patients with systemic lupus erythematosus. *International Journal of Behavioural Medicine, 19,* 157-164. doi: 10.1007/s12529-011-9154-9

Katz, P.P., & Neugebauer, A. (2001). Does satisfaction with abilities mediate the relationship between the impact of rheumatoid arthritis on valued activities and depressive symptoms? *Arthritis Care & Research, 45,* 263-269. doi:10.1002/1529-0131(200106)45:3<263::AID-ART259>3.0.CO;2-W

Knafl, K.A., & Deatrick, J.A. (2002). The challenge of normalization for families of children with chronic conditions. *Pediatric Nursing, 28(1),* 49-56. Accessed at: www.pediatricnursing.net

Kotsis, K., Voulgari, P.V., Tsifetaki, N., Drosos, A.A., Carvalho, A.F., & Hyphantis, T. (2014). Illness perceptions and psychological distress associated with physical health-related quality of life in primary Sjögren's syndrome compared to systemic lupus erythematosus and rheumatoid arthritis. *Rheumatology International, 34,* 1671-1681. doi: 10.1007/s00296-014-3008-0

Lateef, A., &Petri, M. (2012). Unmet medical needs in systemic lupus erythematosus. *Arthritis Research & Therapy, 14(suppl.4),* S4. doi:10.1186/ar3919

Leventhal, H., Jones, S., & Trembly, G. (1966). Sex differences in attitude and behavior change under conditions of fear and specific instructions. *Journal of Experimental Social Psychology, 2,* 387-399. doi: 10.1016/0022-1031(66)90030-8

Leventhal, H., Meyer, D., & Nerenz, D. (1980). The common sense representation of illness danger. In S. Rachman (Ed.), *Contributions to Medical Psychology, Vol. 2.* (pp. 7-30). Oxford: Pergamon.

Locker, D. (1983). *Disability and Disadvantage: The Consequences of Chronic Illness.* London: Tavistock.

Madsen, M., Jensen, K.V., & Esbensen, B.A. (2015). Men's experiences of living with ankylosing spondylitis: A qualitative study. *Musculoskeletal Care,13(1),* 31-41. doi:10.1002/msc.1082

Matcham, F., Norton, S., Scott, D.L., Steer, & Hotopf, M. (2016). Symptoms of depression and anxiety predict treatment response and long-term physical health outcomes in rheumatoid arthritis: Secondary analysis of a randomized controlled trial. *Rheumatology, 55,* 268-278. doi: 10.1093/rheumatology/kev306

Meade, T., Sharpe, L., Hallab, L., Aspanell, D.,& Manolios, N. (2013). Navigating motherhood choices in the context of rheumatoid arthritis: Women's stories. *Musculoskeletal Care,11(2),73-82.* doi: 10.1002/msc.1031

Meyer, D., Leventhal, H., & Gutmann, M. (1985). Common-sense models of illness: The example of hypertension. *Health Psychology, 4(2),* 115-135. Available via: https://www.ncbi.nlm.nih.gov/pubmed/4018002

Moss-Morris, R., Petrie, K.J., & Weinman, J. (1996). Functioning in chronic fatigue syndrome: Do illness perceptions play a regulatory role? *British Journal of Health Psychology, 1,* 15-25. doi: 10.1111/j.2044-8287.1996.tb00488.x

Moss-Morris, R., Weinman, J., Petrie, K.J., Horne, R., Cameron, L.D., & Buick, D. (2002). The revised illness perception questionnaire (IPQ-R). *Psychology and Health, 17(1),* 1-16. doi: 10.1080/08870440290001494

Nagy, M. H. (1951). Children's ideas of the origin of illness. *Health Education Journal, 9(1),* 6-12. doi: 10.1177/001789695100900103

Nerenz, D.R., & Leventhal, H. (1983). Self-regulation theory in chronic illness. In T.G. Burish & L.A. Bradley (Eds.), *Coping with Chronic Disease: Research and Applications* (pp. 13-37). New York: Academic Press.

Paterson, J., Moss-Morris, R., Butler, S.J. (1999). The effect of illness experience and demographic factors on children's illness representations. *Psychology and Health, 14,* 117-129. doi: 10.1080/08870449908407318

Pimm, T.J., & Weinman, J. (1998). Applying Leventhal's self-regulation model to adaptation and intervention in rheumatic disease. *Clinical Psychology and Psychotherapy, 5,* 62-75. doi: 10.1002/(sici)1099-0879(199806)5:2c62::AID-cpp155>3.0.co;2-L

Potter, P.C., & Roberts, M.C. (1984). Children's perceptions of chronic illness: The roles of disease symptoms, cognitive development, and information. *Journal of Pediatric Psychology, 9(1),* 13-27. doi: 10.1093/jpepsy/9.1.13

Primholdt, N., Primdahl, J., & Hendricks, O. (2016). A difficult diagnosis: A qualitative study of the daily lives of young men diagnosed with ankylosing spondylitis. *Musculoskeletal Care.* Early view online: doi: 10.1002/msc.1155

Pullman-Mooar, S. (1999). Arthritis and rheumatic diseases. In S. J Gates, & P. A. Mooar (Eds.), *Musculoskeletal Primary Care* (pp. 288-326). Philadelphia: Lippincott.

Rehm, R.S., & Franck, L.S. (2000). Long-term goals and normalization strategies of children and families affected by HIV/AIDS. *Advances in Nursing Science, 23(1),* 69-82. Accessed at: journals.lww.com/advancesinnursingscience/Pages/default.aspx

Riemsma, R.P., Taal, E., Wiegman, O., Rasker, J.J., Bruyn, G.A.W., & Van Paassen, H.C. (2000). Problematic and positive support in relation to depression in people with rheumatoid arthritis. *Journal of Health Psychology, 5(2),* 221-230. doi:10.1177/135910530000500212

Robinson, C.A. (1993). Managing life with a chronic condition: The story of normalization. *Qualitative Health Research, 3(1),* 6-28. doi:10.1177/104973239300300102

Rolland, J.S. (1999). Parental illness and disability: A family systems framework. *Journal of Family Therapy, 21,* 242-266. doi: 10.1111/1467-6427.00118

Sanderson, T., Calnan, M., Morris, M., Richards, P., & Hewlett, S. (2011). Shifting normalities: Interactions of changing conceptions of a normal life and the normalisation of symptoms in rheumatoid arthritis. *Sociology of Health and Illness, 33(4),* 618-633. doi:10.1111/j.1467-9566.2010.01305.x

Shaffer, D. (1996). *Developmental Psychology: Childhood and Adolescence* (4th Ed.). Cognitive development: Piaget's theory and Vygotsky's sociocultural viewpoint (pp. 243-275). Pacific Grove: Brooks/Cole.

Symmonds, D. P. M. (2005). Looking back: Rheumatoid arthritis - aetiology, occurrence and mortality. *Rheumatology,44(suppl. 4),*iv14-iv17.doi:10.1093/rheumatology/kei055

Vaughn, R., Morrison, L., & Miller, E. (2003). The illness representations of multiple sclerosis and their relations to outcome. *British Journal of Health Psychology, 8,* 287-301. doi: 10.1348/135910703322370860

Weinman, J., Petrie, K.J., Moss-Morris, R., & Horne, R. (1996). The Illness Perception Questionnaire: A new method for assessing the cognitive representation of illness. *Psychology and Health, (11),* 431-445. doi:10.1080/08870449608400270

Woolf, A.D. (2012). Influential global alliance calls on governments and the World Health Organisation to prioritise musculoskeletal health following findings of Global Burden of Disease Study 2010. *Global Alliance for Musculoskeletal Health* press release 14th December 2012. Truro, UK.

Appendix A - Targeted Journal Style Guidelines

British Journal of Health Psychology

Edited By: Alison Wearden and David French

Impact Factor: 2.895

ISI Journal Citation Reports © Ranking: 2015: 21/121 (Psychology Clinical)

Online ISSN: 2044-8287

Author Guidelines

The aim of the British Journal of Health Psychology is to provide a forum for high quality research relating to health and illness. The scope of the journal includes all areas of health psychology as outlined in the Journal Overview.
The types of paper invited are:

• papers reporting original empirical investigations, using either quantitative or qualitative methods, including reports of interventions in clinical and non-clinical populations;
• theoretical papers which may be analyses or commentaries on established theories in health psychology, or presentations of theoretical innovations;

• we particularly welcome review papers, which should aim to provide systematic overviews, evaluations and interpretations of research in a given field of health psychology; and

• methodological papers dealing with methodological issues of particular relevance to health psychology.

All papers published in The British Journal of Health Psychology are eligible for Panel A: Psychology, Psychiatry and Neuroscience in the Research Excellence Framework (REF).

1. Circulation
The circulation of the Journal is worldwide. Papers are invited and encouraged from authors throughout the world.

2. Length
Papers describing quantitative research (including reviews with quantitative analyses) should be no more than 5000 words (excluding the abstract, reference list, tables and figures). Papers describing qualitative research (including reviews with qualitative analyses) should be no more than 6000 words (including quotes but excluding the abstract, tables, figures and references). The Editors retain discretion to publish papers beyond this length in cases where the clear and concise expression of the scientific content requires greater length.

3. Editorial policy

The Journal receives a large volume of papers to review each year, and in order to make the process as efficient as possible for authors and editors alike, all papers are initially examined by the Editors to ascertain whether the article is suitable for full peer review. In order to qualify for full review, papers must meet the following criteria:

• the content of the paper falls within the scope of the Journal

• the methods and/or sample size are appropriate for the questions being addressed

• research with student populations is appropriately justified

• the word count is within the stated limit for the Journal (i.e. 5000 words)

4. Submission and reviewing
All manuscripts must be submitted via Editorial Manager. The Journal operates a policy of anonymous (double blind) peer review. We also operate a triage process in which submissions that are out of scope or otherwise inappropriate will be rejected by the editors without external peer review to avoid unnecessary delays. Before submitting, please read the terms and conditions of submission and the declaration of competing interests. You may also like to use the Submission Checklist to help your prepare your paper.

5. Manuscript requirements
• Contributions must be typed in double spacing with wide margins. All sheets must be numbered.

• Manuscripts should be preceded by a title page which includes a full list of authors and their affiliations, as well as the corresponding author's contact details. A template can be downloaded from here.
• For articles containing original scientific research, a structured abstract of up to 250 words should be included with the headings: Objectives, Design, Methods, Results, Conclusions. Review articles should use these headings: Purpose, Methods, Results, Conclusions. As the abstract is often the most widely visible part of your paper, it is important that it conveys succinctly all the most important features of your study. You can save words by writing short, direct sentences. Helpful hints about writing the conclusions to abstracts can be found here.
• Statement of Contribution: All authors are required to provide a clear summary of ‘what is already known on this subject?’ and ‘what does this study add?’. Authors should identify existing research knowledge relating to the specific research question and give a summary of the new knowledge added by your study. Under each of these headings, please provide 2-3 (maximum) clear outcome statements (not process statements of what the paper does); the statements for 'what does this study add?' should be presented as bullet points of no more than 100 characters each. The Statement of Contribution should be a separate file.

• Conflict of interest statement: We are now including a brief conflict of interest statement at the end of each accepted manuscript. You will be asked to provide information to generate this statement during the submission process.

• The main document must be anonymous. Please do not mention the authors’ names or affiliations (including in the Method section) and always refer to any previous work in the third person.

• Tables should be typed in double spacing, each on a separate page with a self-explanatory title. Tables should be comprehensible without reference to the text. They should be placed at the end of the manuscript but they must be mentioned in the text.

• Figures can be included at the end of the document or attached as separate files, carefully labelled in initial capital/lower case lettering with symbols in a form consistent with text use. Unnecessary background patterns, lines and shading should be avoided. Captions should be listed on a separate sheet. The resolution of digital images must be at least 300 dpi. All figures must be mentioned in the text.

• For reference citations, please use APA style. Particular care should be taken to ensure that references are accurate and complete. Give all journal titles in full and provide doi numbers where possible for journal articles. For example:

Author, A., Author, B., & Author, C. (1995). *Title of book.* City, Country: Publisher.
Author, A. (2013). Title of journal article. *Name of journal, 1,* 1-16. doi: 10.1111/bjep.12031
• SI units must be used for all measurements, rounded off to practical values if appropriate, with the imperial equivalent in parentheses.

• In normal circumstances, effect size should be incorporated.

• Authors are requested to avoid the use of sexist language.

• Authors are responsible for acquiring written permission to publish lengthy quotations, illustrations, etc. for which they do not own copyright. For guidelines on editorial style, please consult the APA Publication Manual published by the American Psychological Association.
• Manuscripts describing clinical trials are encouraged to submit in accordance with theCONSORT statement on reporting randomised controlled trials.
• Manuscripts reporting systematic reviews and meta-analyses are encouraged to submit in accordance with the PRISMA statement.
• Manuscripts reporting interventions are encouraged to describe them in accordance with the TIDieR checklist.
If you need more information about submitting your manuscript for publication, please email Hannah Wakley, Managing Editor (bjhp@wiley.com) or phone +44 (0) 116 252 9504.

6. Supporting information
Supporting Information can be a useful way for an author to include important but ancillary information with the online version of an article. Examples of Supporting Information include appendices, additional tables, data sets, figures, movie files, audio clips, and other related nonessential multimedia files. Supporting Information should be cited within the article text, and a descriptive legend should be included. Please indicate clearly on submission which material is for online only publication. It is published as supplied by the author, and a proof is not made available prior to publication; for these reasons, authors should provide any Supporting Information in the desired final format.

For further information on recommended file types and requirements for submission, please visit the Supporting Information page on Author Services.

7. OnlineOpen
OnlineOpen is available to authors of primary research articles who wish to make their article available to non-subscribers on publication, or whose funding agency requires grantees to archive the final version of their article. With OnlineOpen, the author, the author's funding agency, or the author's institution pays a fee to ensure that the article is made available to non-subscribers upon publication via Wiley Online Library, as well as deposited in the funding agency's preferred archive. A full list of terms and conditions is available on Wiley Online Library.

Any authors wishing to send their paper OnlineOpen will be required to complete thepayment form.
Prior to acceptance there is no requirement to inform an Editorial Office that you intend to publish your paper OnlineOpen if you do not wish to. All OnlineOpen articles are treated in the same way as any other article. They go through the journal's standard peer-review process and will be accepted or rejected based on their own merit.

8. Author Services
Author Services enables authors to track their article – once it has been accepted – through the production process to publication online and in print. Authors can check the status of their articles online and choose to receive automated e-mails at key stages of production. The author will receive an e-mail with a unique link that enables them to register and have their article automatically added to the system. You can then accessKudos through Author Services, which will help you to increase the impact of your research. Visit Author Services for more details on online production tracking and for a wealth of resources including FAQs and tips on article preparation, submission and more.

9. Copyright and licences
If your paper is accepted, the author identified as the formal corresponding author for the paper will receive an email prompting them to login into Author Services, where via the Wiley Author Licensing Service (WALS) they will be able to complete the licence agreement on behalf of all authors on the paper.

For authors signing the copyright transfer agreement
If the OnlineOpen option is not selected the corresponding author will be presented with the copyright transfer agreement (CTA) to sign. The terms and conditions of the CTA can be previewed in the samples associated with the Copyright FAQs .
For authors choosing OnlineOpen
If the OnlineOpen option is selected the corresponding author will have a choice of the following Creative Commons Licence Open Access Agreements (OAA):

- Creative Commons Attribution Non-Commercial Licence (CC-BY-NC)
- Creative Commons Attribution Non-Commercial -NoDerivs Licence (CC-BY-NC-ND)

To preview the terms and conditions of these open access agreements please visit theCopyright FAQs and you may also like to visit the Wiley Open Access Copyright and Licence page.
If you select the OnlineOpen option and your research is funded by The Wellcome Trust and members of the Research Councils UK (RCUK) or the Austrian Science Fund (FWF) you will be given the opportunity to publish your article under a CC-BY licence supporting you in complying with your Funder requirements. For more information on this policy and the Journal's compliant self-archiving policy please visit our Funder Policy page.

10. Colour illustrations
Colour illustrations can be accepted for publication online. These would be reproduced in greyscale in the print version. If authors would like these figures to be reproduced in colour in print at their expense they should request this by completing a Colour Work Agreement form upon acceptance of the paper.

11. Pre-submission English-language editing
Authors for whom English is a second language may choose to have their manuscript professionally edited before submission to improve the English. A list of independent suppliers

of editing services can be found in Author Services. All services are paid for and arranged by the author, and use of one of these services does not guarantee acceptance or preference for publication.

12. The Later Stages

The corresponding author will receive an email alert containing a link to a web site. The proof can be downloaded as a PDF (portable document format) file from this site. Acrobat Reader will be required in order to read this file. This software can be downloaded (free of charge) from Adobe's web site. This will enable the file to be opened, read on screen and annotated direct in the PDF. Corrections can also be supplied by hard copy if preferred. Further instructions will be sent with the proof. Excessive changes made by the author in the proofs, excluding typesetting errors, will be charged separately.

13. Early View

British Journal of Health Psychology is covered by the Early View service on Wiley Online Library. Early View articles are complete full-text articles published online in advance of their publication in a printed issue. Articles are therefore available as soon as they are ready, rather than having to wait for the next scheduled print issue. Early View articles are complete and final. They have been fully reviewed, revised and edited for publication, and the authors' final corrections have been incorporated. Because they are in final form, no changes can be made after online publication. The nature of Early View articles means that they do not yet have volume, issue or page numbers, so they cannot be cited in the traditional way. They are cited using their Digital Object Identifier (DOI) with no volume and issue or pagination information. Eg Jones, A.B. (2010). Human rights Issues. *Journal of Human Rights.* Advance online publication. doi:10.1111/j.1467-9299.2010.00300.x

Further information about the process of peer review and production can be found in this document. What happens to my paper?

Abstracting and Indexing Information

- Academic Search Alumni Edition (EBSCO Publishing)
- Academic Search Complete (EBSCO Publishing)
- Academic Search Elite (EBSCO Publishing)
- Academic Search Premier (EBSCO Publishing)
- Advanced Placement Source (EBSCO Publishing)
- ArticleFirst (OCLC)
- Arts & Humanities Citation Index (Thomson Reuters)
- ASSIA: Applied Social Sciences Index & Abstracts (ProQuest)
- BNI: British Nursing Index (ProQuest)
- Current Contents: Arts & Humanities (Thomson Reuters)
- Electronic Collections Online (OCLC)
- IBR & IBZ: International Bibliographies of Periodical Literature (KG Saur)
- MEDLINE/PubMed (NLM)
- ProQuest Central (ProQuest)
- ProQuest Central: Professional Edition (ProQuest)
- ProQuest Health & Medical Complete (ProQuest)
- ProQuest Medical Library (ProQuest)
- ProQuest Psychology Journals (ProQuest)
- ProQuest Public Health (ProQuest)
- ProQuest Science Journals (ProQuest)
- Psychology & Behavioral Sciences Collection (EBSCO Publishing)
- PsycINFO/Psychological Abstracts (APA)

- SCOPUS (Elsevier)
- Social Sciences Citation Index (Thomson Reuters)
- SocINDEX (EBSCO Publishing)
- SocINDEX with Full Text (EBSCO Publishing)
- Sociological Abstracts (ProQuest)
- VINITI (All-Russian Institute of Science & Technological Information)
- Web of Science (Thomson Reuters)

Service Evaluation: Executive summary and recommendations

Should there be information and education available for children whose parents or grandparents have chronic rheumatic conditions?

Overview:-

People with chronic rheumatic conditions are often given leaflets or signposted to websites that provide information, education and advice about coping with their condition. However, there are no similar age-appropriate resources available specifically for the children within these families, to help them to understand their parent's or grandparent's condition. Provision of such resources might provide a useful addition, enabling families to have conversations about difficult subjects.

The author is a practising health psychologist working within a secondary care rheumatology service. Using a cross-sectional study design, a questionnaire was distributed to adult patients attending the local rheumatology service and made available to members of four national UK rheumatology charities via "Survey Monkey" ™.This evaluation sought to establish:

- Whether parents/grandparents would welcome such resources.
- What information would be useful to their children/grandchildren.
- The format, timing and method of delivery of resources.
- Whether children/grandchildren performed 'caring' tasks, specifically due to a parent/grandparent's rheumatic condition.

Results:-

- Participants were strongly in favour of information about rheumatic conditions being made available to their children/grandchildren in a developmentally appropriate way.
- Participants reported that leaflets, websites and having someone for their children to talk to would be most useful, and suggestions were made for the content of such information.
- Participants indicated that the nurse specialist might be appropriate in assisting them to talk to their children/grandchildren about their rheumatic condition.
- Approximately a quarter of children/grandchildren helped with domestic 'caring' tasks.

Recommendations:-

- Further research should now be conducted with families **and their children** for whom these resources are intended, so that they play a key role in shaping the content and development of materials.
- Once resources are developed they should be piloted with relevant families for feedback and review.
- Once available, clinical nurse specialists will be able to provide resources to patients, and signpost families with more complex mental health needs to the psychology service for specialist interventions.
- Resources should be freely available via recognised partners.

Dissemination process:-

Results were presented to the local rheumatoid arthritis support group, the local combined clinical and research group for rheumatology (11th February 2016) and presented as a poster at the *British Society for Rheumatology* conference, Glasgow, 28th April 2016. Summaries have been provided to Arthritis Research UK and the national charities involved.

CHAPTER Two

Service Evaluation

Should there be information and education available for children whose parents or grandparents have chronic rheumatic conditions?

1.0 Introduction:-

It is estimated that approximately 20% of all General Practitioner (GP) consultations with adults in the United Kingdom (UK) each year are for musculoskeletal related conditions (Arthritis Research UK, 2012 and 2014). People who are suspected of having a rheumatic condition are referred to a specialist tertiary care centre for diagnosis and potentially long-term management of their condition. Whilst the prevalence of these conditions rises with age, perhaps contrary to popular opinion, many of those affected will be young adults with children (Parsons, Ingram, Clarke-Cornwell & Symmons, 2011; Stack et al., 2012).

Within the UK, national rheumatology charities such as Arthritis Research UK (ARUK) have developed a range of educational leaflets for patients that outline specific rheumatic diseases, potential treatments, and some psychosocial advice; for example, "Rheumatoid Arthritis" (ARUK, 2011). Such leaflets may assist patients in preparing their families for the on-going effects of their condition, which tends to have periods of quiescence and 'flare' when the disease becomes more active (Barlow & Wright, 1998).

Patient's families also need information and resources to enable them to understand and support the person with the rheumatic condition. However, whilst there are information leaflets for spouses and partners of people with rheumatic conditions, there are no age-appropriate information or educational materials available specifically for children within these families. It is thus not known what information would be useful to children of parents with rheumatic conditions, nor the

content or format in which this should be delivered. Indeed, it has not been established whether parents would welcome such resources for their children to use.

1.1 The impact of rheumatic disease on the family:-

A few studies suggest that parents and grandparents with rheumatic conditions struggle with particular aspects of parenting. Pain, fatigue and difficulties with physical function (like picking up a child, or engaging in and keeping up with physical play) are reported (Barlow, Cullen, Foster, Harrison & Wade, 1999; Grant, Foster, Wright, Barlow & Cullen, 2004). Perceived inability to carry out the parenting role may result in negative emotions such as frustration, anger, guilt and depression, which can impact upon the well-being of the whole family unit. How well a family manage the impact of a rheumatic disease may depend upon how willing and/or capable family members are to provide support in order to retain a cohesive family unit. This may involve the redistribution of children's activities (such as after-school clubs) to others; making the activities occasional, or ceasing some activities altogether (Backman, Del Fabro Smith, Smith, Montie & Suto, 2007).

Additionally, some children may become responsible for more domestic, personal and emotional caring tasks than would normally be expected. However, simply having a parent with a chronic health condition does not make a child a de facto "young carer", although the simple availability of the child may influence this (Aldridge & Becker, 1999).

1.2 Communicating with the children:-

In an early study reported by Barlow et al. (1999), mothers, fathers and grandparents revealed that all had talked to 'older' children (up to 15 years) about their rheumatic condition. They felt that parenting was easier if the children were aware of potential limitations, which also helped with children's expectations and concerns. Without additional resources younger children may find this difficult to understand however, and parents have noted the behaviour of young children deteriorating when the parent with RA was unwell (National Rheumatoid Arthritis Society (NRAS), 2012). Similarly, not talking to children may mean they do not understand the consequences of a rheumatic condition and may not offer the help needed with household chores (Grant et al., 2004).

Within other disease groups, studies have looked at the amount and timing of information provided for children about parental ill health, and the difficulties parents experience in communicating with them, often shielding their child from anticipated anxiety by not informing them about their diagnosis and potential treatments for some time, and not expecting their children to understand anyway (Barnes, Kroll, Burke, Jones & Stein, 2000). Conversely, families who did discuss diagnosis and treatments with children, were driven by the explicit aim of preventing anxiety by encouraging discussion and trust within the family. Children often know something is amiss before they are told, and parents have reported feeling surprised (Barnes et al., 2000) at what children may have known about a disease (such as cancer), and how

shocked and upset the child was, when in fact the parents had thought the child had 'coped well' (Forrest, Plumb, Ziebland & Stein, 2006).

Patients are increasingly well supported at the time of receiving a diagnosis of a chronic health problem and may be encouraged to talk to their families about this, although they may also often be left unprepared and unsupported in how to do this (Kroll, Barnes & Jones, 1998). Parents have called for more support and resources from health professionals to enable them to talk to their children in an appropriate way for their child's stage of development. Knowing 'how to' break the news, what kind of language to use, and guidance on how to cope with children's emotions and reactions have all been identified as important factors (Barnes et al., 2000). Support from a health professional in person may be useful, so that the child can ask questions directly, particularly during the period of diagnosis, when the parent may be "reeling" (Forrest et al., 2006). This is directly relevant for parents newly diagnosed with a chronic rheumatic condition who are likely to be in a good deal of pain, and having to adjust to an intensive and bewildering treatment and medication regimen.

1.3 The need for research: Existing local research

The impetus for the present study came from an earlier qualitative interview study into the healthcare experiences and importance of appearance concerns for women with systemic lupus erythematosus (SLE), (Hale, Treharne, Norton et al., 2006). Although not the purpose of the study, it became apparent that the impact of SLE on the family unit

was a major concern. Mothers stated there were no age-appropriate educational resources or support available to facilitate discussion with their children, in order to help the family discover the children's understanding and concerns. Communication with healthcare providers about these issues did not usually happen, for a variety of reasons. Children themselves are often effectively excluded from the healthcare interactions that their parents have, and may feel uncertain about whether, or how, to raise questions (Hale, Treharne, Lyons et al., 2006; Hale, Treharne, Norton, Mitton et al., 2006).

Whilst the research mentioned above concerns a small study of a group of women with SLE, these issues may be similar across a wide range of rheumatic diseases. The local NHS Foundation Trust (DGH) serves a population of approximately 312,000 inhabitants and has one of the largest adult rheumatology tertiary care centres in England ("Dudley Joint ...", 2012). Unusually for a rheumatology department, there is an in-house part-time health psychology service and referrals are made by any member of the multi-disciplinary team. Within the psychology clinical interview, patients are encouraged to tell their rheumatology "story" from early symptoms to diagnosis, treatment, impact on work, leisure and family life. Patients often refer to difficulties they experience in communicating effectively with their children (and/or grandchildren) about their rheumatic disease and its consequences, and how additional information resources would help them do this.

2.0 Method

2.1 Aims of the study:-

1. to determine attitudes to providing information and resources for children/ grandchildren of people with rheumatic conditions.

2. to obtain views on the content, format, timing and delivery of such information and resources.

3. to establish any tasks performed, and the amount of time children/grandchildren might be spending in 'caring' roles for an adult with a rheumatic condition.

2.2 Study design:-

In order to ascertain the views of a relevant, reasonably large and geographically varied sample, a questionnaire study design was considered appropriate.

2.3 Participants:-

All adult (>18) patients, regardless of rheumatological diagnosis, attending routine follow-up appointments at the DGH rheumatology department, and all members of the National Rheumatoid Arthritis Society (NRAS); National Ankylosing Spondylitis Society (NASS); Raynauds and Scleroderma Association, now Scleroderma Society (SS); Lupus UK and Psoriasis Association were invited to take part in the study. Only the Psoriasis Association did not respond.

2.3.1 Inclusion/Exclusion criteria:-

All members of the above organisations were eligible to access the information sheet and questionnaire via Survey Monkey™ or by requesting the same by post (Lupus UK). Patients attending DGH with diagnosed severe mental health difficulties or learning disabilities were not approached to take part.

2.3.2 Sample size:-

Power calculations indicated that a sample size of 1,079, with a standard α-level of ≤0.05(95% confidence) and power of 0.80 (1-β) would detect a small effect size (w=0.08) in a typical chi-squared 2x2 distribution with one degree of freedom. Increasing the degrees of freedom to 5(for a test of a variable with 6 options) indicates that a sample of 1061 would still produce an effect size of w = 0.11 (Faul, 2012).

A total of 1635 completed questionnaires were received. Of these 1079 were available for analysis (556 contained only demographic information). Nurses recruiting to the study across three DGH sites did not keep a record of the number of questionnaires distributed, therefore it was not possible to record the percentage of the potential sample approached who subsequently participated.

2.4 Measure:

Patient partners from the DGH (patient-led rheumatoid arthritis support group and Lupus UK support group) were involved in the development of the project aims, information sheet (Appendix A) and

questionnaire (Appendix B). The questionnaire contained closed, multiple response and open-ended questions.

Four patient partners read and completed the questionnaire, commenting on its overall structure, and whether each item was understandable using a verbal cognitive tracking process where each question is spoken aloud (Williams, 2001). Patients reported that the questionnaire was clear, easy to understand and appropriate, and took about 5-10 minutes to complete (Kelly & Haidet, 2007; Nevo, 1985).

2.5 Research procedure:-

Patients were approached by the nurse attending the patient prior to their appointment with the doctor or rheumatology clinical nurse specialist (CNS) and given an information sheet, questionnaire and pre-paid reply envelope. Completed questionnaires were returned to the nurse or returned by post. Any patient with a visual impairment was offered the chance to complete the questionnaire with the aid of the researcher. Patients who required the services of an interpreter were offered a questionnaire and a prepaid reply envelope, as these were not available. Recruitment took place between September 2012 and August 2014.

NRAS, NASS, and the Scleroderma Society provided information about the study and a link to the adapted questionnaire hosted on ″SurveyMonkey″ ™ (n.d.) on their websites. Lupus UK posted packs containing the study information sheet, questionnaire and a prepaid

envelope to 500 of their members in the East and West Midlands, UK. Recruitment took place between November 2012 and March 2014.

2.6 Ethical considerations:-

The questionnaires did not ask for any identifiable personal information. All completed questionnaires were securely stored in accordance with NHS guidelines. Following the NHS Health Research Authority website guidance on consent and participant information (n.d.) completion of the questionnaire was deemed informed consent to participate in the study. The Code of Human Research Ethics (British Psychological Society, 2014), and Code of Ethics and Conduct (British Psychological Society, 2009) were adhered to.

Ethical approval was obtained from the NHS Health Research Authority, National Research Ethics Service Committee West Midlands - Staffordshire, UK. Reference number 12/WM/022 (Appendix C).

2.7 Data analysis:-

In order to prepare a coding frame for analysis of open-ended questions, 50 questionnaire responses were examined in detail. Open-ended responses were listed, printed in hard copy, then grouped by theme in order to develop a thematic coding frame. Once this had been developed a further 50 questionnaires were examined against this frame and adjusted accordingly (Bowling, 2002). Examples of free text from questionnaires are used to illustrate responses and thematic coding. To provide a clear audit trail, each example of free text used is coded with the questionnaire number. Further coding illustrates whether the

participant had children/grandchildren < 18 years, or adult children/grandchildren > 18 years.

Quantitative data were analysed using the Statistical Package for the Social Sciences™ (SPSS Version 20). Data were analysed using the Pearson Chi-square test in order to look for associations between categorical variables (Field, 2011). Cramer's V indicates the strength of association between variables (effect size).

3.0 Results

3.1Sociodemographics:-

Participant demographic characteristics are outlined in Table 1. The majority of responses (73%) came from the local hospital. Of those who stated their postcode, we were able to determine we had reached 65% of all UK postcode areas, excluding UK islands (Figure 1). We also received 13 international responses.

As expected, nearly 74% of participants were female, reflecting the general predominance of women within the rheumatic diseases. Questionnaires were more likely to be completed by people aged over 55 (65%) reflecting age-related prevalence in the rheumatic diseases. Most participants had either a child or grandchild under the age of eighteen (93%).

