

## CASE REPORT STUDY

### Rare Case Of Maxillary Osteomyelitis: A Confluence Of Sinusitis, Dental Caries, Covid-19, and Diabetes

#### ABSTRACT

In the realm of maxillofacial medicine, osteomyelitis, a disorder marked by inflammation of the bone and bone marrow, presents serious challenges. Even though the medical community is aware of this disorder, it is relatively prevalent, especially in developing nations like India, to occur in the maxillofacial skeleton, primarily the mandible. Since the maxilla has a large collateral blood flow, thin cortical bones, and strutted bone marrow that reduces infection risk, it tends to occur more frequently in the mandible, than the maxilla. Owing to the rarity of maxillary osteomyelitis, a thorough study into the illness's potential underlying causes is necessary for a definite diagnosis. A severe infection usually requires an immunocompromised state, even in those impacted by these potential causes. Antimicrobial therapy and debridement, along with the management of ensuing dead space and bone stability, are among the susceptibilities and treatments. Here, we describe a rare instance of maxillary osteomyelitis in an older patient with a weakened immune system, highlighting our approach.

**Keywords:** Osteomyelitis, maxillary infection, immunocompromised state, debridement, sequestrectomy.

#### INTRODUCTION

Osteomyelitis is an inflammatory bone disease that starts as a haversian system related infection in the medullary cavity and spreads to the periosteum. Even so, the incidence of maxillofacial osteomyelitis decreased following the advent of antibiotic therapy.<sup>1</sup> Numerous systemic illnesses, such as HIV, diabetes, malnourishment, and chemotherapy drugs that impair the patient's immune system and exacerbate

osteomyelitis, are also present. It typically manifests in the fifth and sixth decade of life, and patients who visit a tertiary hospital always describe problems and late stages. In India, 45.1% of the rural diabetic population has cases of maxillary osteomyelitis being reported. Due to nonanastomoses of the inferior alveolar artery and dense cortical plates that restrict pus drainage through sinus development and infection buildup, the mandible is the most frequently affected region. Because of the maxilla's large blood circulation, thin cortex, and lack of medullary tissues, it rarely affects them.<sup>2</sup> The mandible's body, symphysis, angle, ramus, and condyle are the areas that are impacted in decreasing order of frequency. On the other hand, if the illness affects the maxilla, it could result in dangerous outcomes such as brain and cranial cavity infections. It is vital to identify maxillary osteomyelitis and begin urgent treatment to prevent that.<sup>3</sup>

In 1852, Edouard Chassaignac, a French physician, provided the first description of osteomyelitis. Rees reported maxillary osteomyelitis in 1847. In 1864, John Hunter created the phrases sequestra and involucrum.<sup>1-3</sup> Owing to the rarity of maxillary osteomyelitis, a thorough study into the illness's potential underlying causes is necessary for a definite diagnosis. A severe infection usually requires an immunocompromised state, even in those impacted by these potential causes. To ensure proper treatment and prevent life-threatening consequences, early detection is essential. The presence of "sequestra," or dead bone, and "involucrum," or new bone, are common radiologic findings. In cases with unusual presentations, higher imaging modalities like cone beam computed tomography scan (CBCT) may be used to confirm the existence and extent of the lesion.<sup>4</sup>

The three main etiologies of maxillary osteomyelitis are traumatic, rhinogenic, and odontogenic (as stated by Macbeth in 1952). Sequestrectomy, debridement of the necrosed bone, and extraction of the affected teeth together with antibiotic therapy and adjuvant therapy are the methods used to treat this ailment.<sup>4-5</sup> We hereby report a rare case of maxillary osteomyelitis in an immunocompromised elderly patient and emphasizing on how did we tackle it.

## **CASE REPORT**

A 57-year-old man was referred to the Department of Oral and Maxillofacial Surgery, with the chief complaints of fluid coming out of his left nostril and pain in the upper left rear area of his jaw. He had been exhibiting these symptoms for the previous six months, along with a watery discharge in his mouth and a white, foul-smelling

substance. The patient's nasal regurgitation ceased when he had a tooth extracted from the upper left side of his jaw at a hospital located nearby. For the year prior and a half, he had been confirmed to have hypertension and diabetes, for which he was on irregular oral medications. He also reported having a two-year history of dust allergies, and a year ago, had contracted COVID-19, which required three days of oxygen and steroid medication, under a hospital stay. Additionally, the patient had surgery for sinusitis eighteen months ago.

Upon inspection, there was a mild diffuse swelling in the upper left cheek area, which was tender in nature on palpation. A 1 cm by 1 cm patch of yellowish necrotic bone covered in slough was seen intraorally in the 26 region. From the mid-palatal raphe to the associated gingiva, a palatal swelling was observed extending from the 23 to 28 region in an anterior-posterior orientation. When the palatal swelling was compressed, pus discharge was seen to arise buccally. The water-holding test produced positive findings, and the cold test showed that none of the teeth in the region were vital. Radiographic investigation involved a 3D-CT of the face, which revealed a typical moth eaten appearance.

Crevicular and crestal incisions from the right lateral incisor to the third molar were used to reach the lesion. In addition to the sequestrectomy, the damaged sinus lining was removed. Neosporin-impregnated gauze was used to pack the sinus, and a layer of extracted buccal pad fat was used to establish primary closure. Two days later, the gauze was taken off. After the procedure, the patient was given a palatal obturator and an intravenous steroid, and antibiotics in addition to painkillers for 10 days. The patient was also subjected to oxymetazoline nasal drops for 5 days. A vigilant check on the patient's glycemic levels was done, with him being under a regular oral hypoglycemic regime with strict dietary restrictions. The post-operative period was uneventful. Upon being clinically stable and symptomatically