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Case Report

Diagnostic Challenges Associated with Osteomyelitis of Maxilla: A Surgical Approach

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Abstract

This case report presents a 59 year old male patient who reported to the dental OP with a complaint of pain and swelling in the right side of face for the past one month. History reveals that he underwent extraction of his teeth 6 months back. Followed by the extraction he experienced pain, foul smell and pus discharge from the extraction site. On examination tenderness was elicited over the right side of face, right maxillary vestibule and maxillary alveolus with exposed necrotic right alveolar bone. Radiographic findings revealed radio opaque areas involving right maxillary alveolus along the lateral nasal wall extending up to infraorbital rim. Based on the clinical examination and extent of invasion through computed tomography the case was diagnosed to be mucormycosis of right maxilla. However, histopathological examination confirmed it to be osteomyelitis of right maxilla. Surgical exploration with marginal resection of right maxilla followed by primary closure was planned as the treatment.

Keywords: *Diabetes Mellitus; Osteomyelitis; Mucormycosis*

Introduction

Osteomyelitis is the inflammation of bone marrow and its contents, haversian system and medullary cavity. Osteomyelitis commonly affects the mandible and a rare occurrence is seen in maxilla which is due to the collateral blood flow in midface, porous nature of membranous bone of maxilla, thin cortical plates and abundant medullary spaces [1]. Bacterial infection of odontogenic origin is considered to be the prime etiological factor that leads to complication after dental extractions and surgeries [2]. Primary stages of osteomyelitis can be prevented at an early stage with appropriate antibiotics, while secondary osteomyelitis may create serious consequences that originate from the primary site which eventually extends to cranial cavity and brain. Management of osteomyelitis may vary from antibiotic regimen to surgical interventions based on the site and severity of invasion.

Case Presentation

A 59 year old male patient reported to the department of oral and maxillofacial surgery with a complaint of pain and swelling in the right side of his face for the past one month. History reveals that he underwent extraction of his teeth 6 months back and followed by the extraction he experienced pain, foul smell and pus discharge from the extraction site. He is a known diabetic patient for the past 20

years and is on medication for the same. On intraoral examination erythema was seen in the right maxillary vestibule and alveolus. Alveolar bone over the right canine and premolar region was necrosed and exposed (Figure 1). On palpation tenderness was elicited over the maxillary sinus region, lateral wall of the nose and infra orbital region extraorally. Based on the above findings the case was clinically diagnosed as mucormycosis of right maxilla. Computed tomographic imaging showed hypodense areas in the right maxilla involving the right maxillary sinus extending from infra orbital rim to maxillary alveolus and from right palate to zygomatico maxillary buttress extending up to the pterygoid plates (Figure 2 and 3).

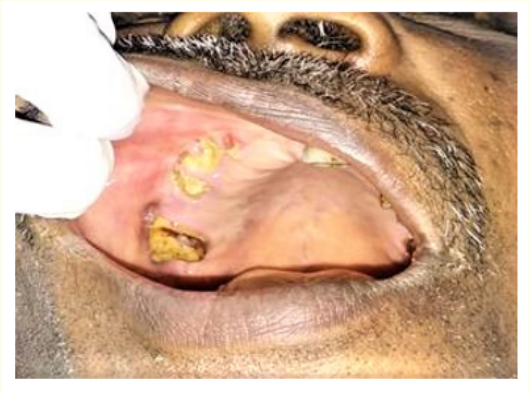

Figure 1: *Exposed necrotic alveolar bone in right side maxilla.*

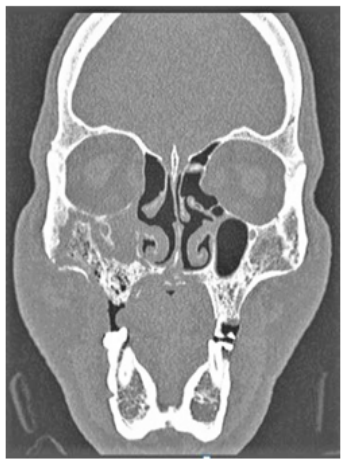

Figure 2: *CT coronal section shows hypodense lesion in the right maxilla involving the entire maxillary sinus extending superoinferiorly from infraorbital rim till alveolar bone and mediolaterally from palate to zygomaticomaxillary buttress. Cortical bone loss involving dentoalveolar region, palate, medial and superior wall of sinus.*