The questionnaire asked participants to state the name of their rheumatic condition resulting in 23 named conditions, although some

participants had 'none' and completed the questionnaire on behalf of a spouse or partner (either living or deceased). With guidance (Firestein,

Table 1. Participant demographics

	N	*%*
Recruitment		
Local hospital DGH	783	73
National charities (websites)	164	15
National charities (postal response)	132	12
Gender		
Male	277	26.1
Female	783	73.9
Age		
18-24	13	1.5
25-34	48	5.6
35-44	91	10.6
45-54	152	17.7
55-64	225	26.2
65-74	211	24.6
75-84	118	13.8
Marital Status		
Single	95	9
Co-Habiting/Married	825	77.8
Separated/Divorced	97	9.2
Widowed	44	4.1
Qualifications		
None	88	12.2
GCSE	152	21.1
Post 16 education	270	37.5
University degree	120	16.6
Postgraduate	54	7.5
Other	37	5.1
Diagnosis		
Inflammatory musculoskeletal diseases	622	60.3
Osteoarthritis	104	10.1
Metabolic bone diseases	15	1.5
Connective tissue diseases	250	24.2
Vascular diseases	18	1.7
Gout	7	0.7
Fibromyalgia	5	0.5
Soft tissue rheumatism	1	0.1
None	10	1

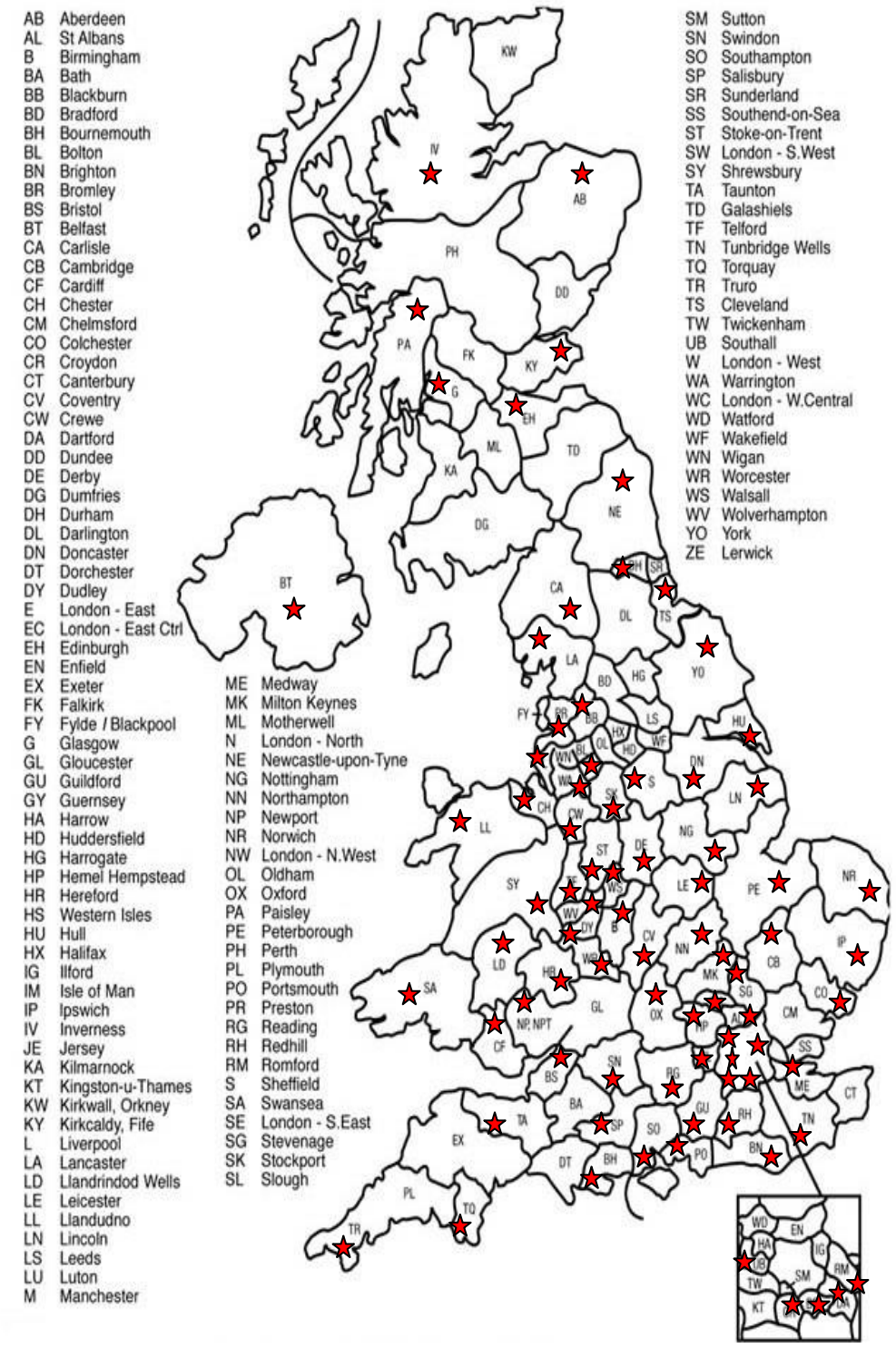


Figure 1. United Kingdom Postcode Index - Questionnaire responses received from starred areas.

Budd, Gabriel, McInnes & O'Dell, 2013) and input from a consultant rheumatologist (GDK), the 23 conditions were collapsed into nine coherent categories consistent with current clinical practice. The sample closely resembled the general rheumatology population, with most participants (60.3%) having an inflammatory musculoskeletal condition, a connective tissue disease (24.2%) or degenerative Osteoarthritis (10.1%). Overall, disease duration ranged from ≤12 months to ≥63 years.

3.2 Information needs:-

In support of our clinical evidence and research literature from other fields, over 90% of participants thought that there should be information available for children/grandchildren whose parents/grandparents have a chronic rheumatic condition (*n* = 955) and answered 'Yes' to this key question. Parental status (i.e., not having children; being a parent only; being a grandparent) and educational qualifications, did not influence responses to this question (NS).

Participants were invited to use free text to explain why they answered 'Yes' or 'No' to our key question. These responses were analysed using the procedure described earlier, which yielded eight possible thematic categories, one for explanations of 'No' answers, and seven for explanations of 'Yes' answers, (see Table 2).

3.2.1 Explaining 'No' answers:-

Using the protective strategy of not wanting to worry a child by providing information characterised 'No' answers: "No. Would not want them to worry about condition" (076/AC/GC1).

3.2.2 Explaining 'Yes' answers:-

Most participants indicated that improving the child's 'general' understanding of their rheumatic condition would be useful, as would an understanding of how that condition impacts upon the person: "Because my 11yr old daughter gets v resentful that I can't keep up with her and her friend's mums. She thinks I am there to do everything for her and she resents having to do things for herself"(032AC/C17/11) and "To take away any anxiety the child might have about their parent or grandparent and to help them understand why granny might be in pain or having an 'off' day" (546AC/GC12).

3.3 The kind of information provided:-

Participants were asked to give free text answers about the kind of information that would be useful for a child/grandchild to have about a parent/grandparent's rheumatic condition. As previously, thematic categories were developed.

A few participants (2%), were unsure, whilst others answered simply 'give all information' (17.1%). Others felt that information should be individualised to the child and contain historical family health information, usually due to heritability concerns (3.4%).

Table 2. Reasons for giving/not giving information about a parent/grandparent's chronic rheumatic condition

	Thematic categories	***n***	***Valid %***
1	**Not giving** information protects the children from worry	47	5.4
2	Giving information stops the child from worrying	75	8.6
3	Giving information improves general understanding	236	27.2
4	Giving information helps the child to understand the impact of the condition	216	24.9
5	Giving information helps, because there are *specific* heritability concerns	92	10.6
6	Giving information helps, because there are *general* heritability concerns	54	6.2
7	Giving information will help the child to take preventative action	46	5.3
8	Giving information will aid early screening/diagnosis	103	11.9
	Total	**869**	**100**

Note. Missing data *n*=210.

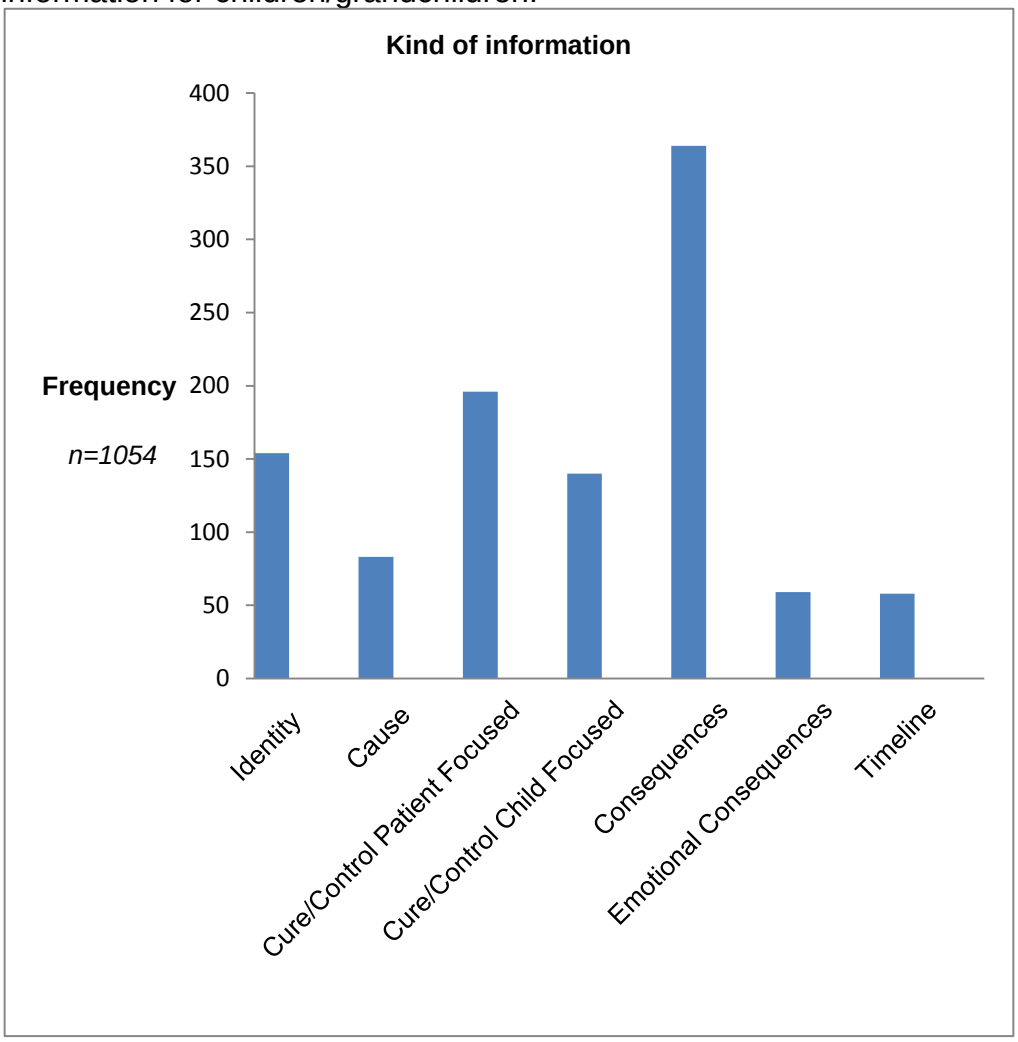
Table 3 illustrates the seven main themes or topics of information that participants thought should be included in information for children/grandchildren, in 'easier' or age-appropriate formats. The most frequent suggestion (33.7%) related to the physical impact of their

Table 3. Main topics of information for children

Theme	Concept contained within the theme
Identity	Names for the condition(s) and the symptoms that may go with it/them. The age at which people might develop these conditions.
Cause	Where possible, information should outline the possible causes of rheumatic conditions.
Curability/ Controllability ***(Patient-focused)***	Information should explain how the condition might be cured or controlled, including any treatment and monitoring. There should be practical suggestions how the child/grandchild can help the parent/grandparent.
Curability/ Controllability ***(Child-focused)***	Information should include preventative steps the child could take to avoid similar conditions. There should be encouragement to seek early 'screening' or diagnosis. There should be information on the likelihood of heritability.
Consequences	Outline the physical impact of the condition and treatments.
Emotional Consequences	Explain the emotional impact of the condition. Why parent/grandparent may be 'irritable'; 'grumpy'; 'sad'
Timeline	How long conditions may last - i.e., that they are long-term, with related support needed.

condition or 'consequences', and the *impact* of treatment (see Figure 2.) This included information that would help a child understand when a parent/grandparent was in pain, stiff, or very fatigued; and when they would be unable to do certain things at these times like lift, carry or play physical games. Some treatments regularly produce nausea and

Figure 2. Main topics of information that should be covered in information for children/grandchildren.



fatigue, and parents/grandparents will be less able to engage with family and other commitments on those days. Parents who had a child under 18 years were more likely to want information about the consequences of a rheumatic condition than if the child was over 18 years (χ^2(1, N = 900) = 23.1, *p*= .001(Cramer's V= 0.016) indicating a small effect size.

Whilst more grandparents indicated the same, this was non-significant (χ^2 (1, N = 526) = 3.347, *p*= .075 (Cramer's V = 0.08) again with a small effect size.

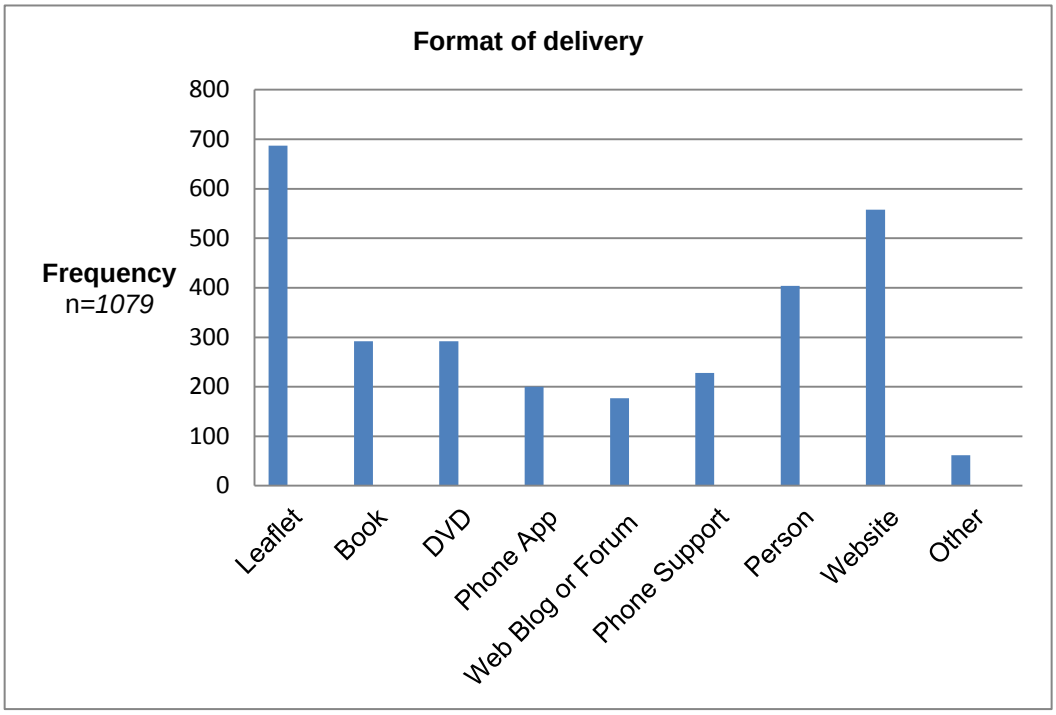
Participants thought it would be important to include information about the controllability or curability of a rheumatic condition by discussing what treatments are for, how they are monitored, and their likely outcomes (18.2%). Helping to control the condition included practical management suggestions about how a child can help a parent/grandparent, such as fetching and carrying things and being careful not to play 'roughly'.

Following on from this, participants felt it would be important to have a name or 'identity' for the rheumatic condition, and a description of symptoms to expect. Information about the general age of onset might also be useful (14.3%). This relates to the next most common response *curability/controllability- child focused*, which suggested children should be made aware of any preventative steps they could take to avoid the onset of a rheumatic condition themselves later in life. Some felt that there should be encouragement to seek early 'screening' or to seek early diagnosis if symptoms occur. There were often mentions of specific heredity concerns and the possibility of genetic links that people felt children should be aware of (13%).

3.4 Format of information:-

Leaflets, websites or delivery by a person were the most popular formats (Figure 3). Comparisons between those with children/grandchildren aged either under or over eighteen revealed no significant differences in overall preference of format, although those

Figure 3. Preferred format of delivery of information



with younger children/grandchildren were more likely to say 'Yes' to any of the formats, perhaps reflecting a belief that younger people are more open to multiple modes of delivery including those using internet and social media technology (Table 3). Proportionally more people with a University education were in favour of websites compared to those with pre- and post-16 education (χ^2 (2, N = 720) = 12.520, p<.05 (Cramer's V = .132); standardised residual -2.1.

As with the answers detailing the kind of information that should be provided, people reiterated that information "must be age-appropriate" (322AC/GC0/2/4/8) and how this might be achieved, "For younger ones, a nice story picture book. For older ones, a helpline is important" (318C0/2/5). Other suggestions were television; video on website, YouTube or DVD; Facebook and other social media; lessons in school; comic style books and posters, "colourful posters, possibly

Table 4. Information format preference by age of child/grandchild

Format	**Child** n = 900		% of Total	**Grandchild** n = 526		% of Total
	Over 18 *(% within age)*	Under 18 *(% within age)*		Over 18 *(% within age)*	Under 18 *(% within age)*	
Leaflet	62.0	73.2	**64.9**	51.9	63.3	**62.2**
Book	23.8	37.2	27.2	7.7	29.7	27.6
DVD	22.6	39.8	27.0	17.3	25.5	24.7
Phone App	11.8	32.0	17.0	3.8	12.2	11.4
Website	47.7	63.2	**51.7**	38.5	46.6	**45.8**
Web Blog/Forum	12.3	25.5	15.7	7.7	13.5	12.9
Phone support	20.3	21.6	20.7	7.7	21.1	19.8
Person	35.1	38.5	**36.0**	25.0	35.2	**34.2**
Other delivery	5.5	5.6	5.6	3.8	6.5	6.3

annotated, telling a story of a day in the life of a rheumatic grandparent and grandchild" (276AC/GC2/5).

3.5 Delivery of information:-

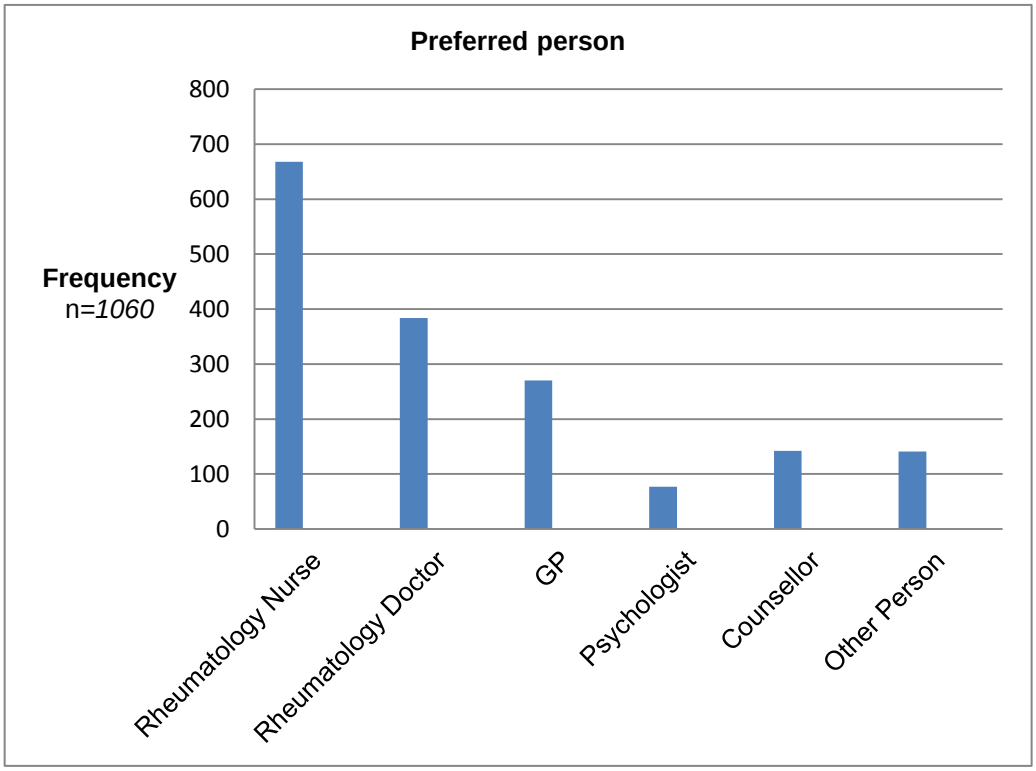
Participants felt that the best people to help them talk to a child/grandchild would be the rheumatology nurse (62.9%), rheumatology doctor (36.1%) and GP (25.8%) see Figure 4. There were no gender differences. Participants with children aged both under and over 18 felt that the rheumatology nurse would be the best person to

help them talk to them about their rheumatic condition, significantly so in the under 18 age group (χ^2 (1, N = 900) = 20.081, *p*= .000 (Cramer's V = 0.149).

3.6 The timing of information delivery:-

Participants suggested that information should be given when the parent or child asked for it (48.1%), at the time of diagnosis (36.8%) or at both instances (11.7%), "or when there are difficulties explaining and understanding" (041AC/GC0/8/10).

Figure 4. Preferred person to help parents/grandparents talk to child/grandchild



3.7 Helping at home:-

Participants who had children/grandchildren aged *under* 18 were asked to indicate whether their children/grandchildren helped around the

house *specifically* because of their rheumatic condition. Few participants answered this section (20.5%), although it was also completed by some with children/grandchildren aged over 18. The amount of help provided ranged from 1 to 120 hours per week, with a median of 4 hours.

Overall, only 25.8% of children (n=900) and 23.6% of grandchildren (n=526) carried out any domestic chores, with the under 18's in both groups providing more help. A small number of children were said to provide personal care for the parent with a rheumatic condition (8.8%) or for themselves (8.7%) or others (1.1%), with the under 18's providing more help. The results were similar for personal care provided by grandchildren to a grandparent with a rheumatic condition (8.0%), although they were less likely to be caring for themselves (4.4%) or others (1.1%).

Free text answers suggested that adult children helped with "driving around" (029AC); "transport to hospital" (081AC); "Opening things"(259C8). Others said that children/grandchildren helped by "keeping spirits up"! (329AC/GC6/5/2). Parents acknowledged they would use their rheumatic condition if necessary to get their children to help out more: "I would expect them to do these as a normal part of growing up, but I use RA as an extra reason for them to help out" (337AC/C15/13).

Some people wanted to explain why their children and grandchildren did not help out: "I do not ask them for help - they do not understand my difficulties, and they do not ask" (604AC/GC0/9/11) and

"The children help with reluctance and don't consciously think of it as helping their mum because of her condition - but in fact that is the reason she needs help. She needs more help than the children actually give" (491C16/15/12).

3.8 Support for young carers:-

Participants were asked if their child or grandchild had ever attended a young carer's support group. Most people did not answer this question (60.7%). Fewer than 1% answered 'Yes' and most did not elaborate beyond this. The remainder (38.6%) answered 'No'.

One person who answered 'Yes' said that her children now aged 18 and 22 years still performed between 24 and 120 hours per week (1 to 5 full days) of caring tasks both for her and themselves. She felt that the best person to help parents/grandparents to talk to their children/grandchildren about their rheumatic condition would come from a "young carer support mentor/befriender ". Earlier in the questionnaire she said:

> "If there had been information, it would have made the journey easier for both my children and myself. My children had to deal with a lot of the unknown for years without help or support.
> Life is now easier but difficult, but when I remember their younger years, they were painful and they lost a great deal of their childhood" (777AC).

3.9 Other comments

Some reiterated or expanded on earlier sections "schools should be aware of children who help to care for parents" (658AC/C17/16), offered support for the study "I think this would be a very good idea, my children have grown up with me having this and understand a lot but have trouble explaining to others" (552C12/9). Others suggested particular areas that should be covered in the information "Info should stress that the sufferer is still the same person inside and, if they can't cuddle at any time, it is not a rejection and another day will bring a different reaction" (625AC/AGC/GC17/16).

4.0 Discussion

It is important to respond to the needs of patients and their families who access health psychology services in rheumatology, by developing such services appropriately. Our study sought to determine attitudes to providing information and resources for children/ grandchildren of people with rheumatic conditions, and obtained views on the content, format, timing and delivery of such information and resources. We also asked adults about the amount of time children/grandchildren might be spending in 'caring' roles for an adult with a rheumatic condition, and the kind of tasks they performed.

The results of our service evaluation indicate that participants were strongly in favour of information about rheumatic conditions being made available to their children/grandchildren in a developmentally appropriate way, for all age groups, something which is not currently

provided. Approximately a quarter of participants' children/grandchildren were said to provide help with domestic chores, with younger (under age 18) providing the most help.

Information from leaflets, websites and 'someone to talk to in person' were the most preferred formats of delivery, available from diagnosis onwards. The information content should include names of conditions, common symptoms, and likely 'consequences' or impact of rheumatic conditions, together with practical suggestions on how the child might help their parent/grandparent. There should be emotional reassurance for the child/grandchild about changes in the parent/grandparent's behaviour. Common treatments used and their monitoring and impact should be outlined. The information could include a health promotion aspect regarding self-care for the child/grandchild that might help to avoid a rheumatic condition in the future, and an emphasis on seeking help early if symptoms should occur. Most participants felt that the rheumatology nurse would be best suited to help them talk to their child/grandchild about their rheumatic condition if required. This discussion will focus upon three key points: the argument for providing information to children, the content and format of such information.

4.1 To tell or not to tell:-

It was clear that participants were keen to protect children from concerns about their health. Interestingly, whilst the majority thought that children would be reassured and helped by the provision of information

and support, those who did not want such information and support available also suggested this was motivated by a desire to protect them from 'worry' or 'burden'.

Studies with parents who have serious or life-limiting physical illness have indicated similar ambiguity in deciding whether to tell their children, and if so, what and how to tell them. Dale and Altschuler (1999) discuss childhood as a time of innocence, noting that disclosing difficult and potentially distressing information to children damaged that innocence. It may be the case that our participants who did not want resources to help them have health-related conversations with their own children/grandchildren felt the same way. As one participant said, *"Children need to enjoy their childhood, and parents and grandparents can tell them as they ask questions" (282).*

Equally, 'not knowing' can carry its own worries, particularly when children have a sense that something is wrong or changed. Children may be unprepared for the impact that a parent's illness may have on the family unit. Parents who try to create a sense of 'distance' between their illness and their child may thus raise both their own and their child's anxiety and sense of isolation, if information is not forthcoming or questions go unanswered (Altschuler & Dale, 1999). Responses suggested that the 'unknown' was far harder to negotiate a path through, hence the overall support for the provision of information. This may explain why our participants suggested that information should be available from diagnosis onwards.

4.2 Content of information: illness cognitions:-

People tend to have individual ways of thinking about health and illness (illness cognitions) which will be based on information and experience from a range of sources, including personal experience of illness, either directly or indirectly. Within health psychology, one illness cognitions model, often known as the Common Sense Model (CSM), proposes that people typically think about illness along five domains: Identity; Cause; Curability/Controllability; Consequences and Timeline (Leventhal et al., 1997), see Table 5. In recent years, researchers have proposed additions to these five domains, namely 'illness coherence' - the degree to which a condition 'makes sense' to an individual and how this might affect adjustment in the long-term; and 'emotional representation', which looks at how emotional responses might affect coping and outcomes, although this may be bi-directional (Moss-Morris et al., 2002); Pimm & Weinman, 1998). Thematic analysis revealed suggestions that closely followed these known domains, including emotional response. Additionally, participants suggested content under the curability/controllability domain, not only in terms of the impact of the condition and its treatment related to themselves, but also from a health promotion/protection point of view for the children.

If the conceptual domains of the CSM indicate the way in which adults tend to think about illness, do children develop the same concepts? And if they do, might this be a useful structure for the development of educational materials? The literature suggests this may be the case. Children's illness cognitions become conceptually more

sophisticated over time, and are linked to their developmental capacity (therefore, not *necessarily* age dependent) and illness experience, i.e. both personal experience and observing it in others (Babooram, Mullan& Sharpe, 2011). Using the Illness Perception Questionnaire (IPQ-R), children aged 11-16 years with juvenile idiopathic arthritis (JIA) perceived causes as genetics (27.1%), immune system (21.3%), accident or injury (15.6%), and infection (15.6%) (Cordingley et al., 2012).

Table 5. Domains of the Common Sense Model (CSM)

Domain	Domain Concept
Identity	The name given to a condition and beliefs about the symptoms that might go with it.
Cause	An individual's ideas about the perceived cause of a condition, not necessarily medically accurate.
Curability/Controllability	Beliefs about whether the condition can be prevented, cured or kept under control and who plays a role in achieving this.
Consequences	What the physical and social impact of a condition might be (and how this might change over time).
Time-line	Predictive beliefs about how long a condition might last (acute *vs* chronic) which again may change over time.

However, even young children demonstrate illness cognitions that reflect CSM domains. Healthy children aged 4-6 years could provide causal explanations for illness, some ideas for cure or resolution, a time line for being unwell and getting better, and specific symptoms to expect (consequences). They could identify 'junk' food and pack healthy and unhealthy lunchboxes, indicating some idea of these concepts, and could identify equipment within the doctor's surgery but did not usually know why they might be used (Goldman, Whitney-Saltiel, Granger & Rodin, 1991).

Whilst both age and developmental level are relevant in the sophistication of children's illness cognitions, the broad categories of the CSM appear to be a useful framework for organising such information. Indeed, it has been suggested that information which slightly challenges a child's level of understanding might help to improve their knowledge of illness, treatment, and disease prevention, something our participants indicated they would like included in the curability/controllability domain for children (Shagena, Sandler & Perrin, 1988). Providing information that can reassure and address possible misunderstandings, for example, ensuring children know that their own behaviour cannot cause or worsen disease outcomes, is easily organised within the CSM (Cross & Rintell, 1999).

4.3 Format of information:-

Leaflets, websites and delivery by 'someone in person' were the most popular choices of format, mirroring current trends for the delivery

of information to adult patients within rheumatology (Arat, Vandenberghe, Moons, & Westhovens, 2015; Arthritis Research UK, 2011). This might suggest our participants answered in terms of those formats with which *they* were most familiar rather than those that might be most appropriate or accessible to their children.

Presently, information for children about difficult subjects such as parental ill-health, witnessing trauma and anticipating or experiencing bereavement is often presented through colourful picture booklets, certainly for the 4 to 12 year-old group (Courtauld & Cobb, 2009; Forrest & Garson, 2008; Holmes, Pillo & Mudlaff, 2002; Johnson, 2009; Thomas, 2000). These are somewhere between leaflets and formal books, and might be what our participants had in mind. Such materials have several advantages: they are designed to be read as a story with a parent/caregiver and allow for questions and discussion; are short and cover only 'essential' topics; and have pictures which younger children like (Huang, Lee, Hu, Gao & O'Connor, 2015). Booklets often include guidance for the parent/caregiver, such as answering questions honestly, not forcing a child to talk more than they want to, and checking their understanding. They are easily carried or stored, and are potentially easy for a child to access by themselves. Some booklets are now available via websites and can be downloaded on multimedia devices like iPad and iPhone (Forrest & Garson, 2008; Johnson, 2009).

The accessibility of information to children can, however, be problematic when information is produced via websites. Whilst even young children may be technologically able, as schools build IT skills, at

home they may well need parental permission and assistance in viewing information and revisiting it, and not all families will have access to the internet. Websites do provide greater scope for more activities that can assist knowledge development, with film graphics, games, quizzes and links to other websites and resources, although these again may need parental supervision (Patient Information Forum, 2014).

One of the problems in assessing the appropriateness and usefulness of current modes of delivery of information to children for our own possible use, is that information of this type is often produced by charities or other organisations, and its evidence-base may be limited. Whilst charities may have medical advisors, or employ professional writers and illustrators, the degree to which children have been involved in developing the content and format of the work, or in peer review is limited (Courtauld & Cobb, 2009).

For example, during the course of this study, a company called *Medikidz* produced a comic book for children about RA, written by doctors and professional medical writers, and peer reviewed by clinicians (Chilman-Blair & DeLoache, 2012). Its target age is unclear, but would appear to be adolescents onwards, and is based upon a single “case study”.

In keeping with research in other chronic illnesses, our participants wished for someone to talk to the child, although it is inconclusive who this person should be or what additional training they would require (Barnes et al., 2000; Forrest et al., 2006). As

parents/grandparents thought that the CNS might be the best person to assist *them* in talking to their child/grandchild, they may also consider the CNS role appropriate for this.

5.0 Implementation issues:-

In practical terms, nurse specialists may have concerns about expanding their current role to accommodate support for children, due to time constraints, lack of training and organizational support for such a role. However, they would be ideally placed to ask parents/grandparents about how they are coping with their parenting and what they have told their children about their health, as this may already occur as part of the clinical interview. Whilst they may be anxious about broaching such questions due to fears about upsetting the patient, Altschuler & Dale (1999) suggested that parents may welcome such questions as a starting point for more focused discussion or signposting to other services (Arat, Vandenberghe, Moons & Westhovens, 2015).

