Short communication

Lingual necrosis secondary to mucormycosis

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Abstract

Mucormycosis is a rare fungal infection with a high mortality that affects immunocompromised patients. This is an unusual case of necrosis of the tongue in a patient with pulmonary mucormycosis, diabetes mellitus, and aplastic anaemia.

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Introduction

Mucormycosis is a rare fungal infection that mainly affects the sinuses, brain, and lungs, often in immunocompromised patients. The major route of infection is inhalation. Effective treatment requires high doses of antifungal agents, correction of risk factors, and surgery.¹ The tongue is a rare location for necrosis because of its excellent blood supply and collateral circulation. Lingual necrosis is most commonly reported secondary to temporal arteritis.

We report a case of pulmonary mucormycosis complicated by oral involvement of the tongue in a patient with aplastic anaemia and poorly controlled diabetes.

Case report

A 69-year-old Asian woman with type I diabetes mellitus was diagnosed with aplastic anaemia. She had recently been treated with antithymocyte globulin. During this course

of treatment she developed fever and sepsis. A thoracic computed tomogram showed consolidation within the middle and lower lobes of the right lung. Sputum culture yielded *Mucor*, *Candida famata*, and mixed coliforms. Blood cultures showed *Klebsiella oxytoca*. She had been started on meropenem, gentamicin, and posaconazole and amphotericin B to treat pulmonary mucormycosis.

Examination of her oral cavity showed a black, necrotic appearance on the anterior two-thirds of the tongue (Fig. 1). A diagnosis of oral mucormycosis was made. This was confirmed histopathologically and was reported as ulcerated and necrotic skeletal muscle and adipose tissue with abundant invading hyphae (Fig. 2).

She was commenced on granulocyte infusions, gentamicin, clarithromycin, voriconazole and amphotericin B given intravenously. Her general condition improved and the oral mucormycosis resolved.

Discussion

Mucormycosis is an opportunistic infection. Causative fungi include *Rhizopus* (most common), *Rhizomucor*, *Cunninghamella*, *Apophysomyces*, *Saksenaia*, *Absidia*, *Mucor*,

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Fig. 1. Black, necrotic tongue on admission (published with the patient's consent).

Syncephalastrum, *Cokeromyces*, and *Mortierella* (least common).² These fungi are found in soil, decaying organic matter, and in manure, fruit, and bread. It is uncommon for mucormycosis to affect healthy individuals. Predisposing medical conditions include poorly controlled diabetes, patients with transplanted organs, cirrhosis, immunosuppressive treatment, and malignancy.¹ Between 40% and 70% of cases have been reported to be associated with diabetes mellitus; hyperglycaemia encourages fungal growth and impairs neutrophil chemotaxis.³

The diagnosis of mucormycosis depends on histological findings of irregularly shaped, broad, non-septate hyphae invading the tissue, and cultures are required to identify the causative fungi.² In our case, the diagnosis of mucormycosis resulted from a positive sample of sputum; the clinical picture and histological findings were consistent with the condition. The organisms have a predilection for directly invading the walls of blood vessels and for producing vascular thrombosis, which leads to ischaemia and necrosis of tissue.⁴

Mucormycosis can present in a variety of clinical forms, the commonest being the rhinocerebral form. Others pre-

Fig. 2. Necrotic tissue showing non-pigmented non-septated hyphae of variable thickness. These morphological features define *Mucor* (haematoxylin and eosin, original magnification 400×).

sentations include cutaneous, gastrointestinal, pulmonary, and disseminated forms.² Cases affecting the oral cavity⁴ have been documented as have those relating to dental extractions,⁵ but we know of only two cases documented on lingual mucormycosis, both of which were thought to have occurred because of the use of infected tongue depressors.^{6,7}

Early diagnosis and rapid initiation of appropriate antifungal agents, correction of the host defect that predisposes to the infection, and surgical debridement of the necrotic tissue can improve prognosis.¹ Studies have found that mortality varies between 50%⁸ and 73%.¹ Survival is likely to depend on concurrent risk factors (such as diabetes or trauma compared with haematological malignancies) which, if corrected, improve outcome.⁸ It is also dependent on the form of disease; pulmonary mucormycosis has been found to have 65% mortality while disseminated mucormycosis has 96%.⁴ Prognosis in diabetic patients is poor.¹ Survival for diabetic patients with rhino-orbital-cerebral mucormycosis is 40–68% when treated with amphotericin B with or without surgery.⁸

Amphotericin B lipid complex is the gold standard in terms of antifungal treatment.¹ Other adjunctive treatments are pro-inflammatory cytokines, such as interferon-gamma and granulocyte macrophage colony-stimulating factor, which enhance the ability of granulocytes to damage the agents of mucormycosis.⁹ Hyperbaric oxygen has been found to have fungistatic and angiogenic effects which improve vascularisation and increase the efficacy of amphotericin B.¹⁰

Treatment should continue until clinical signs and symptoms resolve, radiographic signs of disease have resolved, or residual signs have stabilised on serial imaging, and the patient is no longer immunosuppressed or has recovered from neutropenia.

Our case is worthy of discussion as lingual mucormycosis and lingual necrosis are unusual. The source of infection cannot be found with certainty, but we surmise that it was initially inhaled, which caused the pulmonary mucormycosis.

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