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Case Report
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Palatal Mucormycosis in an Immunocompetent Infant

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Abstract

Mucormycosis in an immunocompetent patient is a rarity. Further, the involvement of palate by the said infection is not a common occurrence. In light of these facts; we present a rare case of palatal mucormycosis in an immunocompetent infant of three months. To our knowledge, such a case has not been previously reported in medical literature.

Keywords: Mucormycosis; Infant; Palate

Introduction

Mucormycosis is a rare opportunistic fungal infection [1-3]. It is usually seen in immunocompromised adults suffering from metabolic disorders like diabetic ketoacidosis and uremia, leukemia or patients on immunosuppressive drugs such as steroids, cytotoxics and antibiotics [1,3-5]. It is caused by a saprophytic fungus that occurs in soil or as a mould on decaying food. Etiopathogenesis of mucormycosis is characterized by angioinvasion, thrombosis, infarction and necrosis of the tissue, at times even leading to death [1,2-4]. Its five major clinical forms are: Rhinocerebral, pulmonary, gastrointestinal, cutaneous and disseminated [1,3,6]. It is diagnosed histopathologically and treated by reversing the source of immune compromise, surgical debridement and systemic antifungal drugs like Amphotericin B [1-3,6].

Mucormycosis in an immunocompetent patient is an extreme rarity [2,3]. Further, the involvement of palate in mucormycosis has also been seldom reported [4,6]. With this background we report one such rare case of palatal mucormycosis in an immunocompetent infant, hitherto unreported in medical literature.

Case Presentation

A three-month-old child reported to the pediatric outpatient department of our tertiary care teaching hospital with the chief complaints of failure to accept maternal feed and nasal regurgitation for the past five days. Further examination revealed a palatal perforation and the case was referred to ENT Department. Detailed clinical evaluation revealed that the lesion was initially a small ulcer which within five days developed into a destructive palatal perforation. There was no history of sinusitis or trauma. The palatal lesion primarily involved the hard palate and the adjoining part of the soft palate (Figure 1). The margins were well defined with no necrotic debris or foul odour. The lesion was about 2.5 cm × 2 cm in its greatest dimensions. The nasopharynx and adenoid tissue were clearly visible through the perforation. The lateral and medial walls of the nasal cavity were normal. A differential diagnosis of granulomatous diseases (like TB and syphilis), midline lethal granuloma and T-cell lymphoma were made. Subsequent PAS staining of the biopsy from the lesion revealed: non-septate thin walled hypha branching at irregular angle and interval consistent with mucormycosis (Figure 2). Hence, the final diagnosis of Palatal mucormycosis was made. It would be prudent to note that the child had no other debilitating disease and rests of the investigations including a CT scan were found to be normal.

The child was given regular feeds by nasogastric feeding tube. The patient was treated with liposomal Amphotericin B in requisite pediatric dosages with all necessary precautions for three weeks. A repeat biopsy from the lesion at the end of 3 weeks was found to be normal. The patient was subsequently discharged with a dental obturator and was kept in regular follow-up for 6 months with no untoward incident to report.

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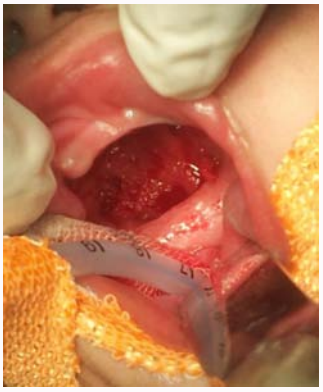

Figure 1: Clinical photograph of the infant showing palatal perforation.

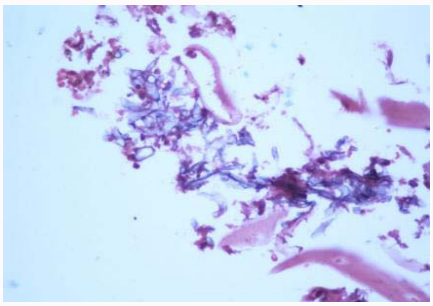

Figure 2: PAS stained section - non-septate thin walled hyphae branching at irregular angle and interval consistent with mucormycosis.

Discussion

Mucormycosis in immunocompetent patients is an intriguing subject. Review of limited literature on the cited subject suggests that in an immunocompetent patient, nose and maxillary sinus are predominant source of infection (sporangia greater than 10 μm get lodged here) [3,7]. After colonizing the nose and sinus, the fungi proliferate and then disseminate to the central nervous system giving a rhino-orbital form or disseminated form. However, it would be important to note that the mucosal/cutaneous epithelium and endothelium are effective barriers to the pathogenicity of the fungus in an immunocompetent person, thus the damage to this barrier by previous infection, trauma and cytotoxic drugs also plays an important role [8]. Moreover, the sporangiospores secrete toxins and proteases to destroy this epithelial barrier [8]. We can all but speculate the etiopathogenesis in our case where the child was an infant with no signs and symptoms of sinusitis or trauma. An entity known as indolent mucormycosis in an immunocompetent patient does find mention in literature, but the age of 3 months in our patient rules that out [2,9]. However, in the context of this case it would be imperative to note that India has recorded the largest number of cases with mucormycosis in immunocompetent patients (44.3%) [3]. This may be due to climatic conditions, malnutrition, poor socio-economic and hygiene status [10].

It would also be pertinent to note that mucormycosis rarely invades the palate. Hard palate mucormycosis in adults is usually an ominous sign of aggressive mucormycosis and even death has been recorded in such cases [4]. It is regarded as a severe manifestation of sino-nasal disease [5,6]. However, in our patient no signs or symptoms of sinonasal infection or orbital infection were observed, yet complete destruction of palate (including hard palate) was observed.

In an internet search using Pubmed/Medline services, author could find only one other case of palatal mucormycosis in an infant which was a manifestation of sinonasal infection requiring surgical debridement of maxilla with voriconazole treatment [1]. In contrast, our case was an immunocompetent child who responded well to Amphotericin B treatment and did not require surgical intervention. The principle of surgical management is to debride until normal bleeding tissue is encountered [6]. Since bleeding was encountered while taking biopsy from the edges of the perforation, further debridement was not undertaken. Also imaging studies showed no disease in the relevant anatomical area.

To summarize, this case merits mention on account of: i) Rarity of the lesion in infants and atypical site ii) treatment of mucormycosis by Amphotericin B only iii) marked paucity of literature on the subject of "Mucormycosis in immunocompetent pediatric patients" in English medical literature, which limits conclusions to be drawn on the clinical course, management and prognosis of the disease.

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