Alternatively, the patient and their family could be referred to psychological services, where they exist. A survey by (Dures et al., 2014) found that psychological provision within rheumatology in England was inadequate, yet required, with further evidence needed for organizational support. Indeed, the participants in our study probably lacked awareness or experience of the kind of help that can be provided by a health or clinical psychologist, therefore they may not have considered them as a potential service option.

Future research should involve families to add depth to these study results, to explore how they talk about parental rheumatic disease, what their children understand, and the content and format of resources.

6.0 Critical appraisal:-

Whilst this study was cross-sectional, it reached a large sample, both locally and nationally, and, to our knowledge, such a study has not been carried out in rheumatology before. Support for the provision of information to children/grandchildren was strong, although it is possible that those disposed to think the research question was a good idea, were more likely to complete the questionnaire, particularly in the responses gathered from “Survey Monkey”™. However, we now have a sound evidence-base in developing resources.

There were certain organizational difficulties encountered in conducting the service evaluation. Firstly, the host NHS Trust requested a more substantial project than the requirements of the PsyD portfolio necessarily intended, which involved obtaining ethical approval from NRES. As this qualified the study for inclusion onto the NHS portfolio study recording system, monthly data entry was required. Some local data administration was available, but often encountered difficulties which had to be managed and resolved.

Secondly, the ethics protocol prevented the researcher from recruiting patients at the local level. Initially we had intended for the questionnaire to be distributed by the receptionist, as patients checked into the rheumatology unit. Receptionists were unwilling to do this,

therefore we had to negotiate with the nursing manager for the out-patient nurses to distribute questionnaires at their pre-consultation check. Unfortunately, this meant that a large number of nurses were involved across the three Trust sites, and we had no record of response rate. Additionally, it was difficult to ensure questionnaires were distributed as intended, prolonging recruitment time.

6.0.1 Personal reflection:-

Conducting this service evaluation has provided me with the opportunity to manage a research project from start to finish, which I had not done before. I have been able to develop skills in a number of areas, such as liaising with patient-focused charities, managing large external databases, attending ethical review meetings, and refreshing my knowledge of SPSS. A further challenge was balancing the requirements of the PsyD, funders, the local NHS Trust and the co-operation of staff. However, ultimately the findings have the potential to produce resources that can be used both locally and nationally, and thus impact on patient and family well-being.

References

Aldridge, J., & Becker, S. (1999). Children as carers: The impact of parental illness and disability on children's caring roles. *Journal of Family Therapy, 21,* 303-320. doi:10.1111/1467-6427.00121

Altschuler, J., & Dale, B. (1999). On being an ill parent. *Clinical Child Psychology and Psychiatry, 4(1),* 23-37. doi:10.1177/1359104599004001003

Arat, S., Vandenberghe, J., Moons, P., & Westhovens, R. (2015). Patients' perceptions of their rheumatic condition: Why does it matter and how can healthcare professionals influence or deal with these perceptions?.*Musculoskeletal Care.* Advance online publication.doi:10.1002/msc.1128

Arthritis Research UK (2011). *Rheumatoid Arthritis.*[Pamphlett].Chesterfield: Author.

Arthritis Research UK (2012). *Annual Review 2011-12*. [Pamphlett]. Chesterfield: Author.

Arthritis Research UK (2014). *Arthritis in the UK - facts and statistics*.[Pamphlett]. Chesterfield: Author.

Babooram, M., Mullan, B.A., & Sharp, L. (2011). Children's perceptions of obesity as explained by the common sense model of illness representation. *British Food Journal, 113(2),* 234-247. doi:10.1108/00070701111105321

Backman, C.L., Del Fabro Smith, L., Smith, S., Montie, P.L., &Suto, M. (2007). Experiences of mothers living with inflammatory arthritis. *Arthritis & Rheumatism, 57(3),* 381-388. doi:10.1002/art.22609

Barlow, J.H., Cullen, L.A., Foster, N.E., Harrison, K., & Wade, M. (1999). Does arthritis influence perceived ability to fulfill a parenting role? Perceptions of mothers, fathers and grandparents. *Patient Education and Counseling, 37(2),* 141-151. doi:10.1016/s0738-3991(98)00136-0

Barlow, J.H., & Wright, C.C. (1998). Knowledge in patients with rheumatoid arthritis: A longer term follow-up of a randomized controlled study of patient education leaflets. *British Journal of Rheumatology, 37,* 373-376. doi:10.1093/rheumatology/37.4.373

Barnes, J., Kroll, L., Burke, O., Jones, A., & Stein, A. (2000). Qualitative interview study of communication between parents and children about maternal breast cancer. *British Medical Journal, 321,* 479-481. doi:10.1136/bmj.321.7259.479

Bowling, A. (2002).*Research Methods in Health: Investigating Health and Health Services* (2nd Ed. pp.341-343). Buckingham: Open University Press.

British Psychological Society (2009). *Code of Ethics and Conduct*. Leicester: Author.

British Psychological Society (2014). *Code of Human Research Ethics*. Leicester: Author.

Chilman-Blair, K., & DeLoache, S. (2012). *Medikidz Explain Rheumatoid Arthritis.* London: Medikidz Publishing.

Courtauld, S., & Cobb, R. (2009). *My Brother and Me.* Buckinghamshire: Ethedo Press.

Cordingley, L., Vracas., T., Baildam, E., Chieng, A., Davidson, J., Foster, H.E., ...Hyrich, K.L. (2012). Juvenile-onset inflammatory arthritis: A study of adolescents' beliefs about underlying cause. *Rheumatology, 51,* 2239-2245. doi:10.1093/rheumatology/kes216

Cross, T., &Rintell, D. (1999). Children's perceptions of parental multiple sclerosis. *Psychology, Health & Medicine, 4(4),* 355-360. doi:10.1080/135485099106090

Dale, B., & Altschuler, J. (1999). 'In sickness and in health': The development of alternative discourses in work with families with parental illness. *Journal of Family Therapy, 21,* 267-283. doi:10.1111/1467-6427.00119

Dudley Joint Strategic Needs Assessment Synthesis (2012). Accessed Jun 04, 2015, at: http://www.dudley.gov.uk/community/initiatives/health-wellbeing/

Dures, E., Almeida, C., Caesley, J., Peterson, A., Ambler, N., Morris, M., Pollock, J. & Hewlett, S. (2014). A survey of psychological support provision for people with inflammatory arthritis in secondary care in England*. Musculoskeletal Care*, *12(3),* 173-181. doi: 10.1002/msc.1071

Faul, F. (2012).G*Power Version 3.1.9.2. Kiel, Germany: Universitat Kiel.

Field, A. (2011). *Discovering Statistics using SPSS* (3rd Ed. pp. 56-58). London: Sage.

Firestein, G.S., Budd, R.C., Gabriel, S.E., McInnes, I.B., &O'Dell, J.R. (2013). *Kelley's Textbook of Rheumatology* (9th Ed. pp. 1109-1719). Philadelphia: Elsevier.

Forrest, G., & Garson, S. (2008). *Mummy's Lump.* Breast Cancer Care: London. Available at: www.breastcancercare.org.uk/information-support/publication/mummys-lump-bcc164

Forrest, G., Plumb, C., Ziebland, S., & Stein, A. (2006). Breast cancer in the family - children's perceptions of their mother's cancer and its initial treatment: Qualitative study. *British Medical Journal, 332,* 998-1003. doi:1136/bmj.38793.567801.AE

Goldman, S.L., Whitney-Saltiel, D., Granger, J., & Rodin, J. (1991).Children's representations of "everyday" aspects of health and illness. *Journal of Pediatric Psychology, 16(6),* 747-766. doi:10.1093/jpepsy/16.6.747

Grant, M.I., Foster, N.E., Wright, C.C., Barlow, J.H., & Cullen, L.A. (2004). Being a parent or grandparent with back pain, ankylosing spondylitis or rheumatoid arthritis: A descriptive postal survey. *Musculoskeletal Care, 2(1),* 17-28. doi:10.1002/msc.53

Hale, E.D., Kitas, G.D., O'Reilly, M., & Vostanis, P. (2016). Should there be information and support available for children whose parents or grandparents have a chronic rheumatic condition? *Rheumatology*, 55 (suppl): i148. doi:10.1093/rheumatology/kew177.001 Retrieved from:

http://rheumatology.oxfordjournals.org/search?submit=yes&tocsectionid=Education&volume=55&issue=suppl%201

Hale, E.D., Treharne, G.J., Lyons, A.C., Mole, S., Mitton, D.L., Douglas, K.M.J., ...Kitas, G.D. (2006).“Joining-the-dots” for systemic lupus erythematosus patients: Personal perspectives of healthcare from a qualitative study. *Annals of the Rheumatic Diseases,65(5),* 585-589. doi:10.1136/ard.2005.037077

Hale, E.D., Treharne, G.J., Norton, Y., Lyons, A.C., Douglas, K.D., Erb, N., & Kitas, G.D. (2006). ‘Concealing the evidence’: The importance of appearance concerns for patients with systemic lupus erythematosus. *Lupus, 15(8),*532-540.doi:10.1191/0961203306lu2310xx

Hale, E.D., Treharne, G.J., Norton, Y., Mitton, D.L., Erb, N., & Kitas, G.D. (2006). ‘Are your hands hurting’? Patients’ perceptions of the impact of lupus on their children and grandchildren. *Rheumatology, 45,* Suppl. 1, il184.Presented at the British Society for Rheumatology Annual Meeting, Glasgow. BHPR Research Poster Prize.

Holmes, M.M., Pillo, C., & Mudlaff, S.J. (2002). *A Terrible Thing Happened.* Washington, DC: Magination Press.

Huang, X., Lee, S., Hu, Y., Gao, H., & O'Connor, M. (2015). Talking about maternal breast cancer with young children: A content analysis of text in children's books. *Journal of Pediatric Psychology, 40(6),* 609-621. doi:10.1093/jpepsy/jsu110

Johnson, J. (2009). *Our Dad Makes the Best Boats.* London: The MS Society.

Kelly, P.A., & Haidet, P. (2007). Physician overestimation of patient literacy: A potential source of health care disparities. *Patient Education and Counseling, 66*, 119-122. doi:10.1016/j.pec.2006.10.007

Kroll, L., Barnes, J., & Jones, A.L. (1998). Editorial: Cancer in parents: telling children. *British Medical Journal, 316,* 880-880. doi:10.1136/bmj.316.7135.880

Leventhal, H., Benyamini, Y., Brownlee, S., Diefenbach, M., Leventhal, E.A., Patrick-Miller, L., & Robitaille, C.(1997). Illness representations: Theoretical foundations. In K.J. Petrie, J.A. Weinman (Eds.), *Perceptions of Health and Illness* (pp. 19-45).Amsterdam: Harwood Academic.

Moss-Morris, R., Weinman, J., Petrie, K.J., Horne, R., Cameron, L.D., & Buick, D. (2002).The revised Illness Perception Questionnaire (IPQ-R). *Psychology & Health, 17(1),* 1-16. doi:10.1080/08870440290001494

National Rheumatoid Arthritis Society (2012). *Family Matters. A major UK wide survey on the impacts on the family of living with rheumatoid arthritis.*[Report]. Maidenhead: Author.

Nevo, B. (1985). Face validity revisited. *Journal of Educational Measurement, 22(4),* 287-293.doi:10.1111/j.1745-3984.1985.tb01065.x/pdf

NHS Health Research Authority (n.d.).Consent and participant information resources page. Accessed Feb 16, 2015, at https://www.hra.nhs.uk/resources/before-you-apply/consent-and-participation/consent-and-participant-information/

Parsons, S., Ingram, M., Clarke-Cornwell, A.M., &Symmons, D.P.M. (2011). A heavy burden: The occurrence and impact of musculoskeletal conditions in the United Kingdom today. The University of Manchester & Arthritis Research UK. Accessed Oct 09, 2015, at http://www.inflammation-repair.manchester.ac.uk/Musculoskeletal/

aboutus/publications/heavyburden.pdf

Patient Information Forum (2014). Summary guide to producing health information for children and young people. Accessed Jan 14, 2016, at http://www.pifonline.org.uk

Pimm, T.J., & Weinman, J. (1998).Applying Leventhal's self regulation model to adaptation and intervention in rheumatic disease. *Clinical Psychology and Psychotherapy, 5,* 62-75. doi: 10.1002/(SICI)1099-0879(199806)5:2<62::AID-CPP155>3.0.CO;2-L

Shagena, M.M., Sandler, H.K., & Perrin, E.C. (1988).Concepts of illness and perception of control in healthy children and in children with chronic illnesses. *Developmental and Behavioral Pediatrics, 9(5),* 252-256. doi:10.1097/00004703-198810000-00002

SPSS. IBM Corp. Released 2011. IBM SPSS Statistics for Windows, Version 20.0. Armonk, NY: IBM Corp.

Stack, R.J., Shaw, K., Mallen, C., Herron-Marx, S., Horne, R., & Raza, K. (2012). Delays in help seeking at the onset of the symptoms of rheumatoid arthritis: A systematic synthesis of qualitative literature. *Annals of the Rheumatic Diseases, 71,* 493-497. doi:10.1136/annrheumdis-2011-155416

SurveyMonkey (n.d.). How it works. Accessed Jan 31, 2015, at https://www.surveymonkey.com/mp/take-a-tour

Thomas, P. (2000). *I Miss You.* London: Hodder Children's Books.

Williams, M. (2001). *Social surveys: design to analysis*. In T. May (Ed.), Social Research: Issues, methods and process (pp. 88-119). Buckingham: O U Press.

Appendix A - Study Information Sheet

NHS TRUST LOGO HERE

Department of Rheumatology
Clinical Research Unit
North Block
XXXXXXXXXX

Date:
Number:
Study reference number: 12/WM/0221

Should there be information and education available for children whose parents or grandparents have chronic rheumatic conditions?

Information Sheet

My name is Liz Hale and I would like to give you some information about a research project that I am carrying out as part of my Doctoral qualification at the University of Leicester.

The research team are myself, Professor XXXX and Dr XXXX from the University, and Professor XXXX from the hospital.

The research is being funded by XXXX UK and The XXXXXX NHS Foundation Trust Charitable Fund.

What is the research about?

People with long-term rheumatic conditions like rheumatoid arthritis, systemic lupus erythematosus (sometimes called Lupus), ankylosing spondylitis and so on, are often given leaflets to read about their condition. These leaflets try to answer some of the questions you, and your family, might ask.

At the moment there is no information available specifically for the **children** or **grandchildren** of our patients who have long-term rheumatic conditions.

We are carrying out a research project to find out if our patients think it would be a good idea to provide this information for children, written in a way that they would understand. We would also like to know the best way to provide this information.

I would like to ask you for a few minutes of your time (5 to 10 minutes) to complete this short questionnaire which asks you for your views. When you

have finished please hand it back to the person who gave it to you or put it into the tray on the reception desk.

Why are you asking me to take part?

I am asking you as I believe you are, or have been, a patient at the rheumatology department at The XXXX Hospitals. It is up to you to choose whether you would like to take part in this aspect of my research project. **The questionnaire does not ask you for your name**.

What if I complete the questionnaire and then change my mind?

You are free to withdraw from this research at any time without giving any reason for your withdrawal. Your care at this hospital will not be affected in any way by your decision to take part, or your decision to withdraw from the research project at a later stage. If you withdraw from the study after completing the questionnaire, I will still need to use the questionnaire data that I have collected from you up to that point.

Is it *really* anonymous?

The questionnaire does not ask you for your name or address so that you can reply anonymously. I have numbered the questionnaires to keep a record of how many we give or send out. We will place a label in your notes to say that you have been given one of the questionnaires, but the completed questionnaire will not be stored in your notes. The completed questionnaires will be kept in a secure place. No-one outside the research team will have access to this information. If you decide to complete the questionnaire I will not notify your GP that you have been involved in this part of our research project.

Will the information I give be kept confidential?

Some of the questions ask you to write an answer in your own words. When I write a report on this research project I may quote some of these statements. As I do not ask you for your name I will do everything I can to protect your confidentiality. If you do mention someone or a place by name, I will change this or delete it so that it will be very difficult for anyone to identify you.

What are the possible disadvantages of taking part?

We hope that completing the questionnaire will cause as little inconvenience in terms of time and effort as possible.

What are the possible advantages of taking part?

While I cannot promise that taking part in this research project will help you or your children (or grandchildren),the information from this questionnaire will help me with the next stage of my study where I will be interviewing families, including the children, to get a more detailed idea of what help we might be able to provide.

If you would like more information, or would like to volunteer to take part in the next stage of the research project, my contact details are given below. There is also a tear-off page on the questionnaire to leave your contact details.

What will happen to the results from this questionnaire?

The results from this questionnaire will be written up into reports which will be sent for publication (e.g. in academic journals), and may be used at conferences and in seminars. We will provide a summary of the results from the study in due course which you may request by contacting me.

Have you had permission to do this research?

Before this research project was allowed to happen we asked an independent review panel called the National Research Ethics Service (NRES) to look at our proposal in detail. We have been given permission by the West Midlands - Staffordshire NRES Committee to carry out this research.

What if I have any questions now or in the future?

If you would like to request any further information or talk to a member of the research team, please ask the receptionist or nurse if anyone is available today. If not, please leave your contact details with them.

If you would prefer to talk to the researcher by telephone, please call, write or email to:

Ms Liz Hale
XXXXX

OR

The Research Team:
XXXXX

Thank-you for taking the time to read this information sheet

(Information sheet for questionnaire v2 31/08/2012)

Appendix B - Questionnaire

Should there be information and education available for children whose parents or grandparents have chronic rheumatic conditions?

Please write your answer or tick the box that applies

Your Age:

Male [ ] Female [ ]

Marital status:

Single [ ] Co-Habiting [ ] Married [ ] Separated [ ] Divorced [ ]

What is your highest educational qualification?

Postcode:

What is the name of your rheumatic condition?*(for example"rheumatoid arthritis")*

When was this diagnosed?

Do you have children?

If YES, what are their ages?

Do you have grandchildren?

If YES, what are their ages?

Do you think that there should be information available for children (or grandchildren) whose parents (or grandparents) have a chronic rheumatic condition?

YES [ ] NO [ ]

Can you tell us in more detail why?

Please write here:

What kind of information do you think would be useful for the children to have about a parent (or grandparents) rheumatic condition?

Please write here:

How should this information be delivered? *(Please tick all that apply)*

Leaflet	[ ]	Website	[ ]	Other:
Book	[ ]	Website Blog or Forum	[ ]	
DVD	[ ]	Telephone support line	[ ]	
Phone App	[ ]	Someone to talk to in person	[ ]	

Who would be the best person to help parents (or grandparents) talk to their child (or grandchild) about their rheumatic condition? *(tick all that apply)*

Rheumatology Nurse	[ ]
Rheumatology Doctor	[ ]
GP	[ ]
Psychologist	[ ]
Other Counsellor	[ ]
Other person	[ ]

When should information be given?

When the parent or grandparent is diagnosed	[ ]
When the parent or child ask for it	[ ]
Other times (please state when)	

Please continue if you have children or grandchildren up to the age of 18 

If your children or grandchildren help you around the house, specifically because of your rheumatic condition, what kind of tasks do they do?

(please tick all that apply)

Domestic chores *(like cleaning, emptying bins, dusting)* [ ]

Preparing food [ ]

Personal care - For you [ ] For themselves [ ] For others [ ]

Gardening [ ]

Shopping [ ]

Other *(please state)*

Roughly how many hours per week would they spend doing these tasks?

Has your child (or grandchild) ever attended a young carer's support group?

Any other comments you would like to make?

Appendix C - Ethical Approval

Revised 12/9/12 – Document table updated

NRES Committee West Midlands - Staffordshire
HRA NRES Centre Manchester
XXXX
Telephone:
Facsimile:

10 September 2012

Ms Elizabeth Hale

Chartered Health Psychologist

The XXXX NHS Foundation Trust

Dear Ms Hale

Study title: **Children of parents with chronic inflammatory musculoskeletal diseases: Experiences, needs and resources.**

REC reference: **12/WM/0221**

Thank you for your letter of 03 September 2012, responding to the Committee's request for further information on the above research and submitting revised documentation. The further information has been considered on behalf of the Committee by the Chair.

Confirmation of ethical opinion

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised, subject to the conditions specified below.

Ethical review of research sites

NHS sites

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

Non-NHS sites

Conditions of the favourable opinion

The favourable opinion is subject to the following conditions being met prior to the start of the study.

Management permission or approval must be obtained from each host organisation prior to the start of the study at the site concerned.

Management permission ("R&D approval") should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements.

Guidance on applying for NHS permission for research is available in the Integrated Research Application System or at http://www.rdforum.nhs.uk.

Where a NHS organisation's role in the study is limited to identifying and referring potential participants to research sites ("participant identification centre"), guidance should be sought from the R&D office on the information it requires to give permission for this activity.

For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.

Sponsors are not required to notify the Committee of approvals from host organisations

It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).

Approved documents

The final list of documents reviewed and approved by the Committee is as follows:

Document	***Version***	***Date***
Interview Schedules/Topic Guides	Semi Structured Interview Guide-Parents 1	20 June 2012
Interview Schedules/Topic Guides	Semi Structured Interview Guide-Child 1	20 June 2012

Investigator CV	Ms Elizabeth D P Hale	03 July 2012
Investigator CV	Professor Panos Vostanis	14 June 2012
Investigator CV	Dr Michelle O'Reilly	14 June 2012
Investigator CV	Professor George Kitas	14 June 2012
Other: Letter from Funder		22 May 2012
Other: Reviewers feedback and responses	1	15 June 2012
Participant Consent Form: Assent Form	1	14 June 2012
Participant Consent Form: Patient and Children	2	20 August 2012
Participant Consent Form: Patients' Spouse/Partner/Significant Other	2	30 August 2012
Participant Information Sheet: Patients' Spouse/Partner/Significant Other for Interview	2	31 August 2012
Participant Information Sheet: Patient and Family for Interview	2	30 August 2012
Participant Information Sheet: Information for Questionnaires	2	31 August 2012
Participant Information Sheet: Information for Questionnaires (Charitable Organisations)	1	31 August 2012

Participant Information Sheet: Children	2	31 August 2012
Protocol	1	14 June 2012
Questionnaire	1	14 June 2012
Questionnaire: Charitable Organisations	1	25 June 2012
REC application	106881/339969/ 1/378	09 July 2012
Response to Request for Further Information		03 September 2012

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

After ethical review

Reporting requirements

The attached document "After ethical review – guidance for researchers" gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports
- Notifying the end of the study

The NRES website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

Feedback

You are invited to give your view of the service that you have received from the National Research Ethics Service and the application procedure. If you wish

to make your views known please use the feedback form available on the website.

Further information is available at National Research Ethics Service website > After Review
12/WM/0221 Please quote this number on all correspondence

With the Committee's best wishes for the success of this project

Yours sincerely

xxxx

On behalf of

Dr XXXX

Chair
Email: XXXX

Enclosures: "After ethical review – guidance for researchers" *[SL-AR2]*

Copy to: XXXX

CHAPTER 3

Research Report

"We do try and do everything that other families would do":

How families manage and talk about parental rheumatic disease.

Introduction

1.0 Overview:-

The rheumatic diseases are a group of over two hundred, mainly autoimmune, chronic health conditions. Most of these are 'inflammatory' in nature and can affect every system of the body; commonly the joints and connective (soft) tissues. As well as pain and swollen joints, people can experience debilitating fatigue and disrupted sleep. Some of the most common are rheumatoid arthritis (RA); psoriatic arthritis (PsA; the spondyarthropathies including ankylosing spondylitis (AS) and the connective tissue disease systemic lupus erythematosus (SLE). Major adjustments to work and social roles may be necessary. Treatment is provided by a multidisciplinary healthcare team. The course of these diseases is unpredictable, with periods of quiescence interspersed with more active disease 'flares'. As yet, there are no cures for any of these conditions (Hill & Ryan, 2000).

There can be a misconception that these conditions only affect people of mature years - in fact they often onset during the childbearing years, when people are considering becoming or are already parents (Parsons, Ingram, Clarke-Cornwell, & Symmons,2011; Stack, et al., 2012). Thus, the onset of such a chronic illness in the context of parenting may present some unique challenges. Whilst there are 'patient education' leaflets for adult patients to help them understand their condition and its treatment, there are no similar information resources

for the young children of these patients (Arthritis Research UK, 2011a; 2011b; 2013).

We do not know whether the provision of information to young children about their parents' rheumatic condition would be welcomed by either parents or their children. As far as we are aware, this is an area yet to be fully investigated. Research in other chronic health conditions, such as breast cancer, suggests that it would be useful and received positively (Forrest, Plumb, Ziebland, & Stein, 2006). A recent study carried out for the Department for Education (2016), focusing on the lives of young carers in England, found that young carers said they wanted to understand their parents' condition better. Parents themselves said that they struggled to discuss their condition with their children, particularly when they were aged under ten or eleven (Aldridge, Clay, Connors, Day, & Gkiza, 2016).

1.1 Managing 'normal' family life:-

The provision of information may help children not only understand their parent's rheumatic condition, but also the impact and responses to it. One of these responses may involve how the family construct, and continue to manage, their 'normal' life (Deatrick, Knafl & Murphy-Moore, 1999). In the sociological literature, authors have examined how individuals and families might experience and cope with the onset of illness for some time. Often this work has focused upon the concept of stigma, both as an acquired state (such as illness and disability), or as a state that otherwise might identify you as 'different' or

'not normal' such as race or sexual orientation. Goffman, (1968) has referred to this as 'spoiled identity'. In order to avoid the stigma of not being considered 'normal', families may manage their identity carefully. Being open about individual disability may require courage; feeling comfortable with your family being identified as having a disabled member may be quite another (Swain & Cameron, 1999).

Other researchers such as Bury, (1982) have focused upon how chronic illness disrupts the existing life 'biography' (biographical disruption) and the sense of bewilderment this can cause. Whilst treatments for rheumatic disease have developed significantly in the twenty-first century with the advent of 'biological therapies', slowing or preventing much of the disability Bury was writing about, certain elements hold true. Rheumatic disease remains a "significant illness" with "uncertain outcome" (Bury p.168), and symptoms still fluctuate without warning. In this context, the individual has to adjust to a new biography, and decide how this new biography will be revealed within the family, shaped in the social world, and be visible to others. All of which may be very confusing for a young child.

Adults in the childbearing years may have particular concerns about how to shape this new biography, fearing the perceived stigma associated with conditions that are often incorrectly associated with advancing years and disability. For example, the general public continue to hold inaccurate views about rheumatic disease, confusing the various types and their age of onset (Bury, 1988; Simons et al., 2017). Additionally, a survey carried out on behalf of SCOPE (Aiden

&McCarthy, 2014) suggested that 85% of the British public felt that disabled people experienced prejudice. It is perhaps not surprising that individuals with disabilities may choose to preserve the family identity of 'normal' as far as possible; walking a line between who they are, and who they are choosing to present to the wider social world.

It has been suggested that in order to manage these issues, individuals and families living with a chronic illness engage in a process that has been termed 'normalisation' (Robinson, 1993). It is worth pointing out that individual's and families define their own normal that works for them (Rehm & Franck, 2000). Normalisation observes how a family preserves or occasionally redefines their family life as normal, or as normal as it can be, in the face of change or challenge. In adult patients with RA, complex interactions have been observed, with normalities often 'shifting' as life, symptoms and treatment change (Sanderson, Calnan, Morris, Richards & Hewlett, 2011).

The concept of normalisation has been explored in families where either the parent (Donalek, 2009) or child (Hart, et al., 2016) is chronically unwell, or both (Rehm & Franck, 2000). Normalisation is often a balance between preserving and foregrounding *positive* pre-existing family routines, events and behaviours as the family normal, whilst *minimising* the difficulties that this might actually present. Where difficulties do eventually arise, the family narrative may be altered to incorporate the change into the new family normal. It is therefore a dynamic and long-term process. What is unclear however, is how families with young children experiencing parental rheumatic disease

manage these situations, particularly when there are no resources for either parent or child to assist them.

1.2 Involving children in research:-

In the United Kingdom (UK) children are increasingly encouraged to form partnerships with their own healthcare providers in order to facilitate informed decision-making when the child themselves are ill (General Medical Council, 2007). Given that children deemed 'competent' are allowed a voice in determining treatment and quality of life issues for themselves, it would seem reasonable that they should also be able to access appropriate information and education about a parent's illness which will also impact upon them. Furthermore, they should be encouraged to take part in research that asks them directly for their views about parental illness and its impact, rather than using parents' as sole 'proxies' for their views (Dixon-Woods, Young & Heney, 1999). Under the United Nations Convention on the Rights of the Child, a child, defined as any person under the age of 18, is entitled to certain rights. Article 12 of the convention states that children have the right to be heard, and that their views must be taken equally into account in all matters that affect them. Indeed, Article 13 gives the child the right to freedom of expression and the right to "seek, receive and impart information and ideas of all kinds, regardless of frontiers, either orally, in writing or in print, in the form of art, or through any other media of the child's choice" (UNCRC,1990).

Children therefore have the right to take part in research that will develop educational materials and interventions that are designed ultimately for their benefit. In fact they should play a key role in the shaping of the content and development of those materials. Children are not ‘mini-adults’ and materials developed *by* adults *for* children may lack key components and hence produce less effective outcomes. A key objective of the present study therefore, was to include the perspective of young children aged between seven and eleven, living within a family where a parent had been diagnosed with an inflammatory rheumatic condition.

The aim of this study was to explore:

How the diagnosis and impact of parental rheumatic disease has been understood, talked about, and managed within families who have young children. In this context we also wanted to know whether providing information and /or support for children about parental rheumatic disease would be useful. If so, suggestions for format, content, and timing of availability were sought.

Method

In order to achieve the aims of our study, our objective was to use a qualitative design informed by an interpretivist framework, employing semi-structured interviews and visual data collection methods. For a wider discussion of the chosen approach, see mandatory Appendix A "Statement of Epistemological Position".

2.0 Participants and sample size:-

Participants were recruited purposively with the aim of obtaining some typicality of experience, as well as depth of information (Braun & Clarke, 2013). Ideally, a sample should produce data that is both "rich" and "thick", having not just quantity but quality to it (Fusch & Ness, 2015, p. 1409). There is a debate about the sample size required for a qualitative methodology, with even experienced researchers noting "it depends" (Baker & Edwards, 2012, p. 42). As qualitative work is exploratory in nature, with no hypotheses to test, we do not have the equivalent of power calculations available (or necessary) to make statistical analyses 'work'. This means that we do not necessarily know at the outset of data collection how large or small our sample may be, unless we intend to specifically use a single case. Additionally, we are not aiming to 'generalise' to a wider population, rather, we are aiming for depth of understanding and the potential transferability of findings (Braun & Clarke, 2013).

Sample size is often determined by the marker of data saturation, when no new codes or themes are being generated (thus new participants provide no new information). However, there remains difficulty in specifying initial sample size at the outset with which to achieve this, as different approaches require different sample sizes. For example Francis et al. (2010) suggests that a sample of 10 plus a possible three extra *may* be required to achieve saturation within a single code, although they relate this specifically to theory-based content analysis.

Guest, Bunce and Johnson (2006) calculated that between 6 and twelve interviews may be adequate for homogenous purposive samples. After 60 interviews, they found that 94% of codes occurred most often within the first six interviews; with 97% occurring after 12. They concluded that very little is added or missed after the first six interviews, and data saturation had occurred, for them, by 12 interviews when no new information was forthcoming. Their work builds upon the concept of 'consensus theory' (Romney, Weller & Batchelder, 1986); a highly quantified technique that attempts to estimate sample size requirements for qualitative work, firmly established in a realist ontology, i.e. 'truth' exists. Guest et al. argued that although their qualitative work (which asked for perceptions and experiences), was not necessarily as 'realist' in its intention, nevertheless revealed common ground across their sample, which comprised the 'truths' of their daily realities.

For the purposes of this study therefore, we anticipated a range of approximately 6-13 families would be required before data saturation would be achieved; data saturation was noticed after 8 interviews (Baker & Edwards, 2012; Braun & Clarke, 2013). A further three interviews were added for confirmation.

2.1 Inclusion/Exclusion criteria:-

All male and female adult patients (>18 years), attending a local secondary care rheumatology service with a confirmed diagnosis of inflammatory arthritis or connective tissue disease (such as SLE; RA; PsA or AS), who had one or more children aged between seven and 11

years, were eligible to take part in the study. We aimed to recruit families with children at the lower, middle and end of the age range stated. As we wanted to be open to involving a range of families that would reflect population diversity, a family is defined as any combination of patient and child(ren) as above, and their spouse/partner/significant family member (whether legally related or not).

As patients need time to adjust to the impact of the onset and diagnosis of a rheumatic disease and become familiar with the demands of treatment, monitoring and healthcare interactions, we did not invite patients with less than a six month diagnosis to participate. Additionally, families where the patient, partner/other or child had a known diagnosed severe mental illness, where mental capacity to understand and give informed consent was in question, or where understanding and speaking English would be limited, were not approached to take part.

