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PHYCOMAXILLA- A RARE CASE REPORT

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ABSTRACT

A compromised immune status can allow the growth of opportunistic pathogens to invade the body. Phycomycosis is an invasive fungal disease affecting the structures of the skin, air sinuses, orbit, and the brain. The leading predisposing factors include uncontrolled diabetes mellitus, lymphoma, leukemia, renal failure, organ transplant, long-term intake of corticosteroids, immunosuppressive therapy and AIDS. The signs and symptoms of intraoral phycomycosis are uncertain. We present an ideal case of phycomycosis with classical features involving the maxilla, invading deep into the zygomatic process and the lower border of orbit in a chronic uncontrolled hyperglycemic patient. The fulminant, non-specific nature of the disease makes the diagnosis and management quite challenging. Treatment includes a combination of surgical debridement of the necrotic debris along with administration of systemic antifungal drugs like Amphotericin, Posaconazole.

INTRODUCTION

Fungal infections manifest in humans clinically in a varied fashion ranging from cutaneous lesions to fatal systemic infections. Basidiobolushaptosporus of order Phycomycetes is the causative organism [Kok Han Ng, 1991]. Cutaneous infections gain entry via cut wounds in the skin or the mucosa. Intraorally, the extraction site acts as an entry portal to invade the underlying structures (Kok Han Ng, 1991; Ferstenfeld, 1977). Predisposing factors include diabetes mellitus, renal insufficiency, organ transplantation, chronic use of iron-chelating agents and declined immune status. Common manifestations include nodular, non-tender swellings on the trunk and extremities. Intraorally, the fulminant necrotic tissue may mimic osteomyelitis, carcinoma, mycotic infections. Microbiological picture and microscopic examination with special stains help to conclude the diagnosis. Treatment may involve a multi-disciplinary approach.

Case Report: A 57-year-old male patient reported to our department with complaints of pain and swelling in the upper right back tooth region for the past 3months. The patient had undergone uneventful extraction of his upper posterior teeth 2weeks ago despite which the symptoms did not subside. Medical history revealed the patient was diabetic and under medication for the past 10years. Extra orally, a diffuse swelling was evident on the right side of the face involving the zygomaticomaxillary region, warm, tender and firm in consistency with no evidence of discharge (Figure 1).

Intraorally, obliteration of the buccal vestibule was evident in relation to 14,15,16 region and was soft to firm upon palpation. A healing extraction socket was seen in relation to 16 (Figure 2). The maxillary occlusal radiograph revealed a diffuse radiolucency on the right side showing extensive bone destruction and the opg showed haziness of the maxillary sinus (Figure 3 & 4). CT scan showed maxillary sinus opacification with a large sequestrum on the right side of the maxilla extending up to the midline (Figure 5). Biochemical investigations revealed elevated fasting blood sugar level of 300 mg/dl. Suspecting an underlying chronic necrotic infection, a working diagnosis of chronic suppurative osteomyelitis was given. An incisional biopsy was done under local anaesthesia and sent for further investigation. The microbiological picture revealed the presence of ribbon-like hyphae. Histopathological examination with H and E, periodic acid-Schiff, Gomori'smethenamine silver and Ziehl-Neelsen stain displayed fungal organisms, branching at right angles, with large aseptate hyphae carrying round to ovoid spores (Figure 6). They were PAS-positive and negative to Ziehl-Neelsen staining. Also, the bed of granulation tissue and necrotic debris contained colonies of large hyalinised basophilic aseptate hyphae. Inflammatory cells prevailed in the peripheral zones of the necrotic tissue with no evidence of dysplasia. Correlating the clinical and histopathological findings, the case was diagnosed as Phycomycosis. Under general anaesthesia, hemi-maxillectomy was performed followed by debridement and curettage of the necrotized tissue.



Figure 1. Reveals presence of a diffuse extraoral swelling on the right side of the face, extending from nasolabial fold to pre auricular region medio-laterally and from infra orbital region involving the inner canthus of the eye to the angle of the mandible supero-inferiorly



Figure 2. Shows a healing extraction socket in relation to 16, with no bleeding or pus discharge



Figure 3. Is a maxillary occlusal cross sectional radiograph which reveals a diffuse radiolucency on the right side showing extensive bone destruction along with vertical bone loss in the maxillary anteriors



Figure 4. Reveals haziness in the maxillary sinuses on either side in the panoramic radiograph