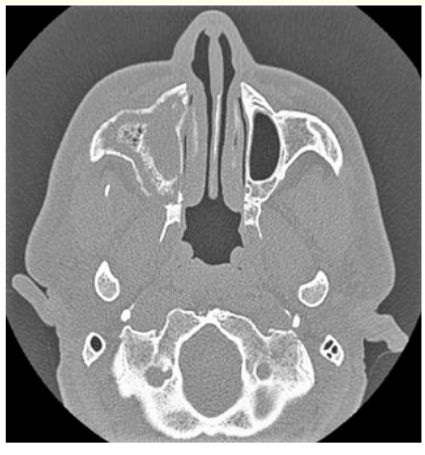

Figure 3: *Axial section showing anteroposterior extension from right anterior wall of maxilla till pterygoid plates.*

Surgical procedure

Treatment was planned for surgical exploration with marginal resection of right maxilla followed by primary closure. Under Orotracheal intubation general anesthesia was administered. Standard painting and draping was done. Markings were placed for Weber-Fergusson incision through the vermilion border along the filtrum of the lip extending around the base of nose and along the facial nasal groove extending infraorbitally 3 - 4 mm below the cilium to the lateral canthus (Figure 4).

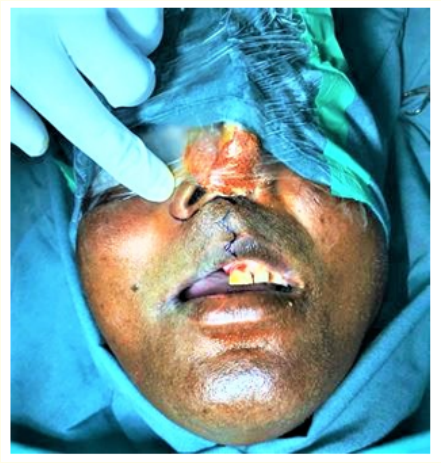

Figure 4: *Markings for weber-fergusson incision.*

Local anesthesia was infiltrated along the areas to be incised. The incision was made through the skin, subcutaneous tissue along the nose. A full thickness upper lip was transected and the labial artery was ligated. Further dissection was carried out along the mucobuccal

fold upto the maxillary tuberosity. The subciliary component was not accessed. The cheek flap was elevated off from the maxilla in a sub periosteal plane (Figure 5).

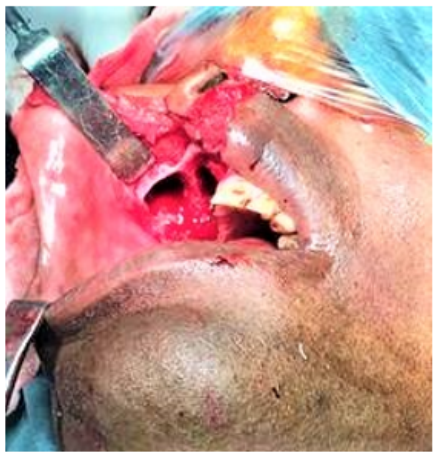

Figure 5: *Full thickness flap was reflected from the right maxilla with exposure of maxillary sinus. The floor of the sinus is completely lost.*

Areas of necrotic bone were surgically explored and marginal resection of right maxilla was performed. The specimen was subjected for histopathological investigation (Figure 6). Intra oral betadine and saline irrigation was done. Closure was done in layers. Subcutaneous suturing was done with 5-0 vicryl. The vermilion border was placed at its anatomical position and secured with 5-0 vicyl and skin suturing was done using 3-0 ethicon. Betadine ointment was applied over the sutures. The patient was extubated and shifted to ICU uneventfully. Postoperative medications were prescribed.

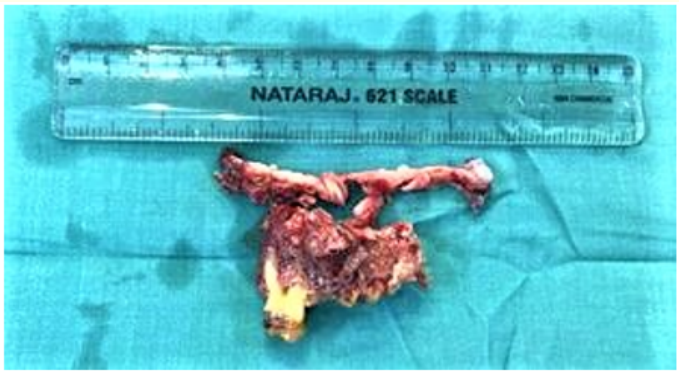

Figure 6: *Hard and soft tissue specimen obtained after marginal resection of right maxilla.*