Patient and family reactions to an inflammatory rheumatic condition may not necessarily match objective indices of disease, therefore by including families with a range of disease duration, we were able to obtain experiences of adaptation and coping that were not necessarily related to disease ‘severity’. As inflammatory rheumatic conditions can onset during early to mid-adulthood, when patients may be either thinking about or will already have started a family, we included families both with relatively recent disease onset (therefore, a ‘change’ to usual family life), and those with longer disease duration (families who have ‘grown-up’ with the disease). Participant and interview details are contained in Table 1.

3.0 Procedure:-

Participant recruitment commenced in November 2012 and ended in December 2013. Semi-structured interviews and visual data collection took place between March 2013 and January 2014.

Patients attending the rheumatology department of the local hospital for a routine appointment were approached by a member of their healthcare team to take part in the study. If the patient expressed interest, they were asked to discuss participation with their family, and were given an information sheet for themselves, their partner/spouse/other and in a format appropriate for their child(ren), (see Appendices B & C). Care was taken to ensure that the information sheet for children was written in language that this age group (UK Primary) would understand, by using an online tool that combined several readability indexes to check that it was pitched at the mid-point of the year group. The calculator suggested that the information sheet would be appropriate for children in Year 3-4 (US Grade 2-3) which meant that it should have been understandable to children from the age of 7 (Readability Calculator, www.online-utility.org).

Patients had the opportunity to discuss the study further with the researcher, if they wished on the day of recruitment, otherwise their contact details were passed to the researcher after the clinic. In line with ethical requirements, the patient had at least 24 hours to discuss the study with their family, or contact the research department for more information, before contact was made to schedule an interview.

Table 1. Participant family groups, indicating rheumatic disease, disease duration, age of children and participation in interviews.

Participant family groups*	Parent with rheumatic disease[a]	Rheumatic disease	Disease duration	Present at interview #1	Present at interview #2	Visual data produced by child
1. PHILLIPS James[a] Kate **Luke** (9 years)	Father	RA	7 years	James Kate Luke	N/A	 Drawing
2. ROBERTS Beatrice[a] Alastair **Tony** (9 years)	Mother	Childhood Stills disease. RA/AS overlap	36 years	Beatrice Alastair Tony	Beatrice *not present* Tony	 Drawing
3. THOMAS Matt[a] Helen **Belle** (7 years)	Father	RA&OA	1 year	Matt *not present* Belle	Matt *not present* Belle	 Drawings
4. JACKMAN Grace[a] Crwys **Sara** (7 years) Bryn (5 years)	Mother	PsA	9 years	Grace Crwys Sara Bryn	Grace *not present* Sara Bryn	 Writing; Drawings; Booklet
5. STEVENS Gail[a] Simon **Ruth** (9 years) **Neil** (9 years)	Mother	RA	27 years	Gail Simon Ruth Neil	Gail Simon Ruth Neil	 Drawing Drawing
6. BAXTER Douglas[a] Felicity **Theo** (11 years)	Father	RA	16 years	Douglas *not present* Theo	Douglas *not present* Theo	 Poster
7. DRAPER Teresa[a] Sam (adult son) (children °/18; 20; 17; 11)	Mother	RA	4 years	Teresa Sam *None present*	N/A	N/A
8. MORRIS Clare[a] Elijah (children 18; 16; 14; 11; 8)	Mother	RA	2 years	Clare *not present* *none present*	N/A	N/A
9. UNDERWOOD Joy[a] Charles **Elise** (9 years) Joel (6 years)	Mother	RA	6 years	Joy Charles Elise Joel	Joy *not present* Elise *not present*	 Poster
10. TIVERTON Laura[a] Donald **Harry** (11 years) Poppy (6 years)	Mother	Mixed CTD/SLE overlap	6 years	Laura Donald Harry Poppy	Laura *not present* Harry Poppy	 Poster
11. WALLACE Karen[a] Paul **William** (7 years Oscar (4 years)	Mother	RA/SLE overlap	12 years	Karen *not present* William *not present*	N/A	 None

Note. *all names are pseudonyms; Child's name in bold is consented study participant.
RA, rheumatoid arthritis; OA,osteoarthritis; SLE, systemic lupus erythematosus; AS, ankylosing spondylitis;CTD, mixed connective tissue disease; PsA, psoriatic arthritis.

Interviews were scheduled for a date and time convenient for the families - usually after school and/or work. No family requested a home interview. Where spouse/partner did not attend eventual interviews, this was always due to work commitments.

3.1 Setting:-

The interviews were carried out in a small, quiet meeting room situated to the rear of the Unit where patients attended for their out-patient clinic, and so was familiar to the patients, and in many cases to their partners/spouses. This area has no access to members of the public. A non-clinical office directly opposite was also available for families who preferred to be interviewed individually, to act as a waiting area; in practice only one family preferred this option. Checks were made before the interviews commenced that audio-recording devices performed satisfactorily in these areas, and "Quiet, interview in progress" signs were placed on the entry doors. Refreshments, snacks and fruit were available for the families; this allowed them to relax after work and school, and provided an initial 'ice breaking' opportunity (Cree, Kay & Tisdall, 2002).

4.0 Data collection:-

The semi-structured interview guides used for the families are shown in Tables 2&3.These provided a guide to the topics of interest according to the existing literature, and allowed the researcher to pursue interesting threads that arose within the interview, change the order of questions, and rephrase questions as necessary (Braun &

Clarke, 2013). In practice, as most families were interviewed together the guides were amalgamated.

At the start of the interview the researcher showed the child(ren) some cards that they could use if they did not wish to speak at any point. The cards said "NO" to indicate they did not wish to answer a question, or could shake their head; "STOP" would end the interview immediately; "??" indicated they did not understand a question, and "ME" indicated they wanted to ask a question themselves, perhaps when interrupting seemed difficult. In practice no child used the cards (Hemming, 2008).

Interviews were digitally audio-recorded and transcribed verbatim by a known professional transcriber with facilities for secure data transfer and storage, using researcher specifications for layout. Digital data files allowed the researcher to listen to the interviews with the transcriptions for analysis. The transcriptions are available as Addenda.

At the start of the interview as part of the 'ice-breaking' process, children were given a pre-printed "spider" diagram with empty circles titled "About ME" (see Figure 1). The researcher encouraged the child(ren) to fill in each box information about themselves, or hobbies they might enjoy. The researcher also completed one to demonstrate how it might be used. These enabled the child(ren) to start to feel comfortable talking about themselves, and provided some useful suggestions for additional topics to follow during the interview. For

Table 2. Interview guide - Parents

1. Can you tell me a little bit about yourself? *Icebreaker questions: age; marital status; employment; disease name (as patient understands it); children's names and ages.*
2. I'd like to ask you a few questions about your [name of rheumatic condition] if that's ok. Can you remember when you received your diagnosis? *To cover onset of symptoms and diagnosis timeline. To establish: relationship with spouse/partner at that time, or if relationship commences later; whether the child(ren) are already in the family (pre-diagnosis) or if the decision to have children occurs later (post-diagnosis).*
3. If applicable: **Did the onset of [condition] affect your decision to become a parent?** *Parenthood: considerations in becoming parents.*
4. Has your [condition] impacted on your role as a parent? *To establish in what ways their condition has had an impact (or not). What are the positive ways; Are there any negative ways? Is there anything as a parent you find difficult to do or have had to stop doing?*
6. Do you think that your [condition] has had an impact on your children? T*o establish both positive and negative perceived outcomes.*
7. Do you tell the children when you feel unwell or are having a bad day? *What do you say/ What stops you from telling them?*
8. Do the children help you specifically because of your [condition]? *What kind of things do they do – for themselves; for others?*
9. How do you refer to your [condition] within the family? Do you have a 'nickname' for it?
10. What do you think your children know about [condition]?
11. Do the children ask questions about [condition]?
12. If applicable: **How do you answer those questions?** *To establish what kind of things the parents say; are they open or are questions deflected/minimized - can they explain why? When did they/will they start to talk to the children about it.*
13. What would help you to talk to your children about [condition]? *To establish what kind of resources would be useful; how confident parents are in their own knowledge.*
14. What kind of information should be given to the children? *To establish what topics should be covered.*
15. How should this information be given? *To establish format of delivery of information – book/DVD/App/website/blog etc*
16. When should this information be given? *To establish best timing of delivery i.e. at diagnosis or later?*
17. Who should give this information to the child? *Parents? Another person outside the family?*
18. Would it be useful for your child to have someone to talk to outside the family for support? *Can you explain who this might be?*
19. Are you aware of any support services that your child could access? *Have any ever been used? Would they like any information?*

instance, a child who enjoys swimming or football might have to pursue these activities with the parent who does not have a rheumatic condition; they may thus be curtailed, or no longer take place because of it.

4.1 Visual data collection:-

All children who attended the interviews (including younger siblings who asked for them) were given a coloured folder which contained A4 paper, together with a set of felt pens and a small tin of coloured pencils in a range of colours. Children were given these during the first interview to facilitate the discussion if necessary, as some may find other forms of expression easier than others. Over reliance on one form of data collection (interview) could introduce bias, particularly when children may not have the verbal skills to answer questions in the way that they want to, particularly if they feel upset, anxious, or fear the parent's reaction - such as upsetting the parent or giving the 'wrong' answer; which may effectively silence them (Eiser & Twamley, 1999; Morrow, 2008; Youssef, Salah, Salem & Megahed, 2010). Additionally, the power dynamic between the researcher and the participant may be particularly important in qualitative research, and even more so where children are involved. The option of producing a drawing can give a child the opportunity to interpret or introduce a topic that is important to them, that they might find difficult to do otherwise (Hemming, 2008; Prosser & Loxley, 2008). In practice, no child used the paper and pens/pencils during the interview.

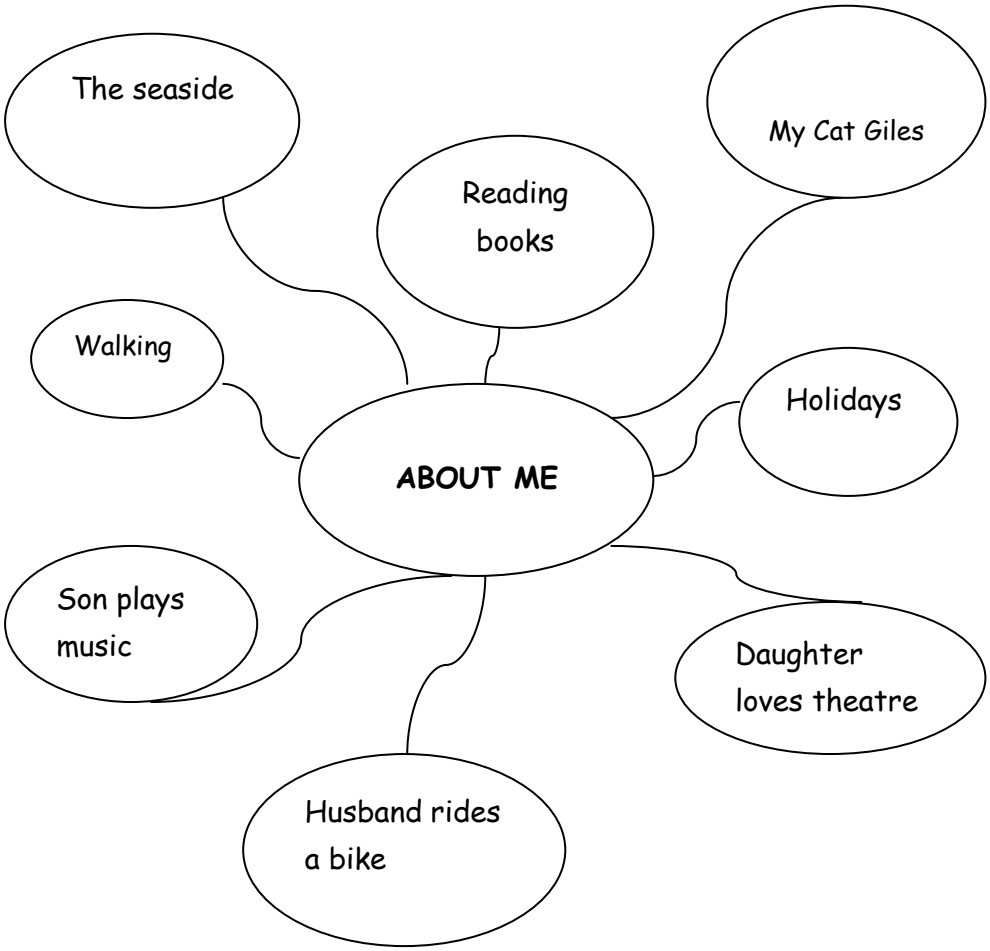
At the end of the interview, children were asked if they would like to take the paper and materials home, and produce either a drawing or poster. They were asked to draw something that was important to them,

Table 3. Semi-structured interview topic guide - child

1. General icebreaker questions to establish rapport and put child at ease:*(Name, age, hobbies, favourite toys/games, favourite subjects at school etc., establishing family members). These can be picked up on in future questions if rheumatic condition impacts on these. Facilitate by drawing if required, using "About ME" or preferred method.* **2. Understanding of parent's rheumatic condition [using 'pet' name if appropriate]:** *What they know about it. Do the parents talk about it and answer their questions.* **3. Impact of parent's rheumatic condition:** *What it is like for mum/dad/other. What it is like for them. How they feel about it.* **4. What is it like when Mum/Dad/Other is poorly?** *What do you do when Mum/Dad/other is poorly? Are there any good things about it? Are there any not so good things about it? (what).* **5. Information and Education:** *What would be useful to know and help other children.* **6. What would be the best way of telling children about [rheumatic condition] if their parent or grandparent had it?** *(if necessary suggest books, posters, DVD's, App's etc but allow child free rein).* **7. What kinds of things should we tell them about?** *Follow discussion so far: Suggest immune system if child has been told; about how parents might behave (grumpy/sad); about how they might help?* **8. Would it be a good idea to have someone to talk to about Mum's or Dad's [rheumatic condition]?** *What kind of person should this be (possibly: in a hospital like a nurse, or somewhere else?)* **9. Would you like to meet other children whose parents have [rheumatic condition]?** *Can you explain why you would like that/not like that?* **10. When should children have information about [parent's rheumatic condition]?** *When parents know they have it? when child(ren) are a certain age? When child(ren) ask questions?*

or that they wanted to convey, about having a mum/dad with 'arthritis' (we used the family terminology for the parent's condition). Alternatively, they were invited to create a poster that showed what they would want another child of their age to know if they had a parent with the same condition. We checked that all the children were familiar with making posters in school that conveyed some information to others.

Figure 1. "About ME"



In practice, only one child (William, aged 7) preferred not to produce a drawing or poster, although he gave details about what he thought a poster should, and should not, contain. Children were allowed to keep all their materials after the interview, whether they had produced any drawings or not.

After approximately a month, the families returned for a second semi-structured interview, so that the child could discuss the content of their drawings. The interviews were audio-recorded and transcribed as

described earlier. An A4 acetate 'grid' was prepared that fitted over the drawings/posters, to help the researcher locate the part of the drawing being spoken about for the audio-recording. The visual data collection was not used as a method of 'triangulation' (gathering data in different ways) in the sense of trying to obtain an overall 'truth', as this sits uncomfortably within an interpretative framework; rather, the method was used to get rich data where verbal accounts may prove difficult. Even with advance information, thought and conversation with the parent, the research interview may stimulate thinking, questions and ideas that the child needs time to process and capture. The second interview (facilitated by a drawing or poster) allowed time for this to happen. One family opted to be interviewed individually (the Phillips family) and Luke, aged 9, did his drawing whilst waiting, and talked about this within his interview. They did not therefore return for a second interview.

5.0 Ethics:-

Ethical approval was obtained from the NHS Health Research Authority (HRA), National Research Ethics Service Committee West Midlands - Staffordshire, UK. Reference number 12/WM/022(See Appendix C in Service Evaluation). Additionally, the Code of Human Research Ethics (British Psychological Society, 2014), and Code of Ethics and Conduct (British Psychological Society, 2009) were adhered to. The information sheets followed the guidance laid down by the NHS Health Research Authority (HRA) on *consent and participant information* (n.d.[a]).

5.1 Consent, Assent and Confidentiality:-

Special attention was given to the consenting of the child(ren) in the family to take part in the research (Modi et al., 2014). Access to the child(ren) as research participants could only come in the first instance from the adult patient who was asked to discuss the study with their families and decide whether to take part. This did not preclude the possibility that a child might not actually wish to take part (Cree et al. 2002). The Medical Research Council (MRC) guidance on competence to give consent to take part in research notes that legally in England, Wales and Northern Ireland a person becomes an adult at the age of 18, and between the ages of 16 and 18 are generally considered to be competent to give consent. The situation is less clear for 'minors' under the age of 16, as no legal statute exists for their right to give consent to take part in research.

However, the principle of 'Gillick competence' is an example of case law that where a child has sufficient intelligence and understanding to grasp what is proposed and required, and they can clearly weigh-up the options given to them, they can give their own consent to take part in research (or not) (Medical Research Council, 2004; Gillick v West Norfolk and Wisbech AHA (1986). The child(ren) were, therefore, given the opportunity to talk through the aim and objectives of the study, ask questions and sign assent forms (Appendix D).The parent with legal authority to give consent for the child(ren) was also asked to read and sign a consent form on behalf of their child(ren). It was made clear to the child(ren) that they could withdraw consent,

however temporarily, at any time during the data collection process. The patient and partner/spouse/significant other were also asked to read and sign consent forms detailing the requirements of the study for themselves.

During the informed consent/assent process, families were reassured that all interview and visual data would be anonymised and any identifying names, places and contacts would be removed.

5.2 Safeguarding welfare of children and adults:-

Participating in research of this nature may be particularly challenging, and give rise to difficult and painful experiences. Details of local organisations that might be helpful to parents and children were available (Young Carer organisations). During the informed consent process, both adults and child(ren) were made aware that there were certain instances such as self-harm or child protection, where the confidentiality of the research interview would be broken.

5.3 Participant Recompense:-

The HRA publish guidance for researchers on what would constitute coercion, inducement and reasonable recompense for adults and children taking part in research in the UK NHS (HRA Ethics Guidance: *Payments and Incentives in Research*, n.d.[b]).It is suggested that families taking part in research that do *not* involve medicinal products should have their expenses refunded to them. Therefore, in an effort to reduce barriers that would prevent the research being as inclusive as possible, participants were given the opportunity to have

their travel costs refunded each time they attended for interview. The researcher was not involved in making the refund, nor was aware who claimed or not. The ethics committee granting approval for this study required that this section of the information sheet be highlighted.

The information sheet made clear that the art materials provided to the children were part of the research process and were not considered an inducement to take part in the study. After the first interview, children were given a 'certificate' for taking part. They (and any younger siblings present) were allowed to choose a 'sticker' to wear, which had 'well-done' or a similar affirmation to thank-them for their contribution.

6.0 Method of analysis:-

The data were analysed using an inductive thematic analysis (TA) (Braun & Clarke, 2006; 2013), chosen during the planning stages of the study. Inductive analysis is a 'bottom-up' style of analysis where the data drive the analysis, rather than a deductive analysis which takes a 'top-down' approach where existing theory or concepts drive analysis.

Thematic analysis is one of a range of approaches for analysing qualitative data, is accessible to novice researchers as a 'foundational' method, appropriate for analysing interview data, and interview data that has been generated via a visual method (drawing or poster). Additionally, TA, as emphasised by Braun and Clarke (2013), is "just a method" (p.178) and, although aligned to no particular ontological or epistemological frameworks, fitted our perspective well. We considered other approaches that look for patterns or themes across data (such as

discourse analysis; grounded theory and interpretative phenomenological analysis), but these did not appear as flexible, and were not necessarily appropriate for answering the research questions.

Other considerations such as taking a realistic view to participant accessibility, timescale, and methodological and analytical expertise were also important. For example, approaches utilising a variety of forms of discourse analysis would require considerable training and practice, and were considered beyond the scope and timescale of the study. Grounded theory (both a methodology and a means of analysis) also requires availability and accessibility of participants, which we felt were unlikely to be achieved within the timeframe for project completion. Additionally, although Corbin and Strauss (2008) acknowledge theories may not provide a view of reality per se (p.55), the approach is essentially a realist one, and thus is at odds with the perspective this study has chosen to adopt.

Interpretative phenomenological analysis (IPA) is an approach well suited to research within health psychology, and has its roots in the traditions of phenomenology and interpretivism (Brocki & Wearden, 2006; Smith 2004). IPA emphasises the personal account of the individual or group, and the researcher's role in accessing this (Smith, Jarman & Osborn, 1999). In analysis, IPA has been at the forefront of acknowledging the 'double hermeneutic', in that the researcher seeks to interpret the participant's interpretation of an event, topic or experience (Rodham, Fox, & Doran, 2013), and as such is relativist in its origins. Whilst IPA would have been a possible approach for our study, it is

arguably more suited to deep analysis and interpretation of much smaller samples (Smith, 2004), and would not have allowed us the breadth of experience we sought to obtain, or flexibility in style of analysis.

TA is recommended for analyses that may wish to report either an overall view of the data set, with emphasis on the important themes, providing both description and some interpretation (a more semantic analysis), or may instead focus upon one or two specific themes in more depth and engage with a more latent analysis. In the second, the researcher describes, interprets and theorizes beyond the data set; examining concepts, assumptions or social structures and practices that may effectively give rise to the data itself (Braun & Clarke, 2006).

6.1 Procedure of analysis:-

A traditional manual coding framework was developed for our analysis of the data, which involved a series of coding practices using printed verbatim transcripts. We broadly followed the approach described by Braun and Clarke (2013), but also integrated our own ways of working, as they suggest.

Interviews were analysed in an idiographic way, with each transcript closely read in the order in which they had occurred (as shown in Table 1). This arguably assists with the identification and development of thematic constructs, both within and across the data set, as questions themselves within the interviews developed from each subsequent interview. The audio-version of each interview was also

listened to as part of this process. Initial notes, thoughts, questions about meaning, concepts or links to the literature were made on each hard copy interview, using a 'complete' approach to coding, where all data that were of interest to the research question were read. We found analysing one interview at a time, and then having a period of 'thought time' allowed for deeper engagement with each interview, rather than rushing to the next. On returning to each interview, this allowed a thoughtful re-reading of the text, and, as each interview was re-read, tentative links or patterns could be formed across the data set.

Following the initial readings and note making, using the word-processed transcript in tandem with the hard copy annotated version for reference, all data that were relevant were extracted, copied, and pasted into a new word-processed file with an identifier and locator, so that the extract could be located back to the text and participant. Alongside each data extract, a summary or 'note' of initial thoughts and potential codes was made for each interview (for example, see Table 3).

Once the interviews were coded, the data were collated, so that each instance of a similar code was collated together with other similar codes across the whole data set. This then enabled codes to be seen as distinct, or clusters that suggested facets of a central concept or 'theme'. We preferred to do this by initially returning to hard copy, printing each item and code out separately, so that they could then be viewed as an overall 'big' picture.

Table 4. Example of early coding

Data Extract	Noticings	Code or Tentative Theme
405 INT: Do you get upset sometimes, does it upset you? **BT:** No. **MT:** It did to start with, didn't it, because **you didn't really know what was going on,** did **we**, because? **BT:** At first you were playing with me and then you stopped playing with me.	- not upset now - child upset through not understanding early symptoms, (father uses "we" - local term - but he did not understand either). Confusion for both. - not understanding early symptoms – impact noticed when play stops	**Children don't really understand** what's going on Noticing impact - **trying to make sense** of parent's behaviour

The second interviews that were based on the visual data the children produced (acting as visual prompts) were analysed in the same way. We did not attempt to carry out a thematic analysis of the drawings and posters themselves, as we were not looking for interpreted meanings or projected emotional states (Merriman & Guerin, 2006). Once an initial code or cluster of codes had been identified, the data items forming these were re-examined to see whether there was a central organising concept (theme), or whether there were facets of that theme explaining different elements of it. When these were determined they were detailed into a separate word processed document in a hierarchical way, to show theme and subtheme. We listed short explanatory lines which captured the essence of the data that contributed to each theme and subtheme for final analysis and writing.

Figure 2 below shows the final overarching theme, themes and subthemes.

During the analysis, when initial coding had been completed, the data were examined by a clinical nurse specialist in rheumatology, familiar with qualitative research but independent of the project. It was useful to have the data read by someone who might have a different perspective. Overall, there were no major areas of difference in interpretation.

Results

3.0 Overarching theme:- *Walking the line*

The overarching theme suggested by the analysis which links all subsequent themes and subthemes is the concept we have called *Walking the Line*. This concept is intended to reflect the *overall* difficulties encountered by both the parents and children in maintaining some kind of balance, or middle ground; a place which was often indistinct and open to challenge. This balance was evident in attempts to define, present and maintain a 'normal' family life; and reflected in the way in which information was communicated to children, or not, and why. This meant that there were inconsistencies within how parents behaved, and in how they communicated with their children about their rheumatic condition. Children's understanding and behaviour relating to the parents' rheumatic condition was therefore similarly inconsistent. Both parents and children felt it was important for children to have access to information and support yet struggled somewhat with defining

the content of that information. Parent's felt that it should be enough, yet not too much. Parents also felt that they needed help themselves in order to achieve this 'balancing line', as current patient education materials for adults did not prepare them sufficiently for this task.

Organising Theme 1: Next to normal*

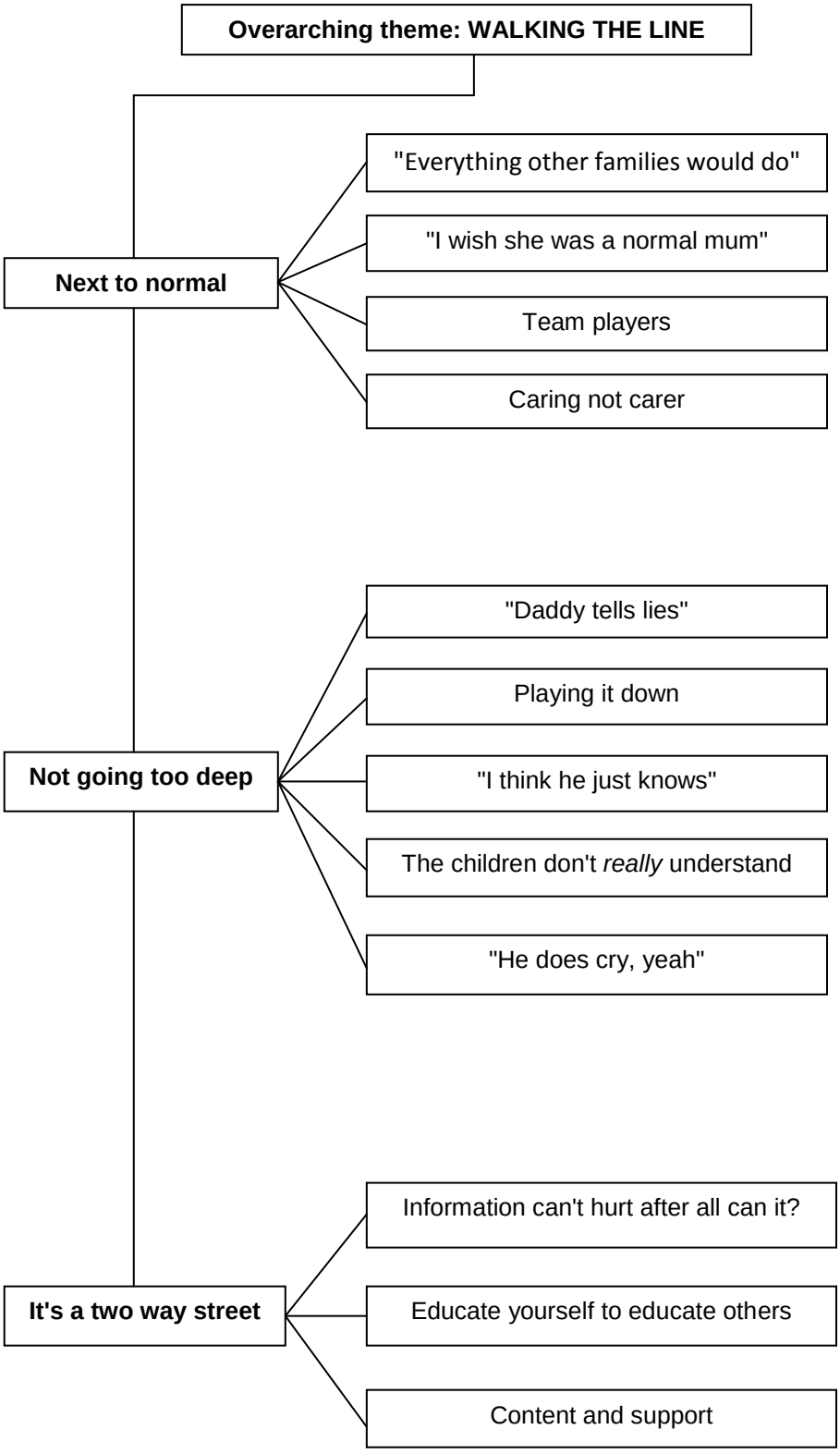
This organising theme encompasses the four subthemes *"Everything other families would do"; "I wish she was a normal Mum"; Team players* and *Caring not carer. Next to normal* illustrates how families would like to attain the idea of 'normal' family life, whilst actively having to cope with challenges to daily living outside the experiences of many 'normal' families. This means there is a careful framing and re-framing of the events of 'normal' family life, to create something that is, for them, next to normal.

1.1 "Everything other families would do"

Parents were keen to emphasize that they tried not to allow their rheumatic condition to impact upon family life wherever possible. Often this involved playing down or ignoring their symptoms for the greater benefit of the family.

> *Joy*: I don't make too much of a big deal about it (...) but I do try and, we do try and do everything that other families would do *(464)*

Figure 2. Themes and subthemes



> *Grace:* Well, I've tried to carry on because it's almost like I don't want it to..I don't want to be that person that it's affecting, and I don't want it to be affecting what I do with them...so I'm trying to stay as, you know, which I am active, it's just that obviously some things, you know, make you more tired, or make things hurt a little bit more, but I try and do as much as I can as normal, to be as normal as possible*(459)*

There were also fears that acknowledging and talking about the condition might in some way allow it to take over, and so where the disease had been a more recent onset, ignoring it maintained the old family 'normality', and allowed them to continue to be like others.

> *Clare:* I don't know if I communicated as much as I should because I was all about…I don't want the attention, I just want to try and get on as normally as I can, and I always had it in the forefront of my mind that if I lay hold of this thing and claim it and keep saying, 'my rheumatoid arthritis', this, this thing is going to zap the life out of me. So I thought, you know, I've got to sort of have a distance with it *(380)*

Where the rheumatic condition had existed prior to the birth of the children, there was also the re-framing of 'normal family' as 'normal for us', acknowledging that this 'normal' might sometimes be different to other families.

> *Karen:* I don't know if it's because I've always had it when I've had him so I suppose I've, he's never known any different *(214)*
>
> *Interviewer:* Do you think that's different to other families?
> *Theo, aged 11*: Erm, in some ways yes and in some ways no*(333*)

There were also attempts to point out that some chores carried out by children were tasks that would be expected within every other family setting, and therefore should be considered 'normal'.

> *Gail*: I like them to bring the plates to me now, but that's just, you know, I think they ought to be doing that anyway *(357)*

1.2 "I wish she was a normal mum"

For some of our participants with pre-existing rheumatic disease prior to parenthood, even the decision to become parents required particularly careful decision making and commitment. Many medications are withdrawn for a significant period pre-pregnancy/conception, resulting in a 'flare' of disease activity, which often returns for women in the post-pregnancy period.

> *Douglas:* you had to be aware of it, because some of the medication I was taking at the time wasn't conducive to becoming a parent so that had to be adjusted..*(105)*

> *Gail:* I did think about it yeah, because I thought(...) I've got this and, you know, children are going to be...it's going to be harder for me, but I've got a great GP and he said, "no, you go for it, because" he said, "I know your family and I know you've got a good network of support", (...)and then when we found out it was twins, I mean it's nice in theory but in practice, it's even harder work,*(64)*

Other parents found the difficulties encountered during the pre and post-pregnancy period too difficult to risk again, whilst others realised that adaptation was the key to coping with family life.

Karen:..now with like the medications and how it affected me having the children, that's why I decided to get sterilised*(66)*

Joy: I was struggling to do the normal parent things..I was struggling with carrying him..just changing a nappy was really hard (...) we had to find different ways of doing things...it was about learning different ways rather than trying to keep going with the old way *(36)*

For some parents, adaptation was difficult to come to terms with, particularly when it involved the loss of valued parenting activities. Parents expressed feelings of guilt - being a "waste of space" *(Clare, 687);* particularly if they felt there might be questions or judgements by other families about why they might not be joining in. Spouses felt that it was "a bit naff" *(Alastair, 569)* when family activities had to be curtailed to accommodate the impact of the condition.