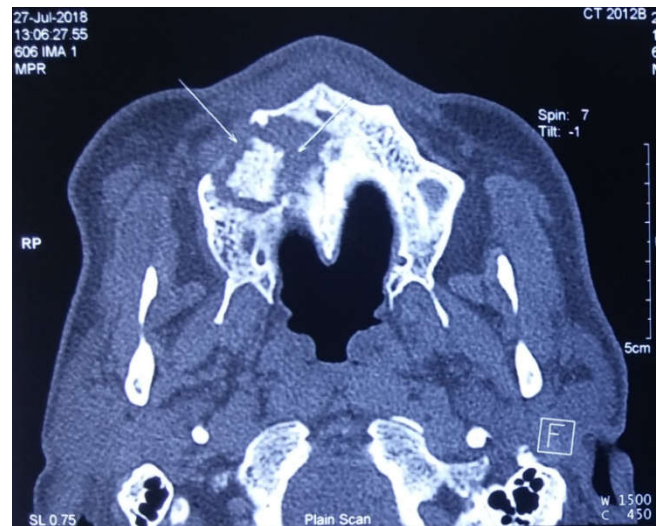


Figure 5. Axial CT shows presence of a large sequestrum on the right side of the maxillary apparatus extending up to the midline

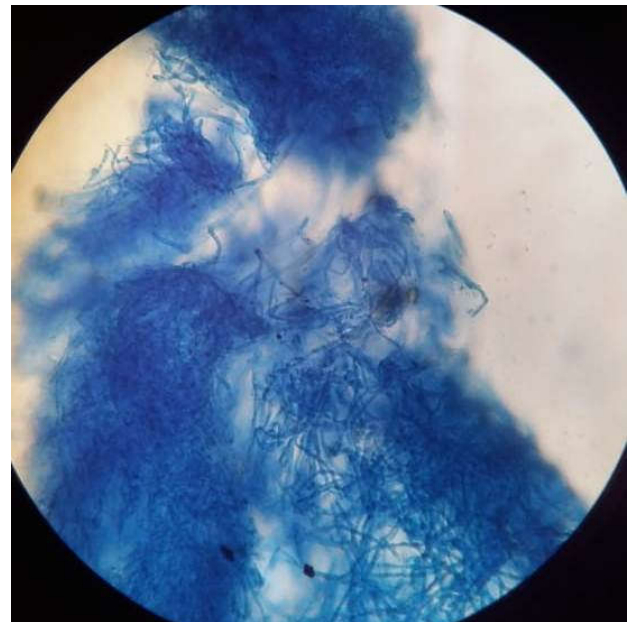


Figure 6. The photomicrograph of the excised necrotic tissue specimen reveals presence of aseptate hyphae of phycomycetes inside the lumen

Under the microscope, the excised tissue concluded the diagnosis as “Chronic osteomyelitis of the maxilla with underlying phycomycosis”. Antifungal chemotherapy consisting of amphotericin B- 50 mg was administered intravenously for one week every day, gradually increasing to 40mg twice a day with continued monitoring of his blood glucose level.

Table 1. Shows the various possible differential diagnosis for intraoral tissue necrosis with its characteristic distinguishing features from phycomycosis

Disease entity	Gender predilection	Age	Most commonly affected jaw	Region	Distinguishing feature
Mucormycosis	No gender predilection	Common in adults	Palate	Extension from maxillary sinus through palate into the mouth	Sloughing ulcers with grey necrotic debris and exposed bone usually the maxilla Unilateral facial pain (8. Anne Cale Jones, 1993).
Osteomyelitis	No gender predilection	From neonates to adults	Mandible	Body of the mandible	Clinical sign: Exposed bone with tissue necrosis Lab evaluation: positive bacterial culture (Aadithya, 2016).
Aspergillosis	No gender predilection	Not specific	Not specific	Paranasal sinuses, nasal cavity, oral mucosa, rarely tongue, soft palate	Periodic acid Schiff stain positive Lesion –yellow or black with necrotic ulcerated base Plaque formation and intense local pain in oral lesions (Aadithya, 1996).
Tuberculosis	Males > Females	Primary form- rare – common in children and adolescents Secondary form- common in middle aged and elderly	Mandible	Alveolus and angle region	Slownecrosis of the bone and formation of a sub-periosteal abscess (lumpy jaw) appearing as a painless, soft swelling. AFB staining positive (Kannaperuman, 2013).
Carcinoma	No gender predilection	Usually adults	Not specific	Any part of the oral cavity	Chronic necrotizing ulcer with everted margins and exposure of underlying bone (Kannaperuman, 2013).

Six days later, his condition was considered stable to allow surgical debridement. The dressings were changed daily, and the wound was irrigated with Hydrogen peroxide.