Macroscopically, multiple [15] hard and soft tissue specimens were obtained which was brownish back in colour. Histologically, the hematoxylin and eosin stained hard tissue section showed multiple irregular shaped necrotic bony trabecular with empty lacunae and

certain region showed osteocytes, large osteolytic bay without osteoblastic rimming separated by a network of fibrous connective tissue containing numerous disseminated RBC's. The given soft tissue section showed a mixture of relatively normal and hypoplastic epithelium, fibrous connective tissue with dense infiltration of chronic inflammatory cells with moderate vascularity (Figure 7). Evidence of new bone formation and fragments of bony trabeculae were evident. The above histopathological investigation confirmed the case to be osteomyelitis.

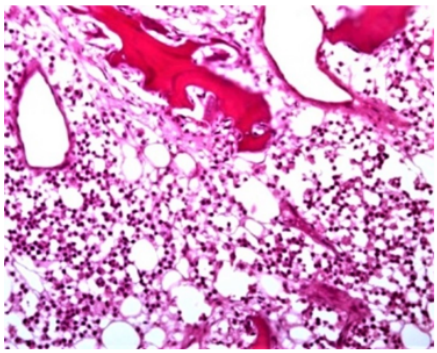

Figure 7: *Multiple irregular shaped necrotic bony trabecular with empty lacunae, large osteolytic bay separated by fibrous connective tissue with dense infiltration of chronic inflammatory cells.*

Discussion

Our case experienced multiple diagnostic challenges to arrive at a provisional diagnosis since the patient reported to us with clinical findings mimicking certain rare entities affecting the maxilla.

Osteomyelitis was first described in 1852 by a French surgeon Edouard Chassaignac [1]. The hallmarks of osteomyelitis are progressive bone destruction and sequestrum formation [3]. Osteomyelitis has various etiological factors such as bacterial infection, vascular deficiency, auto immune diseases [4], trauma to maxillofacial region through domestic violence, sports injury and motor vehicle accidents [5] which initiates inflammation, hyperemia and infiltration of granulocytes due to increased capillary permeability ultimately leading to necrosis of the tissue, vascular thrombosis which over a period of time progresses towards formation of pus. The accumulation of pus gradually increases the intramedullary pressure resulting in vascular collapse, venous stasis and ischemia which eventually accumulates beneath the periosteum resulting in sequestrum formation. Ischemia at the involved site increases the level of carbon dioxide which in turn attracts more calcium causing increase in mineralization of the sequestrum due to change in the level of pH.

The clinical findings of our case was typically in correlation with the findings of mucormycotic infection which showed a positive clinical correlation in a study by Manpreet., *et al.* [3], where she experienced a similar clinical dilemma while diagnosing actinomycosis (fungal infection). Ramesh., *et al.* [6] emphasized that extracted site serves as a nidus for mucor infection particularly in an immunocompromised individual (uncontrolled diabetes mellitus) which alters the immune response of the body primarily causing osteomyelitis with slow progression to mucormycosis. Though the present clinical findings remain questionable, culture and histopathological investigations provide a confirmatory diagnosis.

Chermetz., *et al.* [7] in his study reported that, there was simultaneous presence of both aspergillus as well as mucormycotic infection in 51 patients which was confirmed through histopathological examination. 9 cases showed involvement of oral cavity, maxillary sinus

and orbit while 42 cases had systemic involvement of skin and lungs. Shetty., *et al.* [8] reported a case of non-healing socket where there was exposure of necrotic bone in left maxilla which was clinically diagnosed as osteomyelitis. On further investigation, cytological smear of left maxilla showed a presence of hyphae suggestive of candidiasis. Kumar., *et al.* [9] emphasized that colonization of fungal infection was found to be higher in the oral cavity of diabetic patients than in non-diabetic individuals.

Of various causative bacterial and fungal organisms in osteomyelitis, Asif., *et al.* [10] reported a case of maxillary osteomyelitis caused by *Raoultella planticola* which is a gram negative, aerobic bacillus. Bone biopsy revealed a rare presence of diffuse large B-cell lymphoma (DLBCL). It was described that DLBCL is the most common subtype of non-hodgkins lymphoma affecting oral cavity in 0.1% of cases with maxilla being the most predominant site of occurrence. Upto 40% of non-hodgkins lymphoma show extra nodal involvement, 5% show bone involvement and 2 - 3% show oral cavity and jaw involvement. Treatment includes chemotherapy with or without radiotherapy depending on the type of staging.