Alastair:..we can't go out and do stuff that others can do as much (...)it's easier just to do stuff that works around the wheelchair*(569)*

James: ..when the snow was out last year, she [wife] was over the park with him sledging and <laughs> I want to go

Interviewer: So do you go as a bystander, or do you not go?

James: No, I don't go because I get, I can't do anything

Interviewer: That's hard?

James: Yeah. You know, when all the families are over there as well*..(400)*

Children found the fluctuating, unbalanced nature of rheumatic disease particularly difficult to understand, due to the impact this had on both the parent and then on the rest of the family. Some, like Elise, aged 9, were keen to emphasize they weren't being "mean", but would prefer it if their parent could do the things they wanted to, when they wanted to. They realised that other families did not have to think about these constraints.

> *Elise, aged 9*:..sometimes she can't do all the same things as a proper, erm, mums can, *(170)* ....sometimes I'm okay with it, but, there are some times when I wish she didn't have arthritis and she was a normal, erm, mum, but not like being mean, like just because she can't do something,*(264)*...you're not the same as others, and, but, erm, you can sometimes do the same things, or you sometimes can't do anything the same *(784)*

1.3 Team players

Part of achieving the new 'normal' for each family, involved the process of restructuring their family balance. This involved changing the roles previously undertaken by each member of the family team. Some of the adults reflected upon this as difficult, and unfair for those who had to take on extra work they had not expected.

> *Karen*: Well, when I had Oscar, my husband he more or less didn't work for a full year after I had him because I just… I really did struggle (...)and my mum's 78 but luckily she's really well, and I feel I take over her life enough as it is, and it's not fair really *(134)*

Partners found these changes difficult when the rheumatic condition had onset after the birth of children, both in accepting what the change of role might mean, and in maintaining this change in the long term

> *Charles:* I found it very hard to start with coming into it from like a husband turning into a carer, although I still don't class myself as a carer, I'm still her husband*(1307)*

> *Kate:* Trying < laughs > Yeah, it is, it is very trying (...) I feel like I, I perhaps have to do a little bit more really. Probably a little bit envious of some other couples that we're friends with, you know, where their dads are very hands on and can do lots of swimming and other stuff with them, you know, sort of thing*(35)*

Children responded to this role change in a variety of ways and could be unhappy with the change to their old routine. William, aged 7, was excited when his father collected him from school, but expressed that "it would be better if it was you" [mother], *(229)*. In this recasting of roles, there was a danger that the healthy parent became viewed by the child as the less favoured one, as they had to impose practical rules and boundaries

> *Kate:*..but mornings aren't good for James, because that's when he's in more pain. So it's trying to get Luke to understand these things. He does get kind of upset sometimes, and it's me that's the ogre because it's me that's making him do these things and taking him to places, to school, when really he'd love daddy to do it really*(35)*

For other children, the change in parental roles had proved to be a positive experience. At the time of the interviews, Helen Thomas had

increased her work hours in order to compensate for her husband being unable to work; indeed, she had been unable to attend the interviews as a result. For Belle Thomas, aged 7, this had produced a positive compromise

> *Belle:* Well, I want him to go to work but it's good because I get to play with him *(320)*

> *Matt:* I was always at work (...) we didn't do a lot, did we, now you get me drawing and painting and everything, eh< laughs>*(330)*

1.4 Caring not carer

All of the families in the study talked about negotiating a path through the 'normal' caring for each other that you might expect to see within family life, and the point at which this might stray into the role of a 'carer'. Realizing that the family might need extra help often arrived at the point of crisis

> *Joy*: she did have to do things that other children wouldn't have had to do (...) she had to phone her dad to tell him to come home (...) 'daddy you need to come home, mummy's poorly', and that was when I knew I had to get social services involved*(675)*

Joy had also accessed a young carer's support group for her daughter, recently stopped when funding cuts reduced the meeting frequency to monthly. This coincided with 'Brownies' which met on the same day. Brownie's was viewed as a 'normal' activity for young girls, and more important to keep in lieu of the carer support group.

All of the children reported helping parents by fetching and carrying things at home, and often quickly helped without being asked. Older children were asked to 'keep an eye' on younger siblings, or assist them to bed when a parent was unwell. Some children were very actively involved in helping to administer medications at home; particularly those involving injections

> *Belle, aged 7*: Erm, well, I wipe it [leg] before and then once he's done his injection, I wipe it *(33)*

> *Matt:* She sits there and says, "is it in yet?" and then you count to 10 for me before I pull it out, don't you? *(618)*

Parents did acknowledge however, that this 'fun' was not the norm for most families

> *Joy*: They fight over who's going to help <laughs>, and then they know it has to go in the sharps box (...) so having things like that around the house it's, it's already different to other kids anyway... It is normal for us yeah *(951)*

Parents were very concerned however, that their children should not become carers, and worried about asking them to help too much. Care was taken to try and strike a balance so that help was in some way 'caring' but not 'carer'

> *Gail*: I mean sometimes I feel bad if I'm, if I'm having a bad day and I ask them to do something and I've thought, because you know they say like there's more and more children becoming carers of the parents, but then I think, well, it's only very occasionally you know*(660)*

When this balance was challenged however, parents were also very proud of how their children helped with personal tasks that children usually do not have to do

> *Clare*: feeling very proud that my daughter had to put my knickers on that day, because I was so, in so much agony I couldn't bend down to pull my knickers up (...) I was so proud, I thought, thank God, look at this young lady*(969)*

Organising theme 2: Not going too deep

This organising theme concerns the attempts that parents made to provide the 'right' amount of information to their children about their condition, and their concerns about "going too deep". Five subthemes illustrate the difficulties parents faced in trying to attain this careful balance: *"Daddy tells lies"; Playing it down; "I think he just knows"; The children don't* really *understand; "He does cry, yeah".*

2.1"Daddy tells lies"

Parents found it difficult to define what the 'right' amount of information was (content), and when information should be given. Parents' main concern was to protect their child from unnecessary worry, by restricting information. This was evident both from the point of diagnosis, and as the individual and their family adjusted to the long term nature and management of their condition

*The title *Next to normal* is borrowed from a rock musical which tells the story of a mother with bipolar disorder and her struggles with this and the effects it has on her family (Kitt & Yorkey, 2008).

Beatrice: I think it's a fine balance, because you can prepare children, or prepare anybody, but in doing that, you've got to be careful not to put fear into them (...) but if they're struggling to walk and it's looking like they might start to need the wheel, to need to use a wheelchair, that might be a good time*(922-978)*

There were concerns that information, given too early, might create worry in itself, and should only be available if things deteriorated for the parent

James: enjoy his childhood, you know (...) I wouldn't want my child to worry about it until it was becoming a problem really *(597/656)*.

Children however, were often well aware that parents were not only withholding information about their health from them, but also from extended family members. They also understood the parent's motivation for doing so

Interviewer: How do you know if he's not feeling so good, how can you tell?

Belle: Because he always lies.

Int: He lies, what does he say?

Belle: He tells, he tells mum that he doesn't feel well and then when granddad comes round, he's says that he's fine.

Int: So he doesn't quite tell the truth, why do you think he does that?

Belle: Because he doesn't want people to know because they get worried.

Int: Ah, so dad tries to look after other people; do you think he does that with you as well?

Belle: Yeah*(361)*

Belle's father, Matt, was visibly distressed during the above exchange, as he had not realised the depth of his daughter's understanding. However, he also illustrated the inconsistencies present in how parents sought to protect children from worry by restricting the depth of information. Belle was very active in helping her father carry out his injections at home, and yet:

Matt: when you get round to the medications and that, I don't think they really need to know about the medications, apart from they're there to help*(987)*

2.2 Playing it down

Parents tried to play down the symptoms of their condition, and conceal anything they felt their child might find unpleasant "that would make her worry thinking, that looks a bit nasty" *(Matt, 848)*. There was an active attempt to minimise information about the impact of the condition within the immediate, wider family and social sphere. Whilst children might know the name of the parent's condition, it was often little discussed beyond that

Grace: We just call it arth, arthritis..but we don't really talk about it as,as a thing..as an illness, you know, a condition, we don't talk about it*(75)*

Partners also followed this example, deflecting or minimising questions that a child might ask

> *Kate*: Probably minimise. I probably make light of it *(198)*

Parents also struggled with using aids such as wheelchairs, which made their condition more openly visible. In using these, however, it seemed to allow children to have more open dialogue with those outside the family; sometimes revealing the parent's condition for the first time

> *Joy:* I don't go in my wheelchair unless I really have to, and I don't like people who wouldn't normally see me in it seeing me in it, because it's like admitting there's something there, erm, and for the first time she told her teacher, 'my mum's disabled.' (...) erm, and I don't know, how did they react to that? <asks child>
>
> *Elise, aged 9*: (...) and everyone just went, 'oh my god, your mum's disabled, you should have told us earlier.'*(764)*

Lack of communication, hiding, or playing down symptoms proved to be a double edged sword however, as partners and children were often surprised when difficulties became apparent

> *Clare*: sometimes it doesn't pay always to be so strong, because then when I was really hurting then, it's like, 'what's the fuss about mum?' do you know what I mean, but it's because you've masked it so long *(780)*
>
> *Crwys*: I think it's just you, erm, you don't, you just grin and bear it don't you so I don't (...)but unless you put a post-it note on your head saying, 'I'm having a really bad day'..*(313)*

It appeared that children understood that parents were minimising both the symptoms and impact of the parent's condition, even if they didn't always understand why. This resulted in children adopting the same strategy, by not saying too much about the parent's condition, minimising their own worry, or attempting to reassure the parent that they did not mind about activities being compromised

> *James:* I can't run alongside him and things like that. ..you know, and I get so wound up about things like that (...) because he'll say to me, 'I don't want to ride a bike anyway', and I'll say, 'well, no, you should', 'no, I don't want to, I'm not bothered', but I think he might be, do you know what I mean *(372/384)*

> *Tony, aged 9*: I just think, say to myself that nothing's wrong and everything's alright *(400)*

2.3 "I think he just knows"

It appeared children absorbed information almost by osmosis, from watching, experiencing and listening, rather than from the direct telling of it. Children used various means of gauging a parent's health. Luke, aged 9, noticed "lumps popped out of his elbow" (98); Theo, aged 11 "he's normally limping a lot more than normal" (556); Belle, aged 7 "Because you've got a funny face when you feel bad" (555); Tony, aged 9 "because mummy usually cries and stuff when it's painful" (185); William, aged 7 "I can tell because you'll be (...) shivering a bit and stuff. I can tell (...) by your hands normally" (615); Neil, aged 9 "sometimes I hear you talking on the phone about it" (94). Elise represented what she knew about her mother's rheumatoid arthritis in one of her drawings,

saying "when she gets really ill, she can't, she can't walk or get out of bed...and she has to use crutches" (57).

Parents equally were aware that their children knew they were unwell, not because they had been directly told, but saying the child could 'sense' it, or that they 'just knew'

> *James*: He doesn't really ask, I think he just knows *(316)*
>
> *Teresa*: I think he can sense that. He can, he can sense how, what sort of mood I'm in, if I'm in a, if I'm in a good day or, he can sense it*(458)*
>
> *Karen*: and he picks up when I'm not well and he'll come..and perhaps sit with me and give me a hug *(1319)*

In the absence of much direct information, the children had put together their own illness representations and understandings as much as they were able to, although these were sometimes hard to verbalise

> *Elise, aged 9*: She's sometimes better, erm, I don't know why, she just sometimes is (...) quite less pain than sometimes, especially, when she's ill she's sometimes in a lot of pain, so, erm, when she's not ill she's always a bit better and sometimes, even more better than that.. so she's kind of like better sometimes*(188/199)*

The children were aware that their parent came to the hospital regularly, but were unsure why or what happened when their parent was there. They did suggest that whatever happened was a positive thing however

Sara, aged 7: the doctors do something to, erm, her, and makes her a bit better*(916)*

Luke, aged 9: once he's been here, [hospital] he like feels better sometimes *(164)*

2.4 *The children don't* really *understand*

In the initial stages of diagnosis both parent and child found it difficult to understand what was happening, and again, children noticed in their own way that there were changes, yet didn't know why

Matt: ..you didn't really know what was going on, did **we**, *(author emphasis)* because..

Belle, aged 7: At first you were playing with me and then you stopped playing with me *(405)*

In the longer term, as parents restricted information and minimized their condition (in order to protect their children from worry), children couldn't always tell whether some things were part of the parents' rheumatic disease or not

Belle, aged 7: I don't know if his wrinkles on his head is part of arthritis

Matt: No, no, that's just old *(837)*

Understanding symptoms were difficult too, and whether they should be something to worry about or not. Luke, aged 9, said he found asking his father about it difficult because of this

Well, it kind of, well, it kind of stops me because sometimes when I'm like speak about stuff like that it's… sometimes I just can't understand

> like his hands hurting and stuff and everything. He says it all the time and sometimes I just think he'll be fine, but he speaks about it all the time now*(278)*

And Theo, aged 11:

> the cellulitis I thought it might have been because of the arthritis, and then eventually you did, er, say that when you'd got back, that it was because of a bug, erm, but yeah I've thought of it thinking that he might be very, er, bad, er, and there's other times when I've thought it might not be and it might be going away but…*(1142)*

Children were unsure about why their parent might have arthritis, and interpreted this question in their own frame of reference "I'm not sure; it might be a virus, a bug. I'm not that smart" *(Neil, aged 9; 1063).* Others worried that their parent's might be complicit in cause and maintenance of the condition, which was 'naughty'

> *Luke, aged 9:* You know what I think that caused his like the arthritis and stuff? my dad smokes he does..Yeah and it upsets me a bit..well, dad says he stopped but he's never stops..yes because when mum brings, gets him a packet every three weeks or something..I think it's a bit naughty *(221/228)*

Even where attempts at giving information had been made, this did not necessarily equate to 'understanding'. Lack of understanding meant children had difficulties adjusting to living with the impact of a rheumatic condition. For one single parent, Teresa, the impact of a 'bad day' meant that she would send her son aged 11 to her elder son who

lived nearby. She felt she had to do this as her young son did not understand the limitations of her condition, and reacted badly to this.

> *Teresa:* ..because at the end of the day where my son is, I've tried to be as normal as I can. If I have a bad day I send him to, like he's, like I said, to his brother's *(159)*

Teresa had chosen not to allow her 11 year old son to take part in the interview, as she was very fearful of what he might say about her. He did not understand the fluctuations in Teresa's RA and was distressed when she could go to his sister's parent evening at school, but not his. Teresa felt that his lack of understanding explained his difficult behaviour

> *Teresa:* ..it has got impact on him now because he doesn't, he takes the mickey out of me.
>
> *Int:* Right, what kind of things does he say?
>
> *Teresa:*< Sighs > they're not very nice things that I don't really want to say but he…He's like he, he, he can be nasty with it. ..I mean when I turn round, I mean if he knocks my arm, I mean I do cry when, if I'm, if I'm like in the stage where it's all flared up (...) And he'd be like, if I try and correct him another time, he goes, 'oh my arm', and, and he's like, he takes the mickey out of me*(140)*

Teresa found it very difficult to talk to her son about her condition, and felt that some resources to support her would be very useful. She did not know about young carer support groups, and asked for information at the end of the interview.

2.5 "He does cry, yeah"

Not being sure about parental health was very difficult for some of the children, and resulted in the opposite of what the parents had been trying to achieve. Going to school and worrying about the parent was not uncommon, yet the children did not often speak to their teacher or friends about the real reason for their concerns.

> *William, aged 7*: One day. I was in year 1, she was like really bad and everything I, I pretended to a teacher, 'I feel sick.' I pretended it just because I wanted to go home to like see you, yeah.... I mean sometimes I'll be like… in Year 3 it's not like, it's not like anyone you can just trick 'em if you're feeling sick*...(632)*

William said that now he was in Year 3, he tried to reassure himself that he would see his mother at the end of the day and it would be 'ok' until then. Others like Tony, (aged 9) found that he did not stop worrying at school, but that it at least "takes my mind off, erm, her in pain and stuff" *(438).*

Children could also become very upset if they inadvertently caused the parent pain, particularly if they were unsure why. Even in such situations, parents might brush off the event and minimise the problem

> *James:* he'll just move my hand and it's really painful, you know, and he's done nothing to me but I've jumped, do you know what I mean because it's really painful, and then he'll say, you know, he'll get really upset then because he's hurt me and he doesn't even know, or he'll say

to me, ‘what’s wrong, what I have done?’, you know (...) He does cry yeah, he does cry yeah. I just say, ‘well, don’t worry, it’s fine *(233-9)*

Parents could be contradictory when talking about their children coping with the impact of their condition, claiming that the child both worried and did not worry. There seemed to be little realization that they contradicted themselves, or that their 'protective' stance did not necessarily work

Alastair: You don’t like leaving mummy if she’s bad, do you, you don’t like going to school or you don’t like…

Beatrice: That’s when you get upset, isn’t it? If I’m having a really bad day, he would rather stop at home and help me to look after me, so that them are some days that you struggle, aren’t they?*(384)*

and later

Beatrice: So you don’t really worry, do you, you’re not a worrier. So you get sad, don’t you, but you don’t worry that anything’s going to go wrong, do you? [child indicates 'no'] (...) and sometimes you get a bit, a bit teary (...) you’re a bit sensitive, aren’t you?*(442)*

Organising theme 3: It's a two way street

Again illustrating attempts to hold balanced views, both parents and children felt that information and support should be provided for children, *and* for parents to help them talk to their children. Parents felt that they needed to be educated themselves in how to approach talking to their children, so that they could have effective dialogue with their

children and support them. Parents such as James Phillips felt that this would enable a multi-directional flow of information and support "it would be good to see what the children think about how...they could help the parents, what could they do...or what could the parents do for them, you know *(526)*. Here we see the subthemes *Information can't hurt after all can it?; Educate yourself to educate others* and finally, ideas around *Content and support*.

3.1 Information can't hurt after all can it?

Parents (and their children) all came to the interviews with the expressed view that providing information for children would be a good idea. For one family, the study information sheet and a local media article they had seen about the study, made them wonder if they were achieving the correct balance of information and involvement for their children

> *Grace*:..because until I'd read your article, it never even occurred to me that, you know, I should discuss with them what it, what it was or what it was about *(558)*

The high level of engagement with the research shown by the children clearly influenced the parent's views. Those children who produced a drawing or poster, and returned for another interview, had enabled their parents to have discussions not previously considered. All of the families said that the children had chosen the topic of the drawings or posters themselves, had drawn them, or asked for information from the parent to enable their drawing. Older children like

Theo and Harry, both aged 11, had sought assistance from a parent to search the internet for information to help them. Both admitted this had been the first time they had done so. Parents came to realise that information was not necessarily harmful if presented in the correct way

> *Grace:* I would think it would benefit families. It can't hurt, it can't not benefit can, it can't have a negative effect *(288)*

and Clare:

> If they can understand a lot of things that you don't want them to understand or know about, they full well can understand good, sound information. They can see it, they understand it and they can apply it to say like myself, and then they will know and I think that's excellent *(904)*

The children themselves were clear in their view that parents should be open, and suggested that they had their own ways of finding out information that was being withheld. They reasoned that information, provided early, helped to equip them for the future *even* if it might cause initial worry

> *Elise, aged 9*:.. there's some families I know, erm, they don't like tell, erm, the kids much about, erm, what they do, and the kids end up sneaking downstairs like at night or something trying to listen(...)they don't want them to hear it but they end up hearing it anyway.
>
> *Int:* So you think it's better if your mum and dad are open and tell you anyway?
>
> *Elise*: Yeah *(478)*

And this exchange between Beatrice and Tony, aged 9

> *Beatrice:*..you think it might scare them if they know how bad it can be?
>
> *Tony*: yeah, but also if mummy tells them straight away it might, it could equip them for the future *(1030)*

As Clare says, "I think we underestimate children's ability to understand" (871).

3.2 Educate yourself to educate others

Parents felt that it was their ultimate responsibility to explain their health and its consequences to their children "I think it should be down to the parent to explain" *(Matt,1012)* rather than healthcare professionals. However, parents also felt that they needed assistance in order to be able to do this, and suggested a 'companion' resource. Parents said that they either did not know enough to answer children's questions, did not know the correct way to approach the subject, or did not know what might be the 'right' words or how to use them.

> *Beatrice*:...it's the parents that need to teach the child, it's their responsibility. So maybe it's not just about equipping the children with something to read about this disease that the parents got, but equipping the parent if they are struggling, knowing 'how to' you know *(1013)*
>
> *Karen*: I don't know how to really talk to them about it, especially like Oscar who's 4 when he asks me why I can't sit on the floor and play with him, I find it hard to try and make him… I don't know what to say to help him like understand*(1170)*

Current educational resources, aimed at adult patients, do not help parents in this task. Joy Underwood did not feel that she had received the necessary information she required at diagnosis, and admitted that in her own searches to educate herself ("if you don't educate yourself you can't expect other people to understand", *1043*) the information contained language that was often difficult. Joy suggested that the kind of language appropriate for children, would actually help parent's and other adult's to understand too "sometimes the child like language is better for adults as well" *(1013).*

Children suggested that they would like to sit with their parent when looking at any resources developed for them, so that they could go through it together

> *Theo, aged 11*: I'd probably rather do it with dad and then let him explain what it means *(716)*

> *Elise, aged 9*: I'd like my mum or my dad to go through it with me *(913)*

Parent's also favoured this approach, and suggested that resources could be examined at a pace that suited the individual family, as they would be able to go back to them when they wanted to. They liked the idea that this could be a shared activity that they did together

> *Teresa:* He would sit down, if you says, come and have a look at this book

> *Sam, (adult son):* But you know he'll get bored

Teresa: Yeah but it doesn't mean to say you're going to go all through the book with him in only one go, just to go through in stages, point the important parts out and then just progress on that you could do *(256)*

3.3 Content and support

Suggestions for the content and format of information required in resources came from direct suggestions, ideas that developed by the children and their parents during the drawing exercise, and content derived from the analysis. These were the areas that parents and children felt were important to address, and in such a way that the balance of information was accurate and interesting.

Parents remained concerned that information about medication should not be too detailed "rather than anything too kind of nitty-gritty about maybe medication or anything like that" (*Grace, 579*), and particularly the medication specifically for treating their rheumatic disease "you don't want them to be too sort of focussed on the medication" (*Karen, 1301*) . Both parents and children agreed that one message that would be important to convey was that medication stored at home should be kept safe and not within the reach of children; with the children characterising it as 'strong'.

Belle, aged 7: 'if you see it, don't touch it' *(630)*

Children felt that information should be available at diagnosis whatever the age of the child. Information, and the words used, should be simple and appropriate to the age range of the child. Sara, aged 7, suggested

> we can make like a booklet and say with easy words for like younger children to read saying like...shorter words, like my brother when he first started reading on stage 1, he had like his first words they were easy *(288).*.it could say 'This book belongs to...' and they could write in their name, and then there could be like some colouring of pictures ...and then there could be like a little sentence or two to read *(381)*

Sara felt that the terminology used by adults was difficult to understand, (she felt the word arthritis was "tricky"), and Harry, aged 11, suggested that systemic lupus erythematosus was a big word that people would not understand, so shortening it to 'Lupus' was better. Elise, aged 9, wanted children to understand what 'disabled' meant, emphasising that people with disabilities can look the same as anyone else

> I think some people just think disabled means you're in a wheelchair but it doesn't *(830)*

Equally, Beatrice Roberts felt that children should embrace 'difference'

> It's about being positive that it's okay for your parent to be in a wheelchair. It might look different but it doesn't matter about them being different *(986)*

One analogy often used by parents to explain the immune system, and the role of the immune system in rheumatic disease might not be as effective as thought. Often the activity of the immune system is represented as a battle between 'good' and 'bad', with the 'bad' soldiers representing infections trying to get into the body. In the autoimmune

rheumatic diseases, this is characterised as the 'good' soldiers getting confused about what is good and bad, and attacking the wrong thing. Unfortunately this also equates rheumatic disease with infectious conditions, which is inaccurate. Theo, aged 11, felt that children needed to know that these conditions were not infectious, and that they could not be caught. He also felt that the 'infection battle' analogy might also be frightening for younger children rather than helpful

> I mean it might in a way kind of scare some younger children...thinking that they've got like little creatures fighting inside *(1071)*

It may also be a topic that simply is not interesting to children to know about, even when they were unsure what the immune system was for

> *Belle, aged 7*: Mm, I wouldn't be excited to know about it *(915)*

Again reflecting the desire to achieve a balance, both parents and children felt that any information should emphasize the positive. Some parents felt that an explanation of aids and devices (such as shoe orthoses and wrist splints) could be useful, in order to open up conversations about why the parent might need them, and how they facilitate movement. At diagnosis, where change can be dramatic and confusing, one could acknowledge the long term nature of the condition, but give hope for relief from symptoms that have the most impact upon the family

> *Douglas*: I would say for younger children, you know, the change in a short space of time could be quite, quite scary for them yeah, you know, from going from a normal life to not having a parent there, perhaps, and

> having to go up to hospital every day and visit them. So perhaps the booklet should say, you know, ‘it might not always be like this’, and explain that medicines are getting better all the time and, generally, patients seem to find one that suits them and things like that *(798)*

and Joy

> I think it needs to be..impressed that life does go on, and that it does get better *(1058)*

Both parents and children suggested that key features of rheumatic disease should be mentioned, possibly with suggestions for how the child could help at those times. They also felt that some symptoms needed 'normalising' for the condition itself. Harry, aged 11, had written on his poster:

> "people with lupus will be very tired sometimes so letting them rest, or have a sleep, will help them, or maybe you can do some housework and maybe tidy your room"

Fatigue is a 'normal' feature of most inflammatory rheumatic conditions, and one that children may struggle with understanding the most

> *Joy*: I think the fatigue is probably what frightens children more..and, you know, ‘mum or dad might be like this but it’s nothing to be frightened of’*(1058)*

Parents wanted children to understand that requests for help because of fatigue or pain were because of their condition, and not because they were lazy or trying to "pick at them or get at them" (*Clare, 676*).

Additionally, there were suggestions that children should know that a parent might be very tired and stiff in the morning. This meant that it took them a long time to 'get going', and hence be available to the child.

The impact of parent availability was something the children suggested that information should cover. This should emphasize that parents might not be able to do things as frequently, but this did not mean 'never'

> *Elise, aged 9*: [if] your mum or your dad has got it and they said, "we can't do it as much", it doesn't mean that they can't do it, at all, it just means they can't do it, like if they did it every day, they couldn't be able to do it, every day*(346)*

Parents were concerned that children should understand that when a parent was not able to do something, it was not the child's fault

> *Douglas*: you know, that dad can be tired, be in pain, you know, but it's not the child's fault, you know, if dad doesn't want to do something it's, you know, the fact that he's not feeling well enough to*(544)*

Equally, children felt it was useful to know that when a parent was in a bad mood, feeling low, or was grumpy or 'shouty' (William, aged 7), it was not their fault, but due to the parent feeling unwell.

Children and parents suggested that information could be presented in the form of a book (leaflets were not popular), or in a web-based format available on the computer, iPad or by downloading a phone App. DVD's were also popular, and thought particularly appealing and appropriate for younger children. Books were favoured by child

participants at both ends of the age range. Children were also aware of what their classmates favoured using

> *Elise, aged 9*: Well, thinking about people in my class, it will probably, be best writing it on a phone. Because a lot of people in my class have phones and they don't usually read books much *(357)*

'Colouring in' was thought to be a good way of getting children interested in a topic and learning at the same time. This could be done via interactive pictures "that you could colour in online, like using the mouse and things" (*Joy, 900*). Alternatively, 'activity packs' such as those available in restaurants or 'colour-in sheets' which children might have encountered in other situations and be familiar with were thought useful

> *Gail*: you know, like while you're waiting at the dentist or the doctor's and there's something to colour in...I think that sort of thing is quite good as well *(317)*

Unsurprisingly, children favoured plenty of pictures and diagrams, although one child, William, was clear that pictures should not be personal "a picture, but not of mum" *(1143).* Harry, aged 11, had strong views about the colour scheme used

> Well, it could be good if it's light, light and not dark colours, because lupus you want to have happy feelings for lupus, not like really sad feelings...brighter positive colours more than dark. Yeah, dark is more boring and miserable than bright, it's like shining and it's very nice, I like bright colours...bright colours are more, are better than dark colours because they make you happy *(404/423/465)*

Clare Morris suggested that information could even be kept available by the hospital to be given to the patient and their children "like a loan system and bring it back that would be good" *(1033)* if necessary. Some children were also in favour of having a person at the hospital "who like knows really a lot about arthritis" *(971)* whom they could talk to about their parent's rheumatic disease, particularly "if someone who did have it couldn't actually answer it" [a question] *(1242)*.

Discussion

This study has explored how the diagnosis and impact of parental rheumatic disease has been understood, talked about, and managed within families who have young children aged between seven and eleven years. In this context we have explored attitudes to the provision of information and/or support for children about their parents' rheumatic disease. Suggestions for format, content, and timing of the availability of information were sought. The list of themes and subthemes can be seen in Table 4.

To our knowledge, this is the first study of its kind in the rheumatic diseases, with a focus on the information needs of young children in this way. We have also considered what the implications might be for future research and healthcare practitioners in supporting such families.

Our results suggest that one of the main ways that families managed the impact of rheumatic disease upon the family was in attempts to maintain a 'normal' family life; integrating where necessary

changes that were less 'usual' into the family narrative, so that it became usual or normal for them (Sanderson et al., 2011). This was both a fast (responding to hospitalisations) and slow (realising social services needed to be involved) dynamic process, as the situation changed. Other studies have suggested that families affected by the chronic ill-health of a parent try to continue 'as normal', and are also unsure what, if anything, to tell their children, particularly immediately after diagnosis when there may be a great deal of uncertainty about the future. Children are often shielded from information and explanations about what is happening to the parent, often because they are trying to prevent the child from worrying. This becomes a double-edged sword as the child also attempts to protect the parent by not asking questions (Beckerman & Sarracco, 2012). The role of information provision in our study indicated that shielding, minimising symptoms and other impacts, or withholding information, even with the best of intentions, made the process of adapting and normalising difficult for both parents and children. Our results indicate that families walk a difficult line between providing information and not providing too much.

Conversely, all of the families in our study felt that information should be available to the children to help them understand and cope with their parents' rheumatic condition. In other studies, children themselves have overtly expressed that information about their parent's health and medical treatment, and advice about how to behave when with their parent, would help them (Mukherjee, Sloper & Lewin, 2002). Whilst children were clear that information should be open and honest,

although positive in tone, parents still preferred less detail, particularly where medication was concerned. This is typical of the contradictory behaviour displayed by the parents who may have been allowing children to assist with injections and other treatments, and integrating them into 'normal' family life (Armsden & Lewis, 1993).

Parents also felt that a companion resource would be extremely useful, providing information about their condition in a way that *they* could also understand. Studies suggest that parents may not understand or remember information sufficiently well themselves to pass on to children, particularly soon after diagnosis (Mooney, Poland, Spalding, Scott & Watts, 2013; Veldtman et al., 2000). Our participants suggested that a 'how to' guide to use with their children would be helpful, giving ideas for explaining difficult words and concepts such as the immune system. Examples of such resources already exist in other 'self-help' books written specifically for children, and might provide useful guides in developing resources for families living with parental rheumatic conditions (McKinnon, 2013).

Crucially, for parents as well as children, these resources could help normalise feelings and reactions (Courtauld & Cobb,2009). For example, simply acknowledging that identities can be challenged by ill-health may be reassuring for parents. The resource for parents might include an overview or vignettes describing the difficulties encountered in accepting that a rheumatic condition will be long-term. Refusing to adjust and adapt in the face of a condition that will persist during the life

course, often results in individuals continuing with normal routines that eventually break down or are forced into some form of change.

In her study looking at identity dilemmas in chronically ill men, Charmaz, (1994) noticed that some refused the invitation to take part in the study as it challenged their construction of their illness as an acute episode. In our study, our participant Clare Morris recalled that her own struggle with the denial of her illness as anything other than an acute event, the struggle with the impact this had on her identity, and her struggle to continue family life as 'normal' came to an end when her youngest child became aware of her condition. At this stage she was forced to reconstruct her personal and family narrative to include her illness, and acknowledged that in doing so she had found a great source of pride in her children's abilities to adapt.

In a study looking at everyday life with RA, focus group participants described the "fight between mind and body" (Kristiansen, Primdahl, Antoft & Hørslev-Petersen, 2012, p. 32) that happened in adjusting to rheumatic disease. Children are unlikely to understand the struggle that a parent may be experiencing in making these adjustments, and how this might impact on attempts to preserve the family 'normality'. Parental behaviour may appear to be inconsistent and confusing. Our participant James Phillips, who had no obvious disability, refused to go sledging with his family, partly due to physical limitations, and partly due to worries about not fulfilling his role as father. He feared revealing his 'new' disabled identity, and possibly the stigma of

judgement "you know, when all the families are over there as well..".