DISCUSSION

Phycomycosis, an uncommon fungal infection is caused by organisms under the class Phycomycetes initially described by Paltauf. Originally it was referred to as mucormycosis because the infection was caused by the species belonging to the family Mucoraceae [Kok Han Ng, 1991]. Iron plays an important role in the growth of Phycomycetes. Fungal hyphae produce “rhizoferrin,” which binds to iron fervently. This iron - Rhizoferrin complex is then taken up by the fungus and becomes available for its vital functions. Clinically, Phycomycosis presents in six well-known forms, based on its systemic involvement-namely Rhino-orbito-cerebral, Pulmonary, Gastrointestinal, Cutaneous, Disseminated and Miscellaneous (Kok Han Ng, 1991; Abdulaziz A Bakathir, 2006). Rhino-orbito-cerebral or rhinocerebralphycomycosis is the most common form and is prevalent in uncontrolled diabetics, with evidence of approximately 75% reported cases (Ferstenfeld, 1977) .The usual portal of entry is the nasal sinuses, occasionally the orbit. Initial manifestations include nasal congestion and peri-orbital analgesia. The walls of the blood vessels are permeable to fungal inoculation, penetrating the extraction socket to invade the underlying tissues (Ferstenfeld, 1977; Eugene Abramson, 1967). On the other hand, in regular dental practice Pulmonary form is seen in

result of inhalation or by vascular mode. Gastrointestinal Phycomycosis is rare, but it is believed to occur in extremely malnourished children. Cutaneous and dissemination phycomycosis forms are noted in patients with disruption of the normal protective skin, especially in cases of burns. In the present case, the patient had uncontrolled diabetes, which is a well-known predisposing factor. Diabetes mellitus alters the immunological response of the human body to any persistent infection. Increased glucose levels stimulate the growth of fungi thereby reducing chemotaxis and phagocytosis which in turn permits these organisms to thrive in the acidic environment [McNulty, 1982; Cwroll, 1969].

Table 1 shows the various possible differential diagnosis for intraoral tissue necrosis with its characteristic distinguishing features from phycomycosis. The treatment of Phycomycosis is thus based on three vital measures namely, control of predisposing factors, antifungal therapy and surgical intervention. In the present case, the patient was monitored for a controlled level of diabetes mellitus by his physician, followed by antifungal therapy which includes amphotericin B and surgical intervention of the affected maxilla. Phycomycosis is an aggressive and potentially fatal disease in immunosuppressed patients usually manifesting cutaneously. Intraoral presentation of the lesion is uncommon. Careful clinical, radiographical examination, along with histopathological confirmation supported by special stains, is the key to diagnosis of phycomycosis, leading to early and prompt treatment.

REFERENCES

- Aadithya B Urs, Hanspal Singh, Sujata Mohanty and Pankaj Sharma. 2016. Fungal osteomyelitis of maxillofacial bones: Rare presentation. *Journal of oral and maxillofacial pathology.*, 20,3: 546.
- Abdulaziz A Bakathir. 2006. Mucormycosis of the jaw after dental extractions: Two case reports. *Sultan Qaboos Univ Med J.*, 6(2): 77–82.
- Anne Cale Jones, Teresa Youngblood Bentsen, Paul D. Freedman. 1993. Mucormycosis of the oral cavity. *Oral Surg Oral Med Oral Pathol Oral Radilo Endod.*, 75:455–460.
- Cwroll G. Taylor, Roger E. Alexander, Lieutenad C'onznaamler, Willium H. Green, Lieutewuntand Howard S. Kraner. 1969. Mucormycosis (Phycomycosis) involving the maxilla. *Oral Surgery, Oral Medicine, Oral Pathology.* 27(6): 806-822.
- Eugene Abramson, Dana Wilson, Ronald A. 1967. Arky. Rhinocerebral phycomycosis in association with diabetic ketoacidosis: report of two cases and a review of clinical and experimental experience with amphotericin B therapy. *Annals of Internal Medicine.* 66, 735.
- Ferstenfeld JE., Cohen SH., Rose HD., Rytel MW. 1977. Chronic rhinocerebral phycomycosis in association with diabetes. *Postgraduate Medical Journal.* 53:337-342.
- Kannaperuman J., Gowri Natarajarathinam, Anusha V. 2013. Rao and Sudha Palanimuthu. 2013. Primary tuberculous osteomyelitis of the mandible: A rare case report. *Dental research journal.*, 10,2: 283-6.
- Kok Han NG., Chew Song Chin, Ronald Davis Jalleh, Chong HuatSiar, Chon Hee Ngui, Singaram SP. Nasofacial Zygomycosis. 1991. *Oral Surgery, Oral Medicine, Oral Pathology.* 72(6): 685-688.
- McNulty JS. 1982. Rhinocerebral Mucormycosis: Predisposing factors. *Laryngoscope.*, 92:1140-1144.
- Stavros Tryfon, Ioannis Stanopoulos, Elias Kakavelas Anastasia Nikolaidou Ioannis Kioumis. 2002. Rhinocerebral Mucormycosis in a Patient with latent diabetes mellitus: A Case Report. *J Oral Maxillofac Surg.*, 60:328–330.