Management of osteomyelitis involves treating the underlying systemic condition (diabetes mellitus) [8]. Non- surgical management includes intravenous infusion of antibiotics for 1 week followed by oral antibiotics for a period of 3 weeks [11]. Kim., *et al.* [12] achieved 94.9% of successful treatment outcome with intravenous infusion of augmentin or cefazolin for 2 weeks followed by oral augmentin or roxithromycin for 6 weeks. Fungal osteomyelitis are treated with administration of amphotericin B, voriconazole, posaconazole [3,5,6,13]. Newer antibiotics include Linezolid and tigecycline that showed promising results of 90% of success rates [11]. Surgical management includes debridement of the necrotic site, decortication, sequestrectomy and saucerisation, surgical alveolectomy and hemimaxillectomy in cases with extensive involvement [11,14]. Rehabilitation includes placement of obturator, reconstruction of soft and hard tissues [1,15] and a recurrence rate of 20% is generally observed [1].

Conclusion

Maxillary osteomyelitis is a rare distinct entity which creates a dilemma for the oral surgeons and pathologists in arriving at a diagnosis however correlation of clinical findings with the culture reports and histopathological investigation would provide a confirmatory diagnosis. Early clinical diagnosis and timely intervention may minimize the future complications with periodic and long term follow up.

Conflict of Interest

The authors declare that they have no conflict of interest.

Declaration of Patient Consent

The authors certify that they have obtained written informed patient consent for the surgery under general anesthesia and has given his consent for images and other clinical information to be reported in the journal with an understanding that names and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed. ORCID ID: 0000-0002-9288-6448.

Bibliography

1. Mohite AS., *et al*. "An unusual case of maxillary osteomyelitis in a young female". *Journal of Indian Academy of Oral Medicine and Radiology* 29.2 (2017): 141-144.

2. Muraleedharan A. "An etiological dilemma in osteomyelitis: A case report". *Journal of Indian Academy of Oral Medicine and Radiology* 27.3 (2005): 500-503.

3. Manpreet S., *et al*. "Chronic suppurative osteomyelitis of maxilla mimicking actinimycotic osteomyelitis: A rare case report". *National Journal of Maxillofacial Surgery* 1.2 (2010): 153-156.

4. Patel V., *et al*. "Osteomyelitis presenting in two patients: a challenging disease to manage". *British Dental Journal* 209.8 (2010): 393-396.

5. Oliveira TI. "Maxillary osteomyelitis caused by domestic violence: A diagnostic challenge". *Case Reports in Dentistry* (2014): 930169.

6. Ramesh G., *et al*. "Osteomyelitis of left Maxillary Region due to Mucor Infection". *Rama University Journal of Dental Sciences* 3.4 (2016): 32-36.

7. Chermetz M., *et al*. "Combined orofacial aspergillosis and mucormycosis: Fatal complication of a recurrent paediatric glioma-case report and review of literature". *Mycopathologia* 181.9-10 (2016): 723-733.

8. Shetty L., *et al*. "Maxillary Osteomyelitis with Candidiasis due to Extraction in Uncontrolled Diabetes State-A Case Report". *Dentistry* 5 (2015): 279.

9. Kumar BV., *et al*. "Prevalence of candida in the oral cavity of diabetic subjects". *Journal of the Association of Physicians of India* 53 (2005): 599-602.

10. Asif S., *et al*. "Maxillary osteomyelitis with an incidental diagnosis of maxillary diffuse large B-cell lymphoma: A case report". *Cureus* 11.7 (2019): e5238.

11. Gudmundsson T., *et al*. "Diagnosis and Treatment of Osteomyelitis of the Jaw – A Systematic Review (2002-2015) of the Literature". *Journal of Dentistry and Oral Disorders* 3.4 (2017): 1066.

12. Kim SG., *et al*. "Treatment of chronic osteomyelitis in Korea". *Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology* 92.4 (2001): 394-398.

13. Selvamani M., *et al*. "Mucormycosis causing maxillary osteomyelitis". *Journal of Natural Science, Biology and Medicine* 6.2 (2015): 456-459.

14. Pattnaik B., *et al*. "Osteomyelitis of Maxilla: A rarity". *Annals of International Medical and Dental Research* 3.3 (2017): DE07-DE10.

15. Srivastava A., *et al*. "Maxillary fungal osteomyelitis: A review of literature and report of a rare case". *Annals of Maxillofacial Surgery* 9.1 (2019): 168-173.

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