A recent study by Flurey et al., (2017), exploring the coping styles of men with RA, found that they tried to hide their condition in public in order to maintain a masculine image. The authors suggest that men did not wish to incorporate their rheumatic disease into their public identity, preferring to pass as 'normal'. Their study participants also felt that their RA challenged their role within the family, particularly in active play with their children. There may be genuine concerns about revealing oneself as disabled, fearing the judgement of others. Other people with disabilities have talked about the difficulties in balancing the private and public identity. In Swain and Cameron (1999), Cameron likened his eventual decision to reveal himself as disabled to 'coming out'. He suggests that people with disabilities that are less obvious are less likely to admit to impairments as they can maintain a 'normal' identity for longer.

Struggles with individual identity inevitably impact on family identity and functioning. In clinical practice we may wish to be alert to patients who are having difficulties adjusting to their rheumatic disease and be watchful for how this may be affecting family life. This may determine how receptive patients are to educational resources aimed at educating their children about their condition. The parent companion guide, as noted above, might usefully address this at the start.

3.1 Strengths and weaknesses:-

There are often criticisms of qualitative research having small sample sizes, however, this study employed a clear rationale for

calculating sample size, which achieved the breadth and depth required (O'Reilly & Parker, 2012). Whilst all participants were recruited purposively, they inevitably self-selected into the study. All were in favour of the provision of educational materials for their children which we could not anticipate *a priori*. It is possible that the inclusion of families who had the opposite view would have provided a contrasting perspective, although the expressed view was in keeping with the results of an earlier service evaluation (Hale, Kitas, O'Reilly & Vostanis, 2016).

A study strength was the inclusion of the views of three fathers and five male children. This is useful in a field where historically the male view has been underrepresented, particularly in RA (Stack, et al., 2012). Whilst this is partly due to the preponderance of women with the disease, greater efforts are now being made to engage male participants in research (Lack, Noddings & Hewlett, 2011). This is important as research outcomes should not be assumed as transferable across gender. In the present study, parents of both genders highlighted similar challenges and needs for their families.

Another strength of the study has been a conscious process of reflexivity, often little discussed in published peer reviewed literature. This process is important in everyday life, but may be more so when engaging with clinical work and research, particularly qualitative research:

> "We each have the opportunity in all parts of our lives to bring forward, underscore, articulate, make visible the meaning and importance of other people's utterances, gestures, and actions. We can be witnesses.." *(Weingarten, 2016)*

Being reflective allows us to be aware of those aspects of our experience, knowledge and self that may otherwise unintentionally affect our work. In the present study there was a growing awareness of two issues that were linked. Firstly, the extant literature in other disease groups that suggested families require information and support for their children where a parent has a chronic illness. This knowledge potentially leads to an expectation that the present study sample will suggest the same, leading to an analysis that only looks for themes that confirm this view.

Secondly, a personal reflection of the researcher's own family experiences. The researcher's partner had a congenital heart condition that required surgery when their own children were of similar age to those in the study. It was of utmost importance that personal experiences of being a family with a parent with a chronic health condition, and views on providing children with information and support, did not influence the study analysis. Independent verification of themes by a clinical nurse specialist in rheumatology assisted in monitoring the reflexive process, ensuring the trustworthiness of the analysis. Chapter Four (Critical Appraisal) contains further discussion of study challenges, strengths and weaknesses.

3.2 Implications for practice and future research:-

The researcher has had the advantage of being a psychologist embedded within a multidisciplinary (MDT) team in rheumatology, particularly as a relatively recent survey found that this was unusual (Dures, et al., 2014). The purpose of health psychology is to inform research and practice not just for other psychologists, but for other members of the MDT. Our research with a shared patient population enables us to make recommendations that can be utilised by other members of the team - in fact, may reach the population in need more easily from members of the MDT, rather than from the psychologist (Tonkin-Crine & McSharry, 2017).

The development of educational materials for children might be delivered by any member of the MDT, and additionally by charitable organisations who may be interested stakeholders in funding their production. In rheumatology, the vast majority of nationally available patient education information comes via charitable organisations like *Arthritis Research UK,* and is supplied free of charge. This does not make the role of the practitioner psychologist less valuable, as families in greater distress may still seek the help of specialist psychological services. Rather, in the course of clinical consultation, members of the MDT will be asking about family well-being and support available.

These moments may prove opportune to introduce educational resources for the children in the family, or wider family group (such as grandchildren). A review by Seawell & Danoff-Burg (2004), on psychosocial research in SLE, concluded that healthcare workers were

well-placed to help families increase their levels of communication within the family. They suggested that this may be particularly useful when patients are reluctant to talk to significant family members about their health, which could undermine well-being. Although not specifically stated, we would suggest this should include the children within these families.

Future research development might include three strands. Firstly, the development and piloting of educational materials for children aged seven to eleven years, with companion resources for their parents. Secondly, research with young people aged twelve to eighteen years, who may have different educational resource needs and support requirements when a parent has rheumatic disease. Thirdly, any future development of resources should include research to assess healthcare professional engagement with such resources, as they often act as 'gatekeepers' to such information. Barriers, or perceived barriers, to delivery and support of such resources would need to be evaluated.

Health psychology has a unique opportunity to collaborate with MDT's to help identify and effect behaviour change (parents talking to children about their condition) that improves the overall health and well-being of the family, and also behaviour change that may be required at the level of the MDT to effect best outcomes for all. Translating research into practice should involve the assessment of potential barriers to the provision of educational resources, and might look at a variety of obstacles (Grol & Grimshaw, 2003). Those that exist at the level of the patient, (resistance to accepting the need for educational resources for

themselves and child); those at the level of individual healthcare professionals and/or the health-care team.

Given the difficulty the researcher experienced with assistance from the MDT in recruiting participants to the study, MDT perceptions of increased workload, time availability and 'best use' of it, together with training showing how to provide resources, might prove reassuring and beneficial. Individuals may also fail to distribute information if they are not convinced of the evidence for it, or the value of potential outcomes. Ziegert, (2011), suggests that healthcare professionals may be anxious about being involved in an extended role that provides information aimed specifically at the family of an adult patient. Interactive training for the MDT in small group settings, which can be collaborative and supportive may be most effective (Grol & Grimshaw, 2003).

Worries about the timing of delivery of resources may also need addressing. In a study looking at providing information, and the timing of it, to RA patients about cardiovascular risk, clear differences emerged in individual healthcare professional views, (John, Hale, Treharne, Carroll & Kitas, 2009). The present study suggests that resources should at least be available from diagnosis onwards. If this were the case, clinicians could offer these at the outset, and make gentle reminders of their availability at follow-up consultations.

Clinicians should also be aware of how they talk about 'normal' and 'normalising'. We should listen carefully to how our patients represent their lives, and the outcomes and goals they hope to achieve

from treatments. It is not our remit to tell a patient what 'normal' is, nor how they should respond to their illness, yet, we should be cognisant of any difficulties our patients have in maintaining their normality. Some concerns however, are almost universal across the rheumatic conditions. Fatigue might be given as an example of how a family normalises a new symptom and subsequent behaviour. Particularly where this behaviour can cause concern to children, as our participants noted.

The United Kingdom is proud of its multicultural diversity, and is home to families who speak a range of different languages. Additionally, within these communities some people will speak but not read their first language. The need for provision of culturally appropriate information for adults in a range of languages has been established in rheumatology, with some materials translated into languages other than English, and available in a range of formats such as audio CD (Adab et al., 2004; Kumar et al., 2011; Sanderson et al., 2012). However, as far as we are aware no research has looked at the information needs of children within these communities who have a parent with a rheumatic condition. Such information would need to understand culturally appropriate beliefs and practices, including how normalcy is represented and valued (Deatrick, Knafl & Murphy-Moore, 1999). Future research might consider such work.

References

Adab, P., Rankin, E.C.C., Witney, A.G., Miles, K.A., Bowman, S., Kitas, G.D., Situnayake, D., & Bacon, P.A. (2004).Use of a corporate needs assessment to define the information requirements of an arthritis resource centre in Birmingham: Comparison of patients' and professionals' views. *Rheumatology, 43,* 1513-1518. doi: 10.1093/rheumatology/keh371

Aiden, H., McCarthy, A (2014). Current attitudes to disabled people. SCOPE: about disability. Retrieved from:

http://www.scope.org.uk/Scope/media/Images/Publication%20Directory/Current-attitudes-towards-disabled-people.pdf

Aldridge, J., with TNS-BMRB (2016) Clay, D., Connors, C., Day, N. & Gkiza, M. *The Lives of Young Carers in England*, Department for Education: London. Retrieved from:

https://www.gov.uk/government/uploads/system/uploads/attachment_data/file/498115/DFE-499_The_lives_of_young_carers_in_England.pdf

Armsden, G.C., & Lewis, F.M. (1993). The child's adaptation to parental medical illness: Theory and clinical implications. *Patient Education & Counseling, 22,* 153-165. doi: 10.1016/0738-3991(93)90095-E

Arthritis Research UK. (2011a). *Psoriatic arthritis* [Pamphlett]. Chesterfield: Author.

Arthritis Research UK. (2011b). *Rheumatoid Arthritis.*[Pamphlett].Chesterfield: Author.

Arthritis Research UK. (2013). *Ankylosing spondylitis and related conditions.*[Pamphlett]. Chesterfield: Author.

Baker, S.E., & Edwards, R. (2012). How many qualitative interviews are enough? Expert voices and early career reflections on sampling and cases in qualitative research. Discussion paper. NCRM. (Unpublished). Retrieved from: http://eprints.ncrm.ac.uk/2273/4/how_many_interviews.pdf

Beckerman, N.L., & Sarracco, M. (2012). Listening to lupus patients and families: Fine tuning the assessment. *Social Work in Health Care, 51(7),* 597-612. doi: 10.1080/00981389.2012.683679

Braun, V., & Clarke, V. (2006). Using thematic analysis in psychology. *Qualitative Research in Psychology, 3,* 77-101. doi:10.1191/1478088706qp063oa

Braun, V., &Clarke, V. (2013). *Successful Qualitative Research: A practical guide for beginners.* London: Sage.

British Psychological Society (2009).*Code of Ethics and Conduct*. Leicester: Author.

British Psychological Society (2014).*Code of Human Research Ethics*. Leicester: Author.

Brocki, J.M., & Wearden, A.J. (2006). A critical evaluation of the use of interpretative phenomenological analysis (IPA) in psychology. *Psychology & Health,21(1),*87-108. doi:10.1080/14768320500230185

Broom, A., & Willis, E. (2007) Competing paradigms and health research. In M. Saks & J. Allsop (Eds.), *Researching health: Qualitative, quantitative and mixed methods* (pp.16-31). London: Sage.

Bury, M. (1982). Chronic illness as biographical disruption. *Sociology of Health and Illness, 4(2),* 167-182.

Bury, M. (1988). Meanings at risk: The experience of arthritis. In R. Anderson, M. Bury (Eds.), *Living with Chronic Illness: The Experience of Patients and their Families* (pp. 89-116). London: Unwin Hyman.

Charmaz, K. (1994). Identity dilemmas of chronically ill men. *The Sociological Quarterly, 35(2),* 269-288. doi:10.1111/j.1533-8525.1994.tb00410.x

Corbin, J., & Strauss, A. (2008). *Basics of Qualitative Research*. London: Sage.

Courtauld, S., & Cobb, R. (2009). *My Brother and Me.* Buckinghamshire: Ethedo Press.

Cree, V.E., Kay, H., & Tisdall, K. (2002). Research with children: Sharing the dilemmas. *Child and Family Social Work, 7,* 47-56. doi:10.1046/j.1365-2206.2002.00223.x

Deatrick, J.A., Knafl, K.A., & Murphy-Moore, C. (1999). Clarifying the concept of normalization. *Journal of Nursing Scholarship, 31(3),* 209-214. doi: 10.1111/j.1547-5069.1999.tb00482.x

Dixon-Woods, M., Young, B. & Heney, D. (1999). Partnerships with children. *British Medical Journal, 319,* 778-780. doi:10.1136/bmj.319.7212.778

Donalek, J.G. (2009) When a parent is chronically ill: Chronic fatigue syndrome. *Nursing Research, 58(5),* 332-339. doi:10.1097/NNR.0b013e3181ac156f

Dures, E., Almeida, C., Caesley, J., Peterson, A., Ambler, N., Morris, M., Pollock, J. & Hewlett, S. (2014). A survey of psychological support provision for people with inflammatory arthritis in secondary care in England*. Musculoskeletal Care*, *12(3),* 173-181. doi: 10.1002/msc.1071

Dures, E., Rumsey, N., Morris, M. & Gleeson, K. (2011). Mixed methods in health psychology: Theoretical and practical considerations of the third paradigm. *Journal of Health Psychology, 16,* 332-341*.* doi:10.1177/1359105310377537

Eiser, C.,& Twamley, S. (1999). Talking to children about health and illness. In M. Murray, K. Chamberlain (Eds.), *Qualitative health psychology: Theories and methods* (pp.133-147). London: Sage.

Flurey, C.A., Hewlett, S., Rodham, K., White, A., Noddings, R., & Kirwan, J.R. (2017). "You obviously just have to put on a brave face": A qualitative study of the experiences and coping styles of men with rheumatoid arthritis. *Arthritis Care & Research, 69(3),* 330-337. doi: 10.1002/acr.22951

Forrest, G., Plumb, C., Ziebland, S., & Stein, A. (2006). Breast cancer in the family - children's perceptions of their mother's cancer and its initial treatment: qualitative study. *British Medical Journal, 332,* 998-1003. doi:1136/bmj.38793.567801.AE

Francis, J., Johnston, M., Robertson, C., Glidewell, L., Entwistle, V., Eccles, M.P., & Grimshaw, J.M. (2010). What is an adequate sample size? Operationalising data saturation for theory-based interview studies. *Psychology & Health, 25(10),* 1229-1245. doi: 10.1080/08870440903194015

Fusch, P.I., & Ness, L.R. (2015). Are we there yet? Data saturation in qualitative research. *The Qualitative Report, 20*(9), 1408-1416. Retrieved from http://www.nova.edu/ssss/QR/QR20/9/fusch1.pdf

General Medical Council (2007). 0-18 Years: Guidance for all Doctors. Accessed July 29, 2016, at

http://www.gmc-uk.org/guidance/ethical_guidance/children_guidance_14_21_communication.asp

Gillick v West Norfolk and Wisbech. London: House of Lords, Gillick v West Norfolk and Wisbech AHA (1986) AC 112, [1985] 3 WLR 830, [1985] 3 AII ER 402, HL

Goffman, E. (1968). *Stigma: Notes on the management of spoiled identity.* Penguin: Harmondsworth.

Grol, R., & Grimshaw, J. (2003). From best evidence to best practice: Effective implementation of change in patients' care. *The Lancet, 362,* 1225-1230. doi: 10.1016/50140-6736(03)14546-1

Guest, G., Bunce, A., & Johnson, L. (2006). How many interviews are enough?: An experiment with data saturation and variability. *Field Methods, 18,* 59-82. doi: 10.1177/1525822X05279903

Hale, E.D., Kitas, G.D., O'Reilly, M., & Vostanis, P. (2016). Should there be information and support available for children whose parents or grandparents have a chronic rheumatic condition? *Rheumatology*, 55 (suppl): i148. doi:10.1093/rheumatology/kew177.001 Retrieved from:

http://rheumatology.oxfordjournals.org/search?submit=yes&tocsectionid=Education&volume=55&issue=suppl%201

Hart, R.I., McDonagh, J.E., Thompson, B., Foster, H.E., Kay, L., Myers, A., & Rapley, T. (2016). Being as normal as possible: How young people ages 16-25 years evaluate the risks and benefits of treatment for inflammatory arthritis. *Arthritis Care & Research, 68(9),* 1288-1294. doi:10.1002/acr.22832

Harvey, L., MacDonald, M., & Hill, J. (2000). *Access to Sociology: Theories and Methods.* Abingdon: Hodder & Stoughton.

Hemming, P.J. (2008). Mixing qualitative research methods in children's geographies. *Area, 40(2),* 152-162.

doi:10.1111/j.1475-4762.2008.00798.x

Hill, J,. & Ryan, S. (2000). *Rheumatology: A handbook for community nurses*. London: Whurr.

John, H., Hale, E.D., Treharne, G.J., Carroll, D., & Kitas, G.D. (2009). 'All singing from the same hymn sheet': Health professionals' perceptions of developing patient education material about the cardiovascular aspects of rheumatoid arthritis. Musculoskeletal Care, 7(4), 256-271. doi: 10.1002/msc.157

Kitt, T. (Music),& Yorkey, B. (Book & Lyrics).(2008). *Next to normal. An Original Musical.* New York, NY: Musical Theatre International.

Kristiansen, T.M., Primdahl, J., Antoft, R., & Hørslev-Petersen. (2012). Everyday life with rheumatoid arthritis and implications for patient education and clinical practice: A focus group study. *Musculoskeletal Care, 10,* 29-38. doi: 10.1002/msc.224

Kumar, K., John, H., Gordhan, C., Situnayake, D., Raza, K., & Bacon, P.A. (2011). Breaking communication barriers for RA patients of South Asian origin: The use of a bilingual educational audio CD and linguistically appropriate peer support and education. *Musculoskeletal Care, 9(1),* 11-18. doi: 10.1002/msc.191

Lack, S., Noddings, R., & Hewlett, S. (2011). Men's experiences of rheumatoid arthritis: An inductive thematic analysis. *Musculoskeletal Care, 9(2),* 102-112. doi: 10.1002/msc.203/pdf

Lyons. A. (1999). Shaping health psychology: Qualitative research, evaluation and representation. In M. Murray, K. Chamberlain (Eds.), *Qualitative health psychology: theories and methods* (pp.133-147). London: Sage.

Matthews, J. (2014). Real world research: Epistemology matters. *Qualitative Methods in Psychology (QMiP) Bulletin, 17,* 10-13. See http://qmip.bps.org.uk/

Medical Research Council (MRC). 2004. MRC Ethics Guide: Medical Research Involving Children. London: Accessed August 4, 2016, at

http://www.mrc.ac.uk/documents/pdf/medical-research-involving-children/

McKinnon, H. (2009). Winston's Wish. *The Family Has Been Informed*. Portfolio publishing.

Merriman, B., & Guerin, S. (2006). Using children's drawings as data in child-centred research. *The Irish Journal of Psychology, 27(1-2),* 48-57. doi:10.1080/03033910.2006.10446227

Modi, N., Vohra, J., Preston, J., Elliott, C., Van't Hoff, W., Coad, J....Greenough, A. (2014). Guidance on clinical research involving infants, children and young people: An update for researchers and research ethics committees. Royal College of Paediatrics and Child Health Guidance Report. Accessed July 29, 2016, at

http://www.rcpch.ac.uk/sites/default/files/page/Guidance%20on%20clinical%20research%20involving%20infants,%20children%20and%20young%20people%20v4%20FINAL.pdf

Mooney, J., Poland, F., Spalding., Scott, D.G.I., & Watts, R.A. (2013). 'In one ear and out the other - it's a lot to take in': A qualitative study exploring the informational needs of patients with ANCA-associated vasculitis. *Musculoskeletal Care, 11,* 51-59. doi: 10.1002/msc.1030

Morrow, V. (2008). Ethical dilemmas in research with children and young people about their social environments, *Children's Geographies, 6(1),* 49-61.doi:10.1080/14733280701791918

Mukherjee, S., Sloper, P., & Lewin, R. (2002). The meaning of parental illness to children: The case of inflammatory bowel disease. *Child: Care, Health & Development, 28(6),* 479-485. doi: 10.1046/j.1365-2214.2002.00302.x

NHS Health Research Authority (n.d.[a]). *Consent and participant information sheet guidance: online version.* Accessed July 27, 2016, at

http://www.hra.nhs.uk/resources/before-you-apply/consent-and-participation/consent-and-participant-information/

NHS Health Research Authority(n.d.[b]). HRA Ethics Guidance: *Payments and Incentives in Research.* Accessed July 27, 2016, at http://www.hra.nhs.uk/documents/2014/05/hra-guidance-payments-incentives-research-v1-0-final-2014-05-21.pdf

O'Reilly, M., & Parker, N. (2012). 'Unsatisfactory saturation': A critical exploration of the notion of saturated sample sizes in qualitative research. *Qualitative Research:* 1-8. doi:10.1177/1468794112446106

Parsons, S., Ingram, M., Clarke-Cornwell, A.M., & Symmons, D.P.M. (2011). A heavy burden: The occurrence and impact of musculoskeletal conditions in the United Kingdom today. The University of Manchester & Arthritis Research UK. Accessed Oct 09, 2015, at http://www.inflammation-repair.manchester.ac.uk/Musculoskeletal/aboutus/publications/heavyburden.pdf

Prosser, J., & Loxley, A. (2008). Introducing visual methods. Review paper/010. ESRC National Centre for Research Methods. (Unpublished). Retrieved from: http://eprints.ncrm.ac.uk/420/1/MethodsReviewPaperNCRM-010.pdf

Readability Calculator: Tests document readability. Accessed August 20th, 2012, at https:// www.online-utility.org

Rehm, R.S., & Franck, L.S. (2000). Long-term goals and normalization strategies of children and families affected by HIV/AIDS. *Advances in Nursing Science, 23(1),* 69-82. Accessed at: journals.lww.com/advancesinnursingscience/Pages/default.aspx

Robinson, C.A. (1993). Managing life with a chronic condition: The story of normalization. *Qualitative Health Research, 3(1),* 6-28. doi:10.1177/104973239300300102

Rodham, K., Fox, F., & Doran, N. (2013).Exploring analytical trustworthiness and the process of reaching consensus in interpretative phenomenological analysis: Lost in transcription. *International Journal of Social Research Methodology, 18(1),* 59-71. doi:10.1080/13645579.2013.852368

Romney, A.K., Weller, S.C., & Batchelder, W.H. (1986). Culture as consensus: A theory of culture and informant accuracy. *American Anthropologist, 88(2),* 313-338. doi: 10.1525/aa.1986.88.2.02a00020

Sanderson, T., Calnan, M., Morris, M., Richards, P., & Hewlett, S. (2011). Shifting normalities: Interactions of changing conceptions of a normal life and the normalisation of symptoms in rheumatoid arthritis. *Sociology of Health and Illness, 33(4),* 618-633. doi:10.1111/j.1467-9566.2010.01305.x

Sanderson, T., Hewlett, S., Calnan, M., Morris, M., Raza, K., & Kumar, K. (2012) Exploring the cultural validity of rheumatology outcomes. *British Journal of Nursing, 21,(17),* 1015-1023. Retrieved from:http://eprints.uwe.ac.uk/17472/2/Main%20document%20Br%20J%20Nursing%20FINAL%2011%209%2012.pdf

Seawell, A.H., & Danoff-Burg, S. (2004). Psychosocial research on systemic lupus erythematosus: A literature review. *Lupus, 13,* 891-899. doi: 10.1191/09612033041lu1083rr

Simons, G., Mason, A., Falahee, M., Kumar, K., Mallen, C.D., Raza, K., & Stack, R.J. (2017). Qualitative explorations of Illness perceptions of rheumatoid arthritis in the general public. *Musculoskeletal Care, 15(1),* 13-22. doi: 10.1002/msc.1135

Smith, J.A. (2004). Reflecting on the development of interpretative phenomenological analysis and its contribution to qualitative research in psychology. *Qualitative Research in Psychology, 1(1),* 39-54. doi:10.1191/1478088704qp004oa

Smith, J.A., Jarman, M., & Osborn, M. (1999). Doing interpretative phenomenological analysis. In M. Murray, K. Chamberlain (Eds.), *Qualitative health psychology: Theories and methods* (pp.133-147). London: Sage.

Stack, R. J., Shaw, K., Mallen, C., Herron-Marx, S., Horne, R., & Raza K. (2012). Delays in help seeking at the onset of the symptoms of rheumatoid arthritis: A systematic synthesis of qualitative literature. *Annals of the Rheumatic Diseases*, *71,* 493-497. doi:10.1136/ard.2011.155416

Swain, J., & Cameron, C. (1999). Unless otherwise stated: Discourses of labelling and identity in coming out. In M. Corker, S. French (Eds.), *Disability Discourse* (pp.68-78). Open University Press: Buckingham.

Tonkin-Crine, S., & McSharry, J. (2017). Increasing impact in health psychology: Working in multidisciplinary research teams. *Health Psychology Update, 26(1),* 24-29. A publication of the Division of Health Psychology of the British Psychological Society (www.bps.org.uk).

United Nations Convention on the Rights of the Child (1990). Geneva, OHCHR. Accessed October 10, 2008, at

http://www.ohchr.org/EN/ProfessionalInterest/Pages/CRC.aspx

Veldtman, G.R., Matley, S.L., Kendall, L., Quirk, J., Gibbs, J.L., Parsons, J.M., & Hewison, J. (2000). Illness understanding in children and adolescents with heart disease. *Heart, 84,* 395-397. doi: 10.1136/heart.84.4.395

Weingarten, K. (2016). The art of reflection: Turning the strange into the familiar. *Family Process, 55(2),* 195-209. doi: 10.1111/famp.12158

Yardley, L., & Bishop, F.L. (2015).Using mixed methods in health research: Benefits and challenges. *British Journal of Health Psychology, 20(1),* 1-4. doi:10.1111/bjhp.12126

Youssef, M.M., Salah, E.M., Salem, S.M.E., & Megahed, H.S. (2010). Concepts of health and illness in Egyptian children. *Medical Journal of Cairo University, 78(1),* 77-84. Retrieved from http://cu.edu.eg

Ziegert, K. (2011). Maintaining families' well-being in everyday life. *International Journal of Qualitative Studies in Health and Well-being, 6(2).* doi: 10.3402/qhw.v6i2.7206

Appendix A

Statement of Epistemological Position

Novice researchers start by learning to define a topical research question - what *exactly* is our aim or, what do we want to know or understand? Having achieved this, we are told that we should choose the appropriate method to answer our research question. It is important however, to distinguish between 'method' and 'methodology'. 'Method' relates to how we are going to collect and analyse our data; and the tools or techniques that we will employ such as questionnaires, interviews, observations, or visual or other media. 'Methodology' is our wider approach to collecting data, and involves asking why we are using *these* methods and how they relate to our understanding of the world (Corbin & Strauss, 2008; Dures, Rumsey, Morris & Gleeson, 2011; Harvey, MacDonald & Hill, 2000; Matthews, 2014).

Ideally, our methodology and subsequent choice of method should be related to how we understand the nature of reality (ontology), what we believe about the nature of knowledge and how we can obtain it (epistemology). Whilst there is always a continuum to these positions, *realists* would argue that truth is real, fixed, and discoverable by using the correct techniques, whilst *relativists* would suggest that truth, as such, is only a perspective that exists amongst others (Braun & Clarke, 2013; Broom & Willis, 2007). Researchers working within a realist ontology adopt data collection methods that seek to objectively measure the item or 'variable' of interest, or seek to discover an ultimate 'truth', and may use quantitative or *some* forms of qualitative enquiry and

analytical methods. Relativist researchers often work within interpretive or constructionist epistemologies and use qualitative methods to obtain data. Interpretivists believe that there is no one 'truth', indeed, our 'truth' is our perception alone, fluctuating through experience and time; shaped by social context and the cultures we are part of, and therefore 'relative' to that (Braun & Clarke, 2013; Yardley & Bishop, 2015). Researchers working within an interpretive methodology are hoping to facilitate access to the 'telling' of these experiences and perceptions, via appropriate qualitative method(s); and to actively acknowledge their role in the construction and, later, interpretation through analysis, of these realities. The data generated happens between the researcher and the researched, and the usefulness of the results does not depend upon finding a 'truth', but whether the explanation makes sense and is useful to other people in similar situations (Lyons, 1999). Quantitative researchers however, aim to minimize researcher involvement and influence as much as possible.

Qualitative researchers have different ways of gathering data, and the approach they adopt may in itself reflect their personal way of viewing the world and how they interpret knowledge (a paradigm). Realistically, this may also be dependent upon the demands of funders and/or time available (Corbin & Strauss, 2008). Our study wanted to understand how families with young children aged between seven and 11 years, experience and talk about parental rheumatic disease; particularly whether, and how, they spoke to their children about it. We also wanted to know whether they thought providing information to

children about parental rheumatic disease would generally be a good idea, and if so, what kind of information should be provided and if possible, in what format.

As we wanted to enable children to have a voice in our study, we needed to adopt a research methodology and methods that would be suited to engaging with families with young children aged between seven and 11 years, and would be flexible enough to be useful for all abilities. This led us to adopt a qualitative methodology, using semi-structured interviews and visual data collection to assist children in being as participatory as possible in expressing their views. Our overall paradigm was one of relativist ontology and interpretive epistemology.

Appendix B - Study Information Sheet (patient)

Funding body logo here

NHS Trust Logo here

Department of Rheumatology

Clinical Research Unit

Number:

Study reference number:

XXXX UK Grant Ref:

Date:

Children of parents with chronic (long-term) rheumatic conditions:

Their experiences, needs and resources

Information Sheet

My name is Liz Hale and I would like to give you some information about a research project that I am carrying out as part of my Doctoral qualification at the University of Leicester, and I would like to invite you and your family to take part. The research team are myself, Professor XXXX and Dr XXXX from the University, and Professor XXXX from the hospital.

The research is being funded by XXXX UK and The XXX Charitable Fund.

What is the research about?

People with long-term rheumatic conditions like rheumatoid arthritis, systemic lupus erythematosus (sometimes called SLE or Lupus), ankylosing spondylitis and so on, are often given leaflets to read about their condition. These leaflets try to answer some of the questions you and your family might ask.

However, at the moment there is no information available for the **children** of our patients who have long-term rheumatic conditions. In order to provide the correct information for these children in the best way

possible we need to carry out some research first with families where one of the parents has one of these conditions.

We would like to ask patients what it is like being a parent with a long-term rheumatic condition, and what kind of information they think would be useful for their child(ren) to know about it. We would also like to ask their spouse or partner for their views as well. Most importantly we would like to talk to their child(ren) to ask them for their views. This is because we believe that the child(ren) should have the most input into saying what information would be useful to them and how it should be delivered (and by whom if necessary).

Why are you asking me and my family to take part?

We are asking you to take part in this research project as you have a long-term rheumatic condition and have a child or children aged between 7 and 11 years old, which is the first age group we would like to provide information for.

What would my family have to do?

You and your spouse or partner would be invited to take part in an interview with Liz Hale at a time most convenient to you. This interview could take place individually, or you may prefer to be interviewed together.

The interview would last for approximately one hour. With permission the interview would be audio-recorded. This is nothing to worry about; it is just to make sure that we do not miss anything you and your spouse/partner tell us about, and this will be typed up later.

After this interview we would invite you to bring your child or children to an interview with Liz. This would take place on a different day. If you have one or more children aged between 7 and 11 years they can all take part if they wish, it is up to them.

All of the interviews would be very informal; whilst Liz will have some topics she would like to talk about, you will all be free to discuss the things that you feel are important. You do not have to worry about saying the right or wrong thing as we are just interested in your own experiences and views.

Where would the interviews take place?

We have a quiet room in the hospital that the interview would take place in. This is an office-style room with comfortable chairs and does not have any sinks or beds or medical equipment in it. There will be refreshments provided.

For the interviews with your child(ren), the door to the room would remain open during the interview and you would be asked to wait in the office opposite, also with the door open.

This is so that your child can see you and can come to see you if they wish during the interview. If your child(ren) would prefer you to be in the room during the interview, or would prefer to be interviewed with their brother or sister this is okay, too.

If you would like any of these interviews conducted at home you may be able to arrange this with Liz.

Is it just an interview?

During the interview with your child(ren) we will give them some paper and art materials. There are two reasons for this. Firstly, some children may find it hard to say in words what they would like to communicate, so we are giving them the opportunity to make a drawing to tell us how they feel or what they are thinking about.

Secondly, we would also like to ask them to take the art materials away with them (they can keep them after the project) and to make a drawing, poster or storyboard. They will be asked to imagine that another child has a parent with the same rheumatic condition that you have, and asked:

"What do you think would be helpful for them to know"?

"What would you tell another child" ?

About two weeks after the interview with your child(ren), Liz will collect the artwork and carryout a short interview with them whilst they talk about their drawing, poster or storyboard. This will be audio-recorded as before.

With your and your child's permission, we may use some of these artworks in our study and published materials.

Who gives consent for my family to take part?

If you would like to take part in our research project we would ask you to sign an informed consent form before doing the interview. We would ask your spouse or partner to do the same.

When children under the age of 16 are taking part in a research project, the parents should give written consent for them to do so. Your child(ren) will be given the chance to sign their own assent form as well if they wish.

What if I or my child agrees to take part and then changes their mind?

You and your family are free to withdraw from this study at any time. Your care, and the care of anyone in your family, will not be affected by your decision to take part or to withdraw from the study at a later stage. If you or your family withdraw from the study after completing some or all of the interviews and artwork, we will still need to use the information collected from you up to that point.

Will the information we give be kept confidential?

Nobody except the research team and interview transcriber will have access to what was said during your interviews, either from the audio-recording or in paper copy. The research team are listed at the end of this information sheet.

We will do everything we can to protect your confidentiality. When we write our reports for this study we will not use your real names or the real names of anyone else you identify, or the real names of places (such as schools).

