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Case Report

A rare case of maxillary osteomyelitis caused by *Curvularia* species



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ABSTRACT

Orofacial fungal infections display different etiologies, pathogenesis, and clinical presentations. The incidence of rare mycoses of oral cavity is very low. We report a case of chronic invasive sinusitis caused by *Curvularia* infection which presented as osteomyelitis of maxilla. Intraoral manifestation of *Curvularia* infection is seldom reported in the literature. Most cases of fungal infection in immunocompetent patients present in a noninvasive fashion as an allergic fungal sinusitis. Sinusitis due to unusual fungal pathogens, like *Curvularia*, is thought to occur primarily in immunocompromised individuals and poorly controlled diabetics. Invasive fungal sinus disease has a very high morbidity and mortality. For optimal outcome, rapid diagnosis and reversal of the underlying condition is critical to success.

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1. Introduction

Dematiaceous or darkly pigmented fungi comprise large, heterogeneous group of organisms that have been associated with a variety of clinical syndromes. In recent years, these fungi have been increasingly recognized as important pathogens, and the spectrum of diseases with which they are associated has also broadened. *Curvularia* infections in humans are relatively uncommon despite the ubiquitous presence of this soil-dwelling dematiaceous fungus in the environment. Most cases of sinusitis caused by *Curvularia* are primarily allergic in nature. Involvement of this fungus in invasive sinusitis is quite rare and intraoral involvement is seldom. Diagnosis depends on a high degree of clinical suspicion and appropriate pathological and mycological examination of clinical specimens. Itraconazole and voriconazole demonstrate the most consistent in vitro activity against this group of fungi [1]. We report a case of chronic invasive sinusitis caused by *Curvularia* infection which presented as osteomyelitis of maxilla.

2. Case report

A 77-year-old male reported to the Department of Oral Medicine and Radiology with a complaint of halitosis for a duration of 15 days. The patient reported a history of recurring rhinosinusitis over the preceding several years which had worsened 9 months earlier when he experienced orofacial pain mainly localized to right maxilla associated with a rapidly progressing swelling of the right side of the face. He also reported nasal congestion, posterior nasal discharge and progressive fatigue. The patient had consulted an ENT surgeon and multiple courses of antibiotics had been prescribed. Relief in pain was noticed but the swelling persisted, and therefore, the patient underwent endoscopic sinus surgery following 6 months of medications. Following the sinus surgery, patient was asymptomatic for 2 months, when finally he had approached us with complaints of halitosis.

There was congenital and familial history of hearing loss and aphasia. The patient also reported a past medical history of diabetes mellitus since 20 years. The patient was earlier taking glimepiride 3 mg twice daily before food and metformin 500 mg thrice daily after food, but due to poor glycemic control the patient had been upgraded to insulin by his primary care physician 6 months previously. The patient had been prescribed human mixtard insulin (30:70) administered through subcutaneous route. Presently the patient was taking 18 IU of insulin half an hour before breakfast and 14 IU of insulin half an hour before dinner

* Asian AOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

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along with metformin tablet 500 mg orally twice a day. A history of hypertension since 12 years was also noted.

At the time of presentation, the patient was afebrile, his vital signs were within normal limits. Extraoral examination revealed minimal tenderness over right maxilla. Intraoral examination showed exposure of almost the entire right maxillary alveolus, with necrotic bone which was yellowish green in color, foul smelling and could be broken off easily in small pieces on slightest manipulation (Fig. 1). There was purulent drainage from the wound margins on palpation. The palatal tissues were edematous. The patient gave history of extraction of the teeth in the involved segment 2 years earlier.

Conventional radiographs – orthopantomograph and paranasal sinus view (Fig. 2) demonstrated irregular bony texture of right maxillary alveolus and opacification of right maxillary sinus respectively. An evaluation of computed tomography scan (Fig. 3) showed an irregular moth-eaten cortical destruction of right maxilla, extending into the medullary cavity of the body of maxilla, zygomatic and the alveolar process of the right maxilla and the pterygoid plates. Associated circumferential mucosal thickening of right maxillary sinus was eminent. A soft tissue swelling arising from hard palate and adjacent maxillary alveolus could be appreciated on the right side, measuring 20 mm × 10 mm. There was evidence of focal surgical defect in the antero-lateral wall and floor of the maxillary sinus. Right uncinate process was absent and the nasal septum was deviated to the right with the presence of septal spur.



Fig. 1. Intraoral clinical photograph of the patient showing exposed necrotic bone in the right maxillary alveolus with edema of palatal tissues shown by the arrow.



Fig. 2. Paranasal sinus view radiograph showing opacification and sclerosis of the posterolateral wall of right maxillary sinus, suggestive of chronic sinusitis.



Fig. 3. Computed tomography scan showing irregular moth eaten cortical destruction of right maxilla, extending into the medullary cavity of the body of maxilla, zygomatic and the alveolar process of the right maxilla and the pterygoid plates. Associated soft tissue swelling of hard palate and adjacent maxillary alveolus could be appreciated on the right side. There was evidence of focal surgical defect in the antero-lateral wall and floor of the maxillary sinus.

Chest auscultation and electrocardiogram was normal. No masses or lesions were noted on chest radiograph either. Laboratory studies showed serum electrolytes (S. sodium 139 mEq/L, S. potassium 4.1 mEq/L, S. calcium 8.5 mEq/L) blood glucose level

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