What you have said in your interviews may be quoted directly but we will give you all pretend names to protect your confidentiality. XXXX UK may also use some of this anonymised data in the pursuit of their charitable goals.

The only time we might have to reveal someone's name is if they said they were worried that they were going to be harmed, or that someone else might be harmed. Then we would have to speak to the relevant authorities.

What if my child changes her/his mind during the interview?

We will be very careful to make sure your child is comfortable during the interview. They may choose not to answer some questions which will

beokay. We have also made some cards that the children can use during the interview to show us how they feel if they do not want to speak.

You and your child will be free to stop the interview at any time. The child may re-start the interview only if **they** indicate they wish to do so.

What are the possible disadvantages of taking part in this research?

This study has been carefully designed to make sure that any disadvantages to you and your family are very small.

We will refund you and your family any travel and parking costs you have for coming to the hospital for the interviews.

We understand that talking about personal issues can sometimes be upsetting. If you would like to talk to someone after the interviews, or you feel that your child(ren) would benefit from doing so, we can advise you about arranging this. We can also give you information about support services for your child(ren).

What are the possible advantages of taking part?

Whilst we cannot promise that taking part in this research project will help you and your family now, the information that you share with us may help the children of patients in the future.

In the past, people have reported that taking part in research studies like this was a positive experience.

We will keep in touch with you to let you know how the research is progressing in the future.

What will happen to the results of this research study?

The results from this study will be written up into reports which will be presented at conferences and seminars and will be sent for publication in peer reviewed academic journals. They will guide the development of future studies and resources.

Have you had permission to do this research?

Before this research was allowed to happen we asked an independent review panel called the National Research Ethics Service (NRES) to look at our proposal in detail. We have been given permission by the West Midlands - Staffordshire NRES Committee to carry out this research.

What if there is a problem in the future?

If you have any concerns about any aspect of the research study, or if you feel you or your family have experienced any harm from taking part in the study, you should contact a member of the research team listed at the end of this form. They will do their best to resolve your complaint. You can also contact the Patient Advice and Liaison Service (PALS) at the hospital by telephoning XXXX or XXXX or by email at XXXX

If you remain unhappy and feel that you or your family have been harmed during the research study and that this is due to someone's negligence, then you may have grounds for legal action for compensation against the XXXX NHS Foundation Trust.

What should I do next if I want to take part?

If, after reading this information sheet and discussing it with your family, you think you would like to help us, we would invite you and your family to meet Liz when you can ask any additional questions and arrange convenient times to hold the interviews. It will also be a chance for your children to meet Liz so that they feel more comfortable with her before the actual interview.

You should also have received an information sheet for your spouse or partner. If you do not have a spouse or partner, but there is someone who lives in your household who plays a significant part in your and your family's life, they can volunteer to take part in the study, too.

We have also given you a simplified information sheet to give or read to your child(ren), so that they understand what our research project is about.

What if I have any questions now?

If you would like to request any further information, or talk to a member of the research team please tell the person who gave you this information sheet, or ask the receptionist to contact Liz Hale as she may be able to talk to you now.

You can contact Liz Hale by telephone on:

XXXXextension**XXXX**(there is an answerphone to leave a message if she is not there)

Or e-mail: XXX

Or write to: Ms Elizabeth Hale,Department of Rheumatology,

XXXX NHS Foundation Trust,XXXX Hospital

Thank you for taking time to read this information sheet. I hope you will consider helping us with our research.

The Research Team:

Professor XXXX (Tel: XXXX)

Dr XXXX (Tel: XXXX)

Professor XXXX (Contact via Mr XXXX below)

Research Support Officer: Mr XXXX (Tel:XXXX)

Appendix C - Study Information Sheet (child)

Funding body logo here

NHS Trust logo here

Department of Rheumatology

Clinical Research Unit

Number:

Study reference number:

XXXX UK Grant Ref:

Date:

Study title: Children of parents with chronic (long-term) rheumatic conditions: Their experiences, needs and resources

Information Sheet

Hello my name is Liz Hale,

I have asked your Mum or Dad if your family will help me with a research project. Research is how we find out the answers to questions.

What questions do you have?

Your Mum or Dad has something called "arthritis" (sometimes it has a longer name). Your family may have your own name for it!

Could you tell me what things **you** would like to know about your Mum or Dad's arthritis?

I would also like to know **how** I should tell you these things. You may have lots of ideas – maybe in a book? Or maybe on a DVD?

I would also like to know if you would like someone to talk to about this.

Do I have to take part?

You can decide if you would like to take part. If you do not want to, you do not have to.

What will I have to do?

I would like to talk to you about what it is like being you, and what it is like for you having a Mum or Dad with arthritis.

I will also bring some paper, pens and pencils with me. This is because some people find it easier to draw what they feel or think about something.

I would like you to make a drawing or poster or story board.

I would like you to think about what **you** would tell another child who has a Mum or Dad with arthritis like your Mum or Dad has.

What would be important for them to know? What would you tell them?

After about 2 weeks I will see you again. You will be able to tell me about your drawing. If you agree, I would like to keep your work to show other people.

Where will I talk to you?

At home, or if you like, you can come to the hospital that I work at. My room is like an office and does not have any scary things in it.

There will be drinks and a snack for you if you want one.

Your Mum or Dad will bring you, and will wait while you talk to me. You will be able to see them all the time and go to them if you want to.

If you want your Mum or Dad to stay in the room with you that is ok. If you have a brother or sister and would like them to stay that is ok too.

How will you remember the things I say?

When you talk to me I will use a recorder to help me remember everything you say.

You do not have to worry about saying the wrong thing or drawing the wrong thing, I just want to know about what your life is like and what you think, and this includes good and bad things.

Will taking part upset me?

If I ask you any questions you do not want to answer, you do not have to. You do not have to do the drawing if you do not want to.

Will taking part help me?

I hope you will like talking to me about yourself and doing the drawings.

People say that taking part in a research project can feel good, and you may help a child in the future.

What if I don't want to do the research anymore?

If you change your mind and decide that you don't want to do the research anymore that is ok. Just tell me or ask Mum or Dad to tell me.

Will anyone else know I'm doing this?

Nobody will know you are helping us with our research project apart from your family.

If you tell me the names of your friends, family, school or other people I will not use their names in any of my work.

When I write about my project I will give you and your family pretend names.

The only time I might have to tell someone your real name is if you told me that someone is hurting you, or that you are worried that someone else is being hurt. To protect you, I might have to tell someone else about this.

If you are happy to help me with my research project I will ask you to put your name on another piece of paper. Your Mum or Dad can write your name for you, as long as you agree it is ok.

Thank-you for reading this letter!

Liz Hale ☺

Appendix D - Assent form

Funders Logo here

NHSTrust Logo here

Department of Rheumatology

Clinical Research Unit

Number:
Study reference number:
XXXX UK Grant Ref:
Date:
Study title: Children of parents with chronic (long-term) rheumatic conditions: Their experiences, needs and resources
Name of Researcher: Elizabeth Hale

Assent Form

Please tick the YES box if you agree with what the words say:

I understand why this research is being done ☐

I have asked any questions that I want to ☐

I understand what will happen next ☐

I am happy for my words and pictures to be used for this research ☐

I understand my name will be kept secret ☐

I understand I can stop doing this research if I want to ☐

I am happy to take part ☐

If you are happy to talk to Elizabeth please write your name below. If you do not want to talk to Elizabeth then do not write your name below.

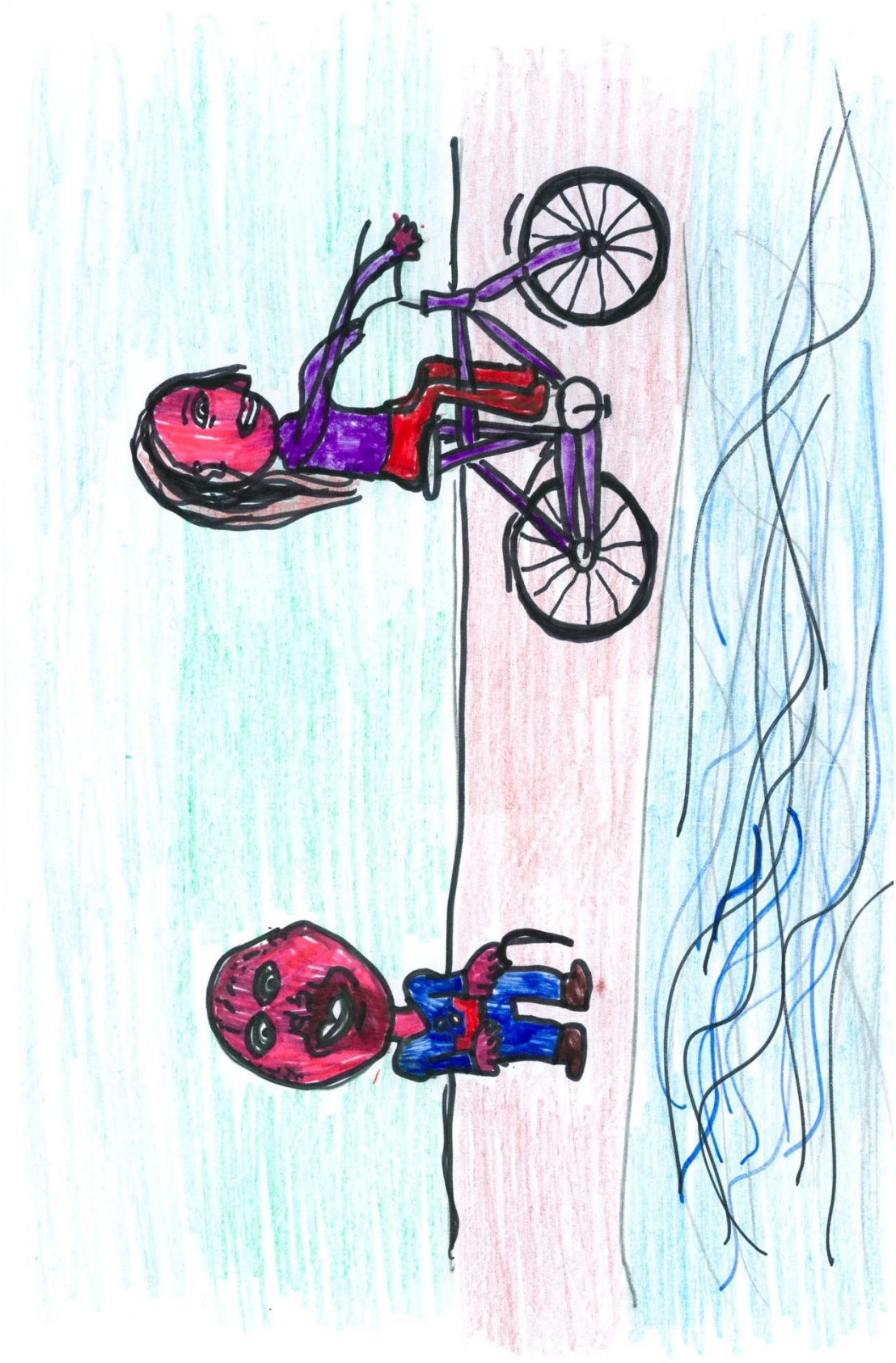
My name is:.....The date is:.....

Appendix E - Drawing by Luke Phillips, aged 9.

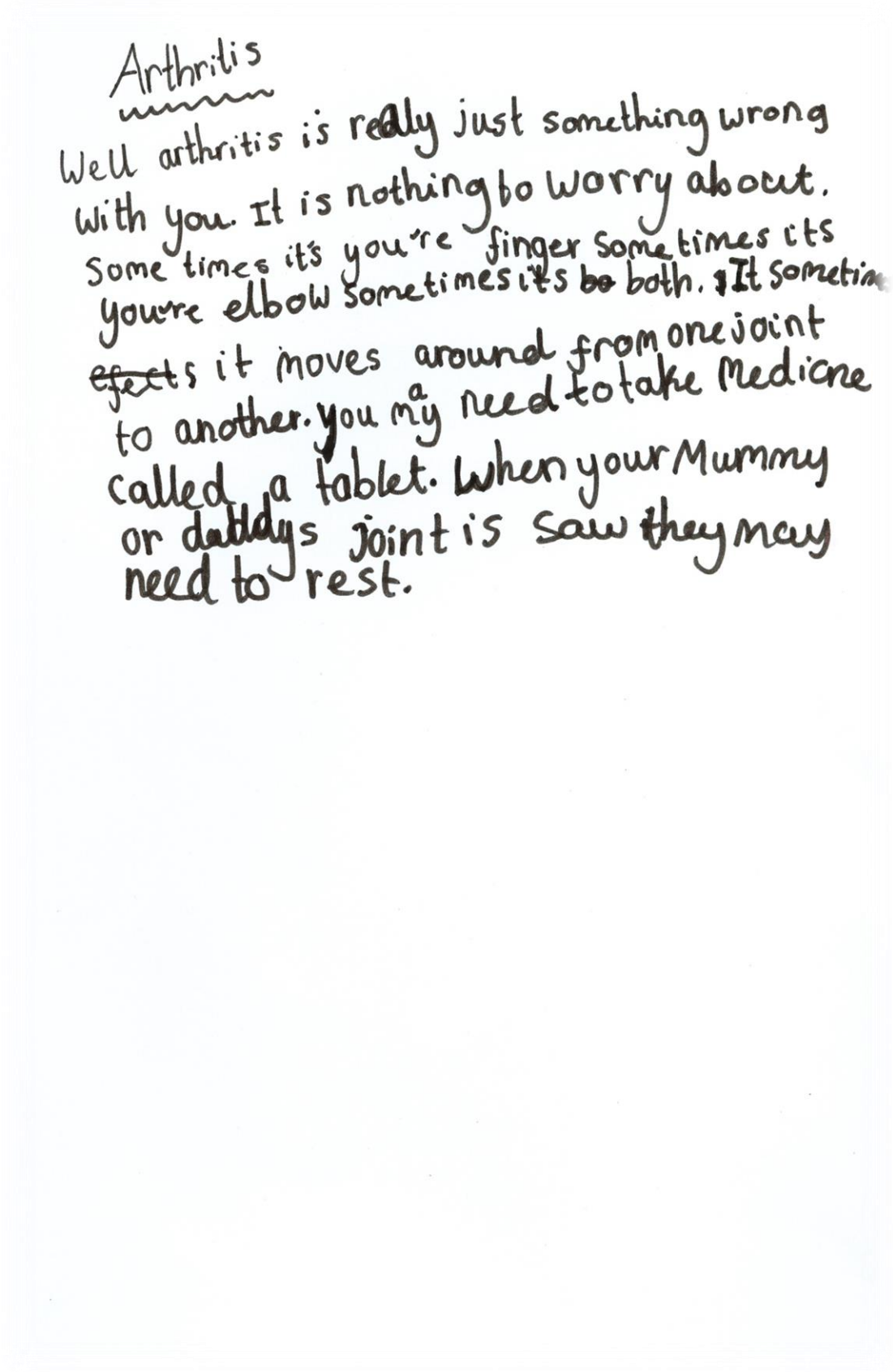
Appendix F - Drawing by Tony Roberts, aged 9.

Appendix G - Drawing 1 by Belle Thomas, aged 7.

Appendix H- Drawing 2 by Belle Thomas, aged 7.



Appendix I - Writing 1 by Sara Jackman, aged 7.



Arthritis

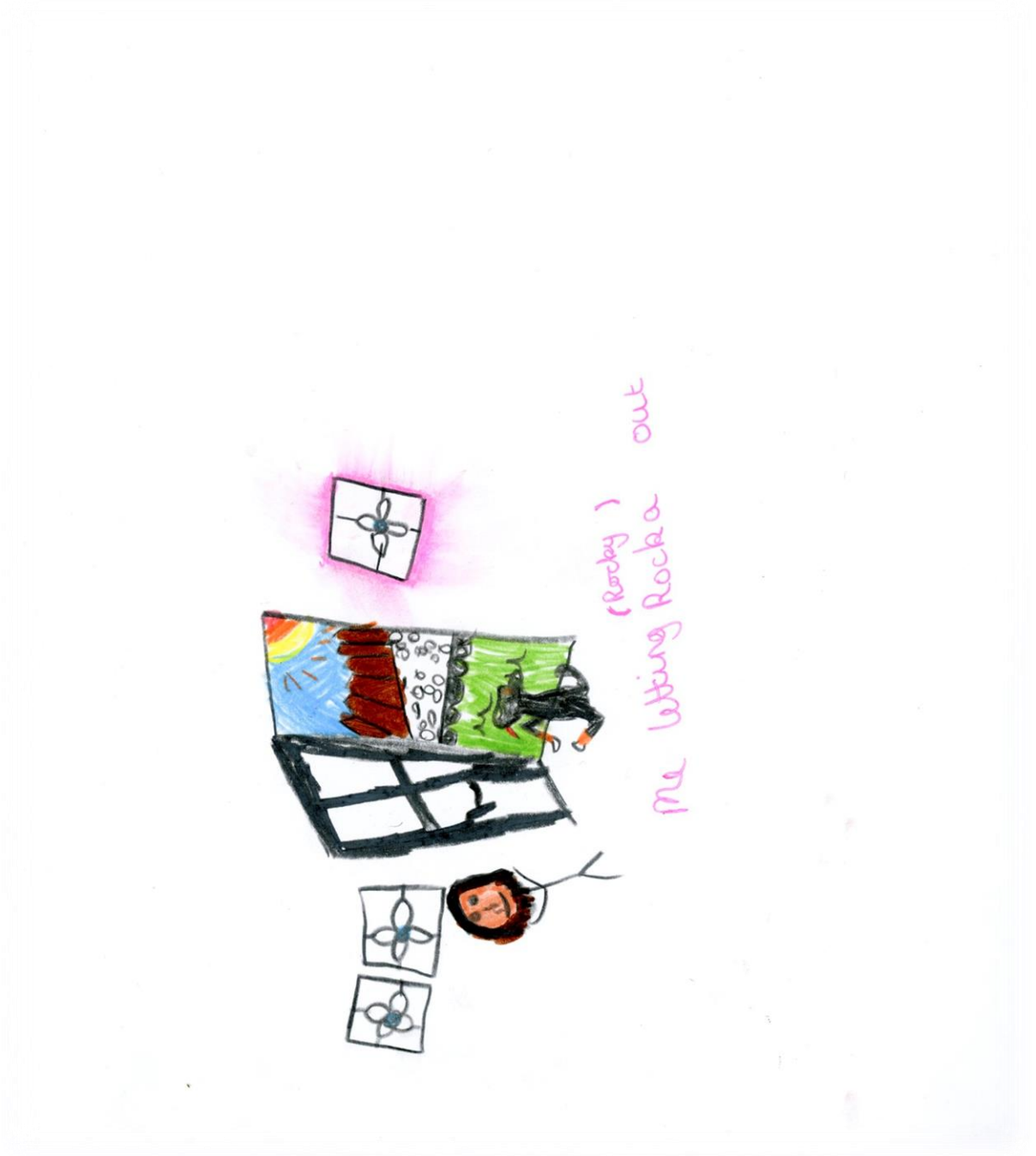
Well arthritis is really just something wrong with you. It is nothing to worry about. Some times it's you're finger some times its youre elbow Some times it's both. It sometim~~efects~~s it moves around from one joint to another. you may need to take Medicne called a tablet. When your Mummy or daddys joint is saw they may need to rest.

Appendix J - Writing 2 by Sara Jackman, aged 7.

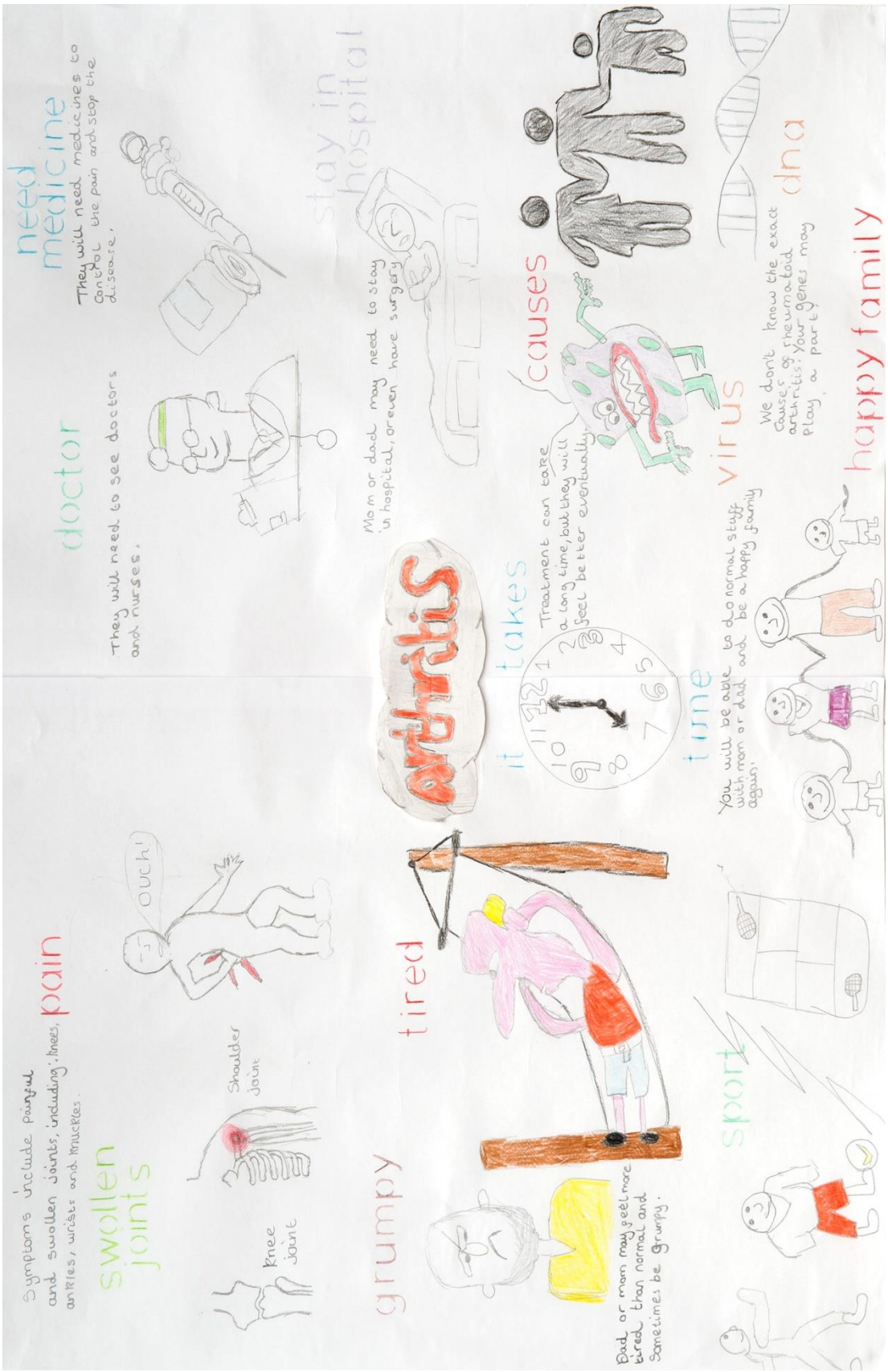
injection

If you have something wrong with your finger you may need to have a needle poked threw your finger this is called an injection It can hurt but the nurse's magic freeze. Hopefully your finger will feel better the next day after some rest.

Appendix K - Drawing by Ruth Stevens, aged 9.

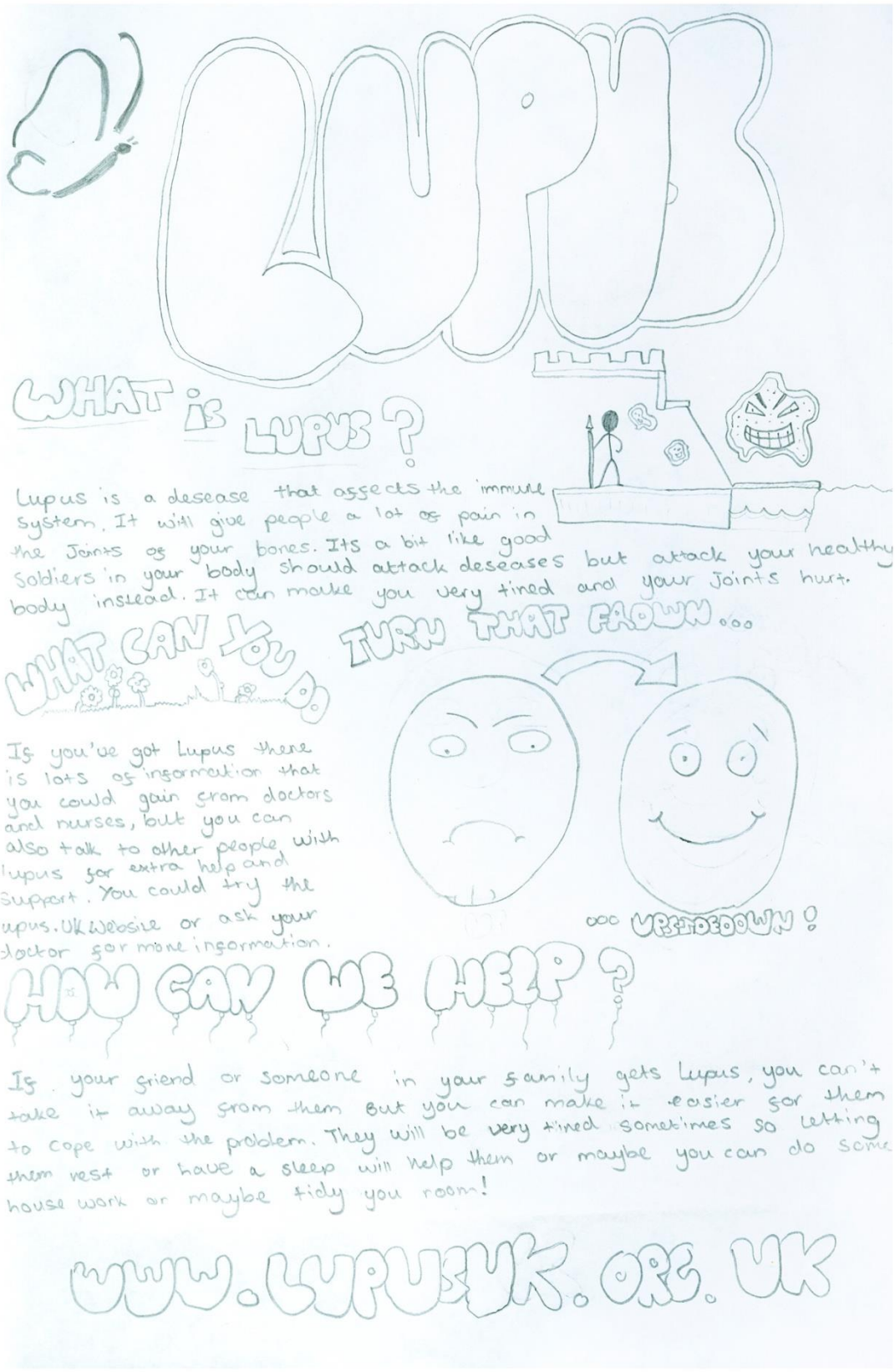


Appendix L - Poster by Theo Baxter, aged 11.



Appendix M - Poster by Elise Underwood, aged 9.

Appendix N - Poster by Harry Tiverton, aged 11.



Appendix O - Targeted Journal Style Guidelines

British Journal of Health Psychology

Edited By: Alison Wearden and David French

Impact Factor: 2.895

ISI Journal Citation Reports © Ranking: 2015: 21/121 (Psychology Clinical)

Online ISSN: 2044-8287

Author Guidelines

The aim of the British Journal of Health Psychology is to provide a forum for high quality research relating to health and illness. The scope of the journal includes all areas of health psychology as outlined in the Journal Overview.
The types of paper invited are:

• papers reporting original empirical investigations, using either quantitative or qualitative methods, including reports of interventions in clinical and non-clinical populations;
• theoretical papers which may be analyses or commentaries on established theories in health psychology, or presentations of theoretical innovations;

• we particularly welcome review papers, which should aim to provide systematic overviews, evaluations and interpretations of research in a given field of health psychology; and

• methodological papers dealing with methodological issues of particular relevance to health psychology.

All papers published in The British Journal of Health Psychology are eligible for Panel A: Psychology, Psychiatry and Neuroscience in the Research Excellence Framework (REF).

1. Circulation
The circulation of the Journal is worldwide. Papers are invited and encouraged from authors throughout the world.

2. Length
Papers describing quantitative research (including reviews with quantitative analyses) should be no more than 5000 words (excluding the abstract, reference list, tables and figures). Papers describing qualitative research (including reviews with qualitative analyses) should be no more than 6000 words (including quotes but excluding the abstract, tables, figures and references). The Editors retain discretion to publish papers beyond this length in cases where the clear and concise expression of the scientific content requires greater length.

3. Editorial policy

The Journal receives a large volume of papers to review each year, and in order to make the process as efficient as possible for authors and editors alike, all papers are initially examined by the Editors to ascertain whether the article is suitable for full peer review. In order to qualify for full review, papers must meet the following criteria:

• the content of the paper falls within the scope of the Journal

• the methods and/or sample size are appropriate for the questions being addressed

• research with student populations is appropriately justified

• the word count is within the stated limit for the Journal (i.e. 5000 words)

4. Submission and reviewing
All manuscripts must be submitted via Editorial Manager. The Journal operates a policy of anonymous (double blind) peer review. We also operate a triage process in which submissions that are out of scope or otherwise inappropriate will be rejected by the editors without external peer review to avoid unnecessary delays. Before submitting, please read the terms and conditions of submission and the declaration of competing interests. You may also like to use the Submission Checklist to help your prepare your paper.

5. Manuscript requirements
• Contributions must be typed in double spacing with wide margins. All sheets must be numbered.

• Manuscripts should be preceded by a title page which includes a full list of authors and their affiliations, as well as the corresponding author's contact details. A template can be downloaded from here.
• For articles containing original scientific research, a structured abstract of up to 250 words should be included with the headings: Objectives, Design, Methods, Results, Conclusions. Review articles should use these headings: Purpose, Methods, Results, Conclusions. As the abstract is often the most widely visible part of your paper, it is important that it conveys succinctly all the most important features of your study. You can save words by writing short, direct sentences. Helpful hints about writing the conclusions to abstracts can be found here.
• Statement of Contribution: All authors are required to provide a clear summary of 'what is already known on this subject?' and 'what does this study add?'. Authors should identify existing research knowledge relating to the specific research question and give a summary of the new knowledge added by your study. Under each of these headings, please provide 2-3 (maximum) clear outcome statements (not process statements of what the paper does); the statements for 'what does this study add?' should be presented as bullet points of no more than 100 characters each. The Statement of Contribution should be a separate file.

• Conflict of interest statement: We are now including a brief conflict of interest statement at the end of each accepted manuscript. You will be asked to provide information to generate this statement during the submission process.

• The main document must be anonymous. Please do not mention the authors' names or affiliations (including in the Method section) and always refer to any previous work in the third person.

• Tables should be typed in double spacing, each on a separate page with a self-explanatory title. Tables should be comprehensible without reference to the text. They should be placed at the end of the manuscript but they must be mentioned in the text.

• Figures can be included at the end of the document or attached as separate files, carefully labelled in initial capital/lower case lettering with symbols in a form consistent with text use. Unnecessary background patterns, lines and shading should be avoided. Captions should be listed on a separate sheet. The resolution of digital images must be at least 300 dpi. All figures must be mentioned in the text.

• For reference citations, please use APA style. Particular care should be taken to ensure that references are accurate and complete. Give all journal titles in full and provide doi numbers where possible for journal articles. For example:

Author, A., Author, B., & Author, C. (1995). *Title of book.* City, Country: Publisher.
Author, A. (2013). Title of journal article. *Name of journal, 1,* 1-16. doi: 10.1111/bjep.12031
• SI units must be used for all measurements, rounded off to practical values if appropriate, with the imperial equivalent in parentheses.

• In normal circumstances, effect size should be incorporated.

• Authors are requested to avoid the use of sexist language.

• Authors are responsible for acquiring written permission to publish lengthy quotations, illustrations, etc. for which they do not own copyright. For guidelines on editorial style, please consult the APA Publication Manual published by the American Psychological Association.
• Manuscripts describing clinical trials are encouraged to submit in accordance with theCONSORT statement on reporting randomised controlled trials.
• Manuscripts reporting systematic reviews and meta-analyses are encouraged to submit in accordance with the PRISMA statement.
• Manuscripts reporting interventions are encouraged to describe them in accordance with the TIDieR checklist.
If you need more information about submitting your manuscript for publication, please email Hannah Wakley, Managing Editor (bjhp@wiley.com) or phone +44 (0) 116 252 9504.

6. Supporting information
Supporting Information can be a useful way for an author to include important but ancillary information with the online version of an article. Examples of Supporting Information include appendices, additional tables, data sets, figures, movie files, audio clips, and other related nonessential multimedia files. Supporting Information should be cited within the article text, and a descriptive legend should be included. Please indicate clearly on submission which material is for online only publication. It is published as supplied by the author, and a proof is not made available prior to publication; for these reasons, authors should provide any Supporting Information in the desired final format.

For further information on recommended file types and requirements for submission, please visit the Supporting Information page on Author Services.

7. OnlineOpen
OnlineOpen is available to authors of primary research articles who wish to make their article available to non-subscribers on publication, or whose funding agency requires grantees to archive the final version of their article. With OnlineOpen, the author, the author's funding agency, or the author's institution pays a fee to ensure that the article is made available to non-subscribers upon publication via Wiley Online Library, as well as deposited in the funding agency's preferred archive. A full list of terms and conditions is available on Wiley Online Library.

Any authors wishing to send their paper OnlineOpen will be required to complete thepayment form.
Prior to acceptance there is no requirement to inform an Editorial Office that you intend to publish your paper OnlineOpen if you do not wish to. All OnlineOpen articles are treated in the same way as any other article. They go through the journal's standard peer-review process and will be accepted or rejected based on their own merit.

8. Author Services
Author Services enables authors to track their article – once it has been accepted – through the production process to publication online and in print. Authors can check the status of their articles online and choose to receive automated e-mails at key stages of production. The author will receive an e-mail with a unique link that enables them to register and have their article automatically added to the system. You can then accessKudos through Author Services, which will help you to increase the impact of your research. Visit Author Services for more details on online production tracking and for a wealth of resources including FAQs and tips on article preparation, submission and more.

9. Copyright and licences
If your paper is accepted, the author identified as the formal corresponding author for the paper will receive an email prompting them to login into Author Services, where via the Wiley Author Licensing Service (WALS) they will be able to complete the licence agreement on behalf of all authors on the paper.

For authors signing the copyright transfer agreement
If the OnlineOpen option is not selected the corresponding author will be presented with the copyright transfer agreement (CTA) to sign. The terms and conditions of the CTA can be previewed in the samples associated with the Copyright FAQs .
For authors choosing OnlineOpen
If the OnlineOpen option is selected the corresponding author will have a choice of the following Creative Commons Licence Open Access Agreements (OAA):

- Creative Commons Attribution Non-Commercial Licence (CC-BY-NC)
- Creative Commons Attribution Non-Commercial -NoDerivs Licence (CC-BY-NC-ND)

To preview the terms and conditions of these open access agreements please visit theCopyright FAQs and you may also like to visit the Wiley Open Access Copyright and Licence page.
If you select the OnlineOpen option and your research is funded by The Wellcome Trust and members of the Research Councils UK (RCUK) or the Austrian Science Fund (FWF) you will be given the opportunity to publish your article under a CC-BY licence supporting you in complying with your Funder requirements. For more information on this policy and the Journal's compliant self-archiving policy please visit our Funder Policy page.

10. Colour illustrations
Colour illustrations can be accepted for publication online. These would be reproduced in greyscale in the print version. If authors would like these figures to be reproduced in colour in print at their expense they should request this by completing a Colour Work Agreement form upon acceptance of the paper.

11. Pre-submission English-language editing
Authors for whom English is a second language may choose to have their manuscript professionally edited before submission to improve the English. A list of independent suppliers

of editing services can be found in Author Services. All services are paid for and arranged by the author, and use of one of these services does not guarantee acceptance or preference for publication.

12. The Later Stages

The corresponding author will receive an email alert containing a link to a web site. The proof can be downloaded as a PDF (portable document format) file from this site. Acrobat Reader will be required in order to read this file. This software can be downloaded (free of charge) from Adobe's web site. This will enable the file to be opened, read on screen and annotated direct in the PDF. Corrections can also be supplied by hard copy if preferred. Further instructions will be sent with the proof. Excessive changes made by the author in the proofs, excluding typesetting errors, will be charged separately.

13. Early View

British Journal of Health Psychology is covered by the Early View service on Wiley Online Library. Early View articles are complete full-text articles published online in advance of their publication in a printed issue. Articles are therefore available as soon as they are ready, rather than having to wait for the next scheduled print issue. Early View articles are complete and final. They have been fully reviewed, revised and edited for publication, and the authors' final corrections have been incorporated. Because they are in final form, no changes can be made after online publication. The nature of Early View articles means that they do not yet have volume, issue or page numbers, so they cannot be cited in the traditional way. They are cited using their Digital Object Identifier (DOI) with no volume and issue or pagination information. Eg Jones, A.B. (2010). Human rights Issues. *Journal of Human Rights.* Advance online publication. doi:10.1111/j.1467-9299.2010.00300.x

Further information about the process of peer review and production can be found in this document. What happens to my paper?

Abstracting and Indexing Information

- Academic Search Alumni Edition (EBSCO Publishing)
- Academic Search Complete (EBSCO Publishing)
- Academic Search Elite (EBSCO Publishing)
- Academic Search Premier (EBSCO Publishing)
- Advanced Placement Source (EBSCO Publishing)
- ArticleFirst (OCLC)
- Arts & Humanities Citation Index (Thomson Reuters)
- ASSIA: Applied Social Sciences Index & Abstracts (ProQuest)
- BNI: British Nursing Index (ProQuest)
- Current Contents: Arts & Humanities (Thomson Reuters)
- Electronic Collections Online (OCLC)
- IBR & IBZ: International Bibliographies of Periodical Literature (KG Saur)
- MEDLINE/PubMed (NLM)
- ProQuest Central (ProQuest)
- ProQuest Central: Professional Edition (ProQuest)
- ProQuest Health & Medical Complete (ProQuest)
- ProQuest Medical Library (ProQuest)
- ProQuest Psychology Journals (ProQuest)
- ProQuest Public Health (ProQuest)
- ProQuest Science Journals (ProQuest)
- Psychology & Behavioral Sciences Collection (EBSCO Publishing)
- PsycINFO/Psychological Abstracts (APA)

- SCOPUS (Elsevier)
- Social Sciences Citation Index (Thomson Reuters)
- SocINDEX (EBSCO Publishing)
- SocINDEX with Full Text (EBSCO Publishing)
- Sociological Abstracts (ProQuest)
- VINITI (All-Russian Institute of Science & Technological Information)
- Web of Science (Thomson Reuters)

CHAPTER FOUR

Research Report Critical Appraisal

Overview

This chapter will offer a reflection on, and critique, of the research methodology used, and a reflection on the experience of the research process as a whole. The implications for future research will also be examined.

1.0 The Research Process:-

It may be useful to provide the context in which the study occurred, as the entire research process was developed over a number of years and whilst at times was exciting, emotional and intensely frustrating; this has ultimately given me a wealth of experience that I can use in the future, and hopefully pass on to others entering the tricky world of 'doing research'. These experiences will also shape how future research strands may grow from this project.

The initial idea for the research, if I am entirely honest, came froma decision to break my usual habits and have breakfast in front of the TV news. An interview with Rasheda Ali, daughter of the late boxer Mohammed Ali, sent me post-haste into work. She was describing how she could find no information for her children to explain what was happening to her father, who by then had developed Parkinson's disease. Finding no resources, she had written her own book for

children (and grandchildren) to help them understand the condition (Ali, 2005). This interview struck a chord. I wanted to go back and re-read data that I had collected a year previously from women with systemic lupus erythematosus (SLE). I remembered that, whilst not the focus of the interviews, some of the mothers had talked about their difficulties in managing their disease with their families, and communicating with their children about it. I also wanted to check what information was available for children to help them understand rheumatic conditions in parents/grandparents. After all, there were estimated to be over 200 rheumatic diseases, surely someone had thought to tackle this important topic? Surprisingly, it appeared not. Whilst there were leaflets for 'carers', these were clearly aimed at adults, and were not suitable for children. My data, read with a fresh perspective, suggested that this was an idea to be explored.

A few years later, the process of trying to obtain funding for the project in order to combine with a part-time research degree, provided the opportunity to write grant proposals, answer peer review questions, attend facilitation meetings, and experience being interviewed by small (Arthritis Research UK [ARUK]) and large (National Institutes for Health Research [NIHR]) funding panels. As part of my 'day job' involves working for the research and development department (R&D) of the local NHS Trust, obtaining funding for the project was very important. Eventually, a call for research for educational project grants by ARUK offered an opportunity for the project (main study) to be part-funded and combined with the professional doctorate in psychology (PsyD).

Undoubtedly, registering for the PsyD added weight to the funding application, but also meant that one had to juggle the requirements of funders, the NHS Trust (R&D duties), the NHS Trust (clinical work) and the requirements of the PsyD, one of which was the time-frame available to carry-out the research project:

> If you ever have the misfortune to need to get interviewees via the NHS Research Ethics Service, then there are two things to bear in mind: first, that you should brace yourself for an endurance test in bureaucracy; and second, that all of those plans that impressed your supervisor/upgrade panel need to be thrown in the bin....after a full year's delay from when you first tried to get permission to speak to people, and having already tested the patience of all the GPs who kindly agreed to let you research their patients, you have to do your research in the fastest and most hassle-free way possible.
>
> (Ben Baumberg, 2012, p.37)

My own perspective was not quite as bad. However, registering for the PsyD, then spending nearly a year finishing the funding process and applying for NHS research ethics approval certainly left less time to complete the research project than I would have liked within the timeframe of the PsyD. I am not unfamiliar with the nuances of the research ethics process, and knew enough, for example, that the participant information sheet guidance is not, in fact, guidance, but rather a required version likely to require amendments if deviated from.

Such anticipation does not protect one from the requests of lay members of the ethics panel, and even slight changes can effectively hold up the project whilst addressing the comments and re-submitting.

I was surprised by the difficulties I then experienced in recruiting participants to the study, given the large potential pool of patients (and families) available. Cree, Kay & Tisdall, (2002) described similar difficulties with recruitment of children, in particular how researchers had to gain access via a series of 'gatekeepers'. Ethically, I was not allowed to approach patients (parents) directly to ask them to take part in the study. I, therefore, relied heavily upon the Consultants, Registrars and Clinical Nurse Specialists (CNS) to ask their patients if they had children between the ages of seven and eleven years at the end of their consultation; and, if they did, hand them the research information pack with a brief overview of the study, and/or an offer of meeting either me or a research facilitator to discuss further. I faced the dual 'gatekeeper': access via health professionals, followed via parents for their children to participate. Parents could easily refuse to take part, and even having chosen to take part, could withdraw their children from the study, as indeed two participants opted to do.

Reflecting upon why few participants were being referred to me, I considered the factors that might be preventing the health care team from recruiting to my study. As I worked in a supportive environment for research, I did not consider that they were simply not engaged with the project, or objected from an ontological position. At least two of the consultants had used qualitative work as part of their own PhD theses,

as had one of the CNSs. I put large signs in the consulting rooms reminding them of the recruitment guidelines and put the information packs within easy reach. Reminder e-mails were sent to the Rheumatology team every Monday morning, verbal reminders were re-iterated at team meetings, and I gave presentations within the education and research meeting times, unfortunately, all to no avail.

I considered whether I was being unrealistic in expecting my colleagues to spend time (albeit brief) outlining the study to their patient and handing over an information pack. After all, clinics in a secondary hospital environment are extremely busy, with the main focus being on patient well-being. Also, this large Rheumatology Department recruits patients and collects data for multiple research studies. Pharmacological trials mean that medics are focused upon certain characteristics within a patient group, and whether they might be suitable for access to medications that may be of immediate benefit, rather than upon psychosocial projects which may be considered useful, but carry little perceived immediate benefit. Additionally, some studies qualify for entry to the NIHR Portfolio of Studies (www.crn.nihr.ac.uk/can-help/funders-academics/nihrcrn-portfolio/),where recruitment numbers effectively earn financial gains for R&D departments; hence there can be pressure (perceived or real) to recruit to such studies that carry future rewards. In this respect, there was little recognition that my study could qualify for entry to the NIHR Portfolio. As the Service Evaluation aspect of the PsyD was included in the portfolio entry, my recruitment figures far surpassed any study that was being undertaken within the department

(and within the hospital at that time). Perhaps, even in such a supportive research environment there remains a hurdle between natural and social sciences that has yet to be overcome fully. Eventually, two CNS's proved the most effective at recruitment, because of their detailed knowledge of family characteristics, and being pro-active before appointments.

One additional factor that impacted on the time-frame for the study was arranging convenient times for families to attend the department for the interviews. This meant that families opted for after school or early evening, so that parents could attend after work; or that they asked for interviews to be held when the children had a day off school or during school holidays. This meant that interviews had to wait for such popular periods, and were often cancelled and re-booked at the last minute. Coupled with the initial slow recruitment, the initial two interviews took much longer to conduct than anticipated.

There are undoubtedly challenges to carrying out research within the NHS. Some stem from my own errors, for example putting up reminder notices in consulting rooms only to find them all removed the next day by the nurse in charge (apparently an infection risk unless laminated first), taught me to always ask before doing *anything* in a clinical area, no matter how innocuous it seemed. Checking the setting is another lesson for the future. Many of the children had never attended the Unit or even the hospital with their parents before. On walking out of the Unit after the first interview, I realised to my horror that our large plastic skeleton used for teaching was hanging in full view. Fortunately,

'Luke' (child) was fascinated and asked to have a closer inspection. Indeed, asking the children as they left "would you like to see our skeleton?" proved something of a success.

Other strategies required increased awareness that tact and diplomacy is needed to achieve your aim whilst not upsetting others, for instance, informing the senior nurse in charge about the project and any potential impact on junior nurses' time; and warning the reception staff that families would be attending late in the day, so that they were not turned away. One unlikely ally came from the domestic staff who carried out their work in the late afternoon/early evening. Aware that audio-recording would pick up noise from vacuuming and cleaning (something I had not thought of), they took care to work at the other end of the floor until I had finished. Additionally, although our Unit is locked after 5.00pm, they generously remained in visible proximity until all participants had left, to ensure my safety.

2.0 Research Methodology:-

Whilst I had used semi-structured interviews with adult participants in the past, I had not used this approach with families. It was interesting, but not surprising, that all but one family opted to be interviewed together. This presented both pros and cons - children were reassured having their mother and father (and in some cases siblings) with them, and so relaxed more easily, but the presence of parents may have restricted what they would otherwise have said. Assurances that they could be honest, and would not get into trouble for any views

expressed, always reiterated by the parents, may not have been enough to let them feel entirely free, and so there is a possibility that there were limitations within their views. Equally, the parents appeared to occasionally modify what they wanted to say, or how they wanted to say it, in the presence of their children, particularly where younger siblings were present. I also had concerns that children might agree too easily with me, being conscious of the power dynamic inherent in these situations.

Nevertheless, it was clear that, once children settled, they were quite happy to disagree when I suggested something they were not keen on - for example, in a discussion about whether to include information on the immune system in any resources for them, one child said "Mm, I wouldn’t be excited to know about it" (Belle). This is an excellent reminder that materials written *by* adults *for* children could easily contain information that children themselves do not find relevant or interesting. To facilitate questioning, the approach suggested by Danby, Ewing & Thorpe (2011) was useful, i.e. trying a 'thinking out loud' way of asking a question, which shows that you really want to hear what the child has to say, and is more likely to produce a more open response. Ultimately, the trustworthiness of the data relies in part on ensuring that it does reflect the genuine views of the participants.

Children, like their parents, found it difficult to articulate what the content of resources should be. Adults were concerned that they were pitched at the right 'level' but found it hard to articulate what this should be. Equally, children were unsure what they would like to know about. I

found that I had to make suggestions to them, which may have appeared leading. However, these suggestions emerged from the service evaluation results, and so were grounded (at least for the parents perspectives) in research. Equally, awareness of the emerging illness perceptions model guided the later introduction of topics into the interviews. Difficulties in verbalising ideas were in some part resolved by using visual methods by the children, which was the reason they had been initially chosen.

The introduction of visual methods as part of the data collection process was carefully considered. Travers (2011) asks whether visual methods add anything substantial to a study, or are they in danger of being simply decorative and diversionary from the 'real' point? They might be 'innovative', but there has to be a real reason for using them. Visual methods have been recommended as a way of engaging children in 'true' participatory child-centred methods to give them a voice about matters that concern and have impact on them (Coad, Plumridge & Metcalfe, 2009). The 'spider' diagram I used was intended to be a 'warm up' exercise, given that I had no opportunity to meet or spend time with the children prior to the interviews to put them at ease with me. The idea for this came from research training, where I learned about the use of genograms to facilitate clinical interviews. I could see that the method was adaptable for research to engage children, as well as to gain information that would be useful to have included in the interview itself. The diagram was pre-prepared for structure and guidance, and allowed me to observe how the child engaged and responded to questions, and

how the family communicated, in addition to being an ice-breaking exercise.

Visual methods are generally popular, not perceived as difficult or threatening, and appropriate for all cultural backgrounds (Merriman & Guerin, 2006). As there are different ways of using drawing as a research method, this was given some thought. Two similar techniques, the 'draw and tell' (Amuyunzu-Nyamongo et al., 2011) or 'draw and write' (Youssef, Salah, Salem & Megahed, 2010) have been used where children talk about what they are drawing and why, as they do it, or similarly draw and write about their pictures as they do them. I did not consider that these methods would produce useful results; firstly, because the families would have had to spend much longer at the hospital at the end of the day, and; secondly, because writing assumes that a child has the ability to express themselves well via this medium which could have put a barrier rather than a facilitator into the process. Other forms of visual data collection were considered such as 'photo-voice', where the children could have taken photos that they felt were relevant to their situation; this, however, raises difficulties in terms of expertise, and the ethics of anonymity and informed consent. Consent has to be sought from anyone contained within a photograph. Furthermore, consent is required for dissemination of the photograph. Today, when the visual image can be taken and shared easily across social media without consent necessarily being sought by third parties, there are real ethical issues to be considered. As children as young as seven years may not fully understand the concept of 'consent', as well

as being unable to gain it, this was not an appropriate method to use in this age group. Children also struggled with the concept of anonymity, not understanding why they were not allowed to put their names on their drawings and posters for people to see, as they were used to doing in school.

Researchers have found differing attitudes to the maintenance of anonymity within the research setting. Corden and Sainsbury (2006) found that participants disliked the idea of pseudonym's and preferred quotations to be unattributed, whilst the participants in the study carried out by Saunders, Kitzinger and Kitzinger (2015) asked for pseudonym's to be used as they felt that they reminded readers that there were real people behind the words.

Mindful that the children would be potentially sharing sensitive information via their drawings, I requested that they completed those at home, where their parent would be available should they become distressed or have any questions (Coad, 2009). This, of course, presented something of a double-edged sword, as I could not be sure to what degree parents would interfere in the content of the drawings, and could instead represent their views rather than those of their child, against what Mayaba and Wood (2015) call 'scaffolding', where the parent supports the child in their task.

It was clear that children who engaged with drawing or preparing a poster were happy to return with it and talk about it. I found that parents were at pains to state that the child had either completed the drawing

unsupervised, or had determined the content for themselves. This was then explored with the parent, particularly when internet research was used to find and develop their own information. The drawing task undoubtedly facilitated family conversations about the parents' rheumatic condition that they had not had before, and in a way acted as an intervention in its own right. The children actively constructed and interpreted their knowledge through the task set, thus providing a unique view of their 'reality'.

3.0 Demonstrating rigour:

Firstly, it is worth addressing the ontological and epistemological position taken a little further (See Appendix A, Research report) from a personal viewpoint. As Corbin and Strauss (2008) note, whilst researchers will answer research questions appropriately, often they have a preferred way of viewing the world, and how we can come to understand it. I have to agree that instinctively I am at heart a relativist, and enjoy qualitative methods. The simplicity of "if you want to know what a person thinks about something, ask them; they *may* tell you" (Merriman & Guerin, p.49, 2006) has always appealed. This does not mean that one shoehorns one's project into a particular stance regardless; but rather that the process of judging the quality and rigour of qualitative research, and the likelihood of it being useful, applicable in the real world and, ultimately publishable in peer reviewed academic journals and proceedings, lies in the openness of the researcher in acknowledging these perspectives amongst other issues.

This also involves being clear about the *active* element of qualitative analysis, as one brings experience and knowledge to the enterprise. Braun and Clarke (2013) are clear that, whilst the analysis is rooted in the data, you create the analysis from this *and* your expertise and skills, choosing what is relevant and what is not. During the process of analysing the data, I reflected upon whether I was seeing patterns in the data that were not there. Would another researcher draw the same conclusions, although they may not have been expressed in the same way, if *they* looked at the same data set? To guard against this, as well as asking an experienced CNS with qualitative research experience to read my interpretations of the data, I asked "what is going on here"? "what are these people saying"? Thus guarding against 'bracketing' my knowledge, whilst ensuring that the knowledge was not clouding the *actual* interpretation of the data.

The importance of ensuring rigour in qualitative approaches, and the difficulties quantitative researchers have with evaluating qualitative styles of research (particularly with anything so novel as visual data), because of the dominance of using positivist ways of evaluating research methods that use anything other than empirical quantitative methods, was made clear to me during an encounter with a consultant rheumatologist at the British Society for Rheumatology conference where I was presenting a poster based on my research project, which included some of the children's drawings. After responding to questions about measurement, reliability, validity and generalizability (all from a

quantitative positivist stance), there was a short pause before the pronouncement "no, you'll never get this published ".

As a health psychologist working within a medical specialty, I have long recognised that when it comes to dissemination of research, if you want to reach the audience that can help to facilitate the changes you recommend, you are going to be talking to, and publishing largely within the world of academic medicine. Historically, psychology was keen to align itself with positivism, which sought to use the realist ontology and epistemologies of natural sciences, thus defining itself as a 'scientific' discipline. The quantitative methods adopted have remained largely the dominant paradigm in the teaching and training of psychologists ever since. The specialist discipline of health psychology has followed this trend, partly due to its close alliance to medicine (Murray & Chamberlain, 1999) and the reluctance of other allied medical professions to challenge the dominant paradigm. For me, this has begun to feel like something of an 'own goal', as it is so difficult to emerge from the quantitative paradigms dominating the profession and to convince others of the value of qualitative methodologies.

I have attempted to demonstrate the quality of the study via a number of means. Firstly, many of the traditional ways of evaluating qualitative work are not applicable, as they rest upon positivist realist assumptions, for example the concept of generalizability. As my epistemological position is one of accepting there are multiple ways of viewing the world, I do not expect my results to be generalizable in the traditional sense, but I do hope that they are detailed enough to prove

trustworthy and credible within the situation (valid and reliable). They may, therefore, be useful or 'transferable' for others in similar situations.

Secondly, quality may be shown by transparency throughout the research process. Recruitment and interviews were carried out until no new information was forthcoming, and the accounts provided a depth of information to answer the research question. I acknowledged my ontological and epistemological views, as well as my active role in the analysis. Clearly outlining the analytic procedure would allow another researcher to follow my processes, and there is a clear audit trail locating the data excerpts back to the text. The study is the first of its kind in rheumatology, and therefore its findings may have significant impact for others in a similar context.

4.0 Future research:-

The study has offered some key messages for the future. It is clear that parents and children would welcome resources that enable them to have discussions about their rheumatic disease, and it is also clear that joint resources - one for parents and one for children - would be welcomed. This is because parents need guidance about how to answer questions in a simple and appropriate way. Illness coherence, or understanding one's condition, is key to enabling this to happen. As you cannot tell others about something you do not understand sufficiently well yourself, the goal should be to enable children to develop an illness coherence about their parent's condition. The illness perceptions model

would be an appropriate way to develop topics within the resource, using its framework to structure information.

Future research should focus on, firstly, developing and piloting resources for children aged seven to eleven years, and secondly developing a new strand of research that looks at the resource requirements of adolescents aged 12 to 18. Barriers and facilitators to the eventual delivery of resources within the healthcare setting would also need assessing. Opportunities from existing funders and from national rheumatology charities will be sought to develop and pilot resources using the recommendations from the parents and children who took part in the study.

References

Ali, R. (2005). *I'll hold your hand so you won't fall: a child's guide to Parkinsons disease.* Merit publishing International.

See also: https://www.theguardian.com/uk/2005/apr/06/sport.books

Amuyunzu-Nyamongo, M., Tchounkeu, Y., Oyugi, R., Kabali, A., Okeibunor, J.C., Manianga, C., & Amazigo, U. (2011). Drawing and interpreting data: Children's impressions of onchoceriasis and community-directed treatment with ivermectin (CDTI) in four onchoceriasis endemic countries in Africa. *International Journal of Qualitative Studies in Health and Well-being, 6,* 5918. doi: 10.3402/qhw.v6i2.5918

Baumberg, B. (2012). In S.E Baker & R. Edwards (Eds.), *How many qualitative interviews are enough? Expert voices and early career reflections on sampling and cases in qualitative research.* Discussion paper. NCRM. (Unpublished). Retrieved from http://eprints.ncrm.ac.uk/2273/4/how_many_interviews.pdf

Braun, V., & Clarke, V. (2013). *Successful Qualitative Research: A practical guide for beginners.* London: Sage.

Coad, J., Plumridge, G.,& Metcalfe, A. (2009) Involving children and young people in the development of art-based research tools. Nurse *Researcher, 16(4),* 56-64. doi: 10.7748/nr2009.07.16.4.56.c7161

Corbin, J., & Strauss, A. (2008). *Basics of Qualitative Research*. London: Sage.

Corden, A., Sainsbury, R. (2006) Exploring 'Quality': Research Participants' Perspectives on Verbatim Quotations. *International Journal of Social Research Methodology, 9(2),* 97-110.
doi: 10.1080/13645570600595264

Cree, V., Kay, H., & Tisdall, K. (2002). Research with children: Sharing the dilemmas. *Child and Family Social Work, 7,* 47-56.
doi: 10.1046/j.1365-2206.2002.00223.x

Danby, S., Ewing, L.,& Thorpe, L. (2011). The novice researcher: Interviewing young children. *Qualitative Inquiry, 17(1),* 47-84.
doi: 10.1177/1077800410389754

Mayaba, N.N & Wood, L. (2015). Using drawings and collages as data generation methods with children: Definitely not child's play. *International Journal of Qualitative Methods,*1-10.
doi: 10.1177/1609406915621407

Merriman, B., & Guerin, S. (2006). Using children's drawings as data in child-centred research. *The Irish Journal of Psychology, 27(1-2),* 48-57.
doi:10.1080/03033910.2006.10446227

Murray, M., & Chamberlain, K. (1999). Health psychology and qualitative research. In M. Murray, K. Chamberlain (Eds.), *Qualitative health psychology: Theories and methods* (pp.133-147). London: Sage.

Saunders, B., Kitzinger, J., & Kitzinger, C. (2015). Anonymising interview data: Challenges and compromise in practice. *Qualitative Research,15(5),* 616-632. doi: 10.1177/1468794114550439

The NIHR Clinical Research Network (NIHR CRN) Portfolio

https://www.crn.nihr.ac.uk/can-help/funders-academics/nihrcrn-portfolio/

Travers, M. (2011). Visual methods: Innovation, decoration or distraction? *Qualitative Researcher,13,* 5-7. Accessed at: http://www.wiserd.ac.uk/publications/info/qualitative-researcher/

Youssef, M.M., Salah, E.M., Salem, S.M.E., & Megahed, H.S. (2010). Concepts of health and illness in Egyptian children. *Medical Journal of Cairo University, 78(1),* 77-84. Retrieved from http://cu.edu.eg

Appendix A: Chronology of Research Process

	2011 PRE-PSYD							2012 YEAR ONE													2013 YEAR TWO											
TASK	**J**	**A**	**S**	**O**	**N**	**D**		**J**	**F**	**M**	**A**	**M**	**J**	**J**	**A**	**S**	**O**	**N**	**D**		**J**	**F**	**M**	**A**	**M**	**J**	**J**	**A**	**S**	**O**	**N**	**D**
SUBMIT EXPRESSION OF INTEREST TO FUNDING BODY AR UK	X																															
MENTORING MEETING AR UK				X																												
INVITATION TO APPLY FOR FUNDING AR UK					X	X		X	X																							
PSYD REGISTRATION								X																								
IDENTIFY Y1 TRAINING PLAN									X																							
RESEARCH TRAINING									X		X	X	X	X									X									
LITERATURE SEARCHING TRAINING									X	X																						
SYSTEMATISED LITERATURE REVIEW COMMENCES & DATABASE										X																						
DEVELOPMENT OF INFORMATION SHEETS & QUESTIONNAIRES										X	X																					
DEVELOPMENT OF INTERVIEW SCHEDULE & MATERIALS										X	X	X	X	X	X	X																
FUNDING PEER REVIEW										X																						
FUNDING AWARDED													X																			
FUNDING STARTS																X																
PATIENT PARTNERS APPROVE QUESTIONNAIRE													X	X	X																	

Chronology of Research Process (continued)

	2012 YEAR ONE							2013 YEAR TWO																							
TASK	**J**	**A**	**S**	**O**	**N**	**D**		**J**	**F**	**M**	**A**	**M**	**J**	**J**	**A**	**S**	**O**	**N**	**D**		**J**	**F**	**M**	**A**	**M**	**J**	**J**	**A**	**S**	**O**	**N**	**D**
LOCAL R&D PROCESS FINALISED																**X**																
ELIGIBLE FOR NIHR PORTFOLIO														**X**																		
NHS ETHICAL REVIEW APPLICATION														**X**																		
NHS ETHICAL REVIEW MEETING															**X**																	
NHS ETHICAL REVIEW GRANTED																**X**																
RECRUITMENT COMMENCES DGH																**X**	**X**	**X**	**X**		**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**
RECRUITMENT COMMENCES NRAS																		**X**	**X**		**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**
RECRUITMENT COMMENCES LUPUS UK																		**X**	**X**		**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**
RECRUITMENT COMMENCES NASS																					**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**
RECRUITMENT COMMENCES SCLERODERMA SOCIETY																									**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**
RECRUITMENT INVITED Psoriasis Association																								**X**	**X**							
PORTFOLIO UPLOAD COMMENCES																	**X**	**X**	**X**		**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**

Chronology of Research Process (continued)

	2012 YEAR ONE							2013 YEAR TWO												2014 YEAR THREE												
TASK	**J**	**A**	**S**	**O**	**N**	**D**		**J**	**F**	**M**	**A**	**M**	**J**	**J**	**A**	**S**	**O**	**N**	**D**		**J**	**F**	**M**	**A**	**M**	**J**	**J**	**A**	**S**	**O**	**N**	**D**	
INTERVIEW RECRUITMENT COMMENCES DGH					**X**	**X**		**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**															
INTERVIEWS										**X**		**X**	**X**	**X**	**X**				**X**		**X**												
VISUAL DATA COLLECTION										**X**		**X**	**X**	**X**	**X**	**X**	**X**		**X**														
INTERVIEW TRANSCRIPTIONS																	**X**	**X**										**X**			**X**		
N-VIVO TRAINING													**X**																				
SPSS TRAINING												**X**										**X**				**X**				**X**			

	2015 YEAR FOUR													2016 YEAR FIVE																			
TASK	**J**	**F**	**M**	**A**	**M**	**J**	**J**	**A**	**S**	**O**	**N**	**D**		**J**	**F**	**M**	**A**	**M**	**J**	**J**	**A**	**S**	**O**	**N**	**D**								
SPSS ANALYSIS	**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**																							
WRITING						**X**	**X**	**X**	**X**	**X**	**X**	**X**																						

Chronology of Research Process (continued)

2012 YEAR ONE														2013 YEAR TWO												
PROJECT MANAGEMENT & DEVELOPMENT	**J**	**F**	**M**	**A**	**M**	**J**	**J**	**A**	**S**	**O**	**N**	**D**		**J**	**F**	**M**	**A**	**M**	**J**	**J**	**A**	**S**	**O**	**N**	**D**	
SUPERVISORY MEETINGS		**X**		**X**			**X**																			
PROGRESS REPORTS												**X**													**X**	
REQUIRED REPORTS																								**X**		
PRESENTATIONS																							**X**			
THESIS WRITING																										

2014 YEAR THREE														2015 YEAR FOUR												
PROJECT MANAGEMENT & DEVELOPMENT	**J**	**F**	**M**	**A**	**M**	**J**	**J**	**A**	**S**	**O**	**N**	**D**		**J**	**F**	**M**	**A**	**M**	**J**	**J**	**A**	**S**	**O**	**N**	**D**	
SUPERVISORY MEETINGS																		**X**	**X**		**X**	**X**				
PROGRESS REPORTS												**X**														
REQUIRED REPORTS					**X**																			**X**		
PRESENTATIONS		**X**								**X**				**X**			**X**									
THESIS WRITING			**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**		**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**	

2016 YEAR FIVE																										
PROJECT MANAGEMENT & DEVELOPMENT	**J**	**F**	**M**	**A**	**M**	**J**	**J**	**A**	**S**	**O**	**N**	**D**		**J**	**F**	**M**	**A**	**M**	**J**	**J**	**A**	**S**	**O**	**N**	**D**	
SUPERVISORY MEETINGS	**X**			**X**			**X**			**X**		**X**														
PROGRESS REPORTS	**X**																									
REQUIRED REPORTS																										
PRESENTATIONS	**X**																									
THESIS WRITING	**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**	**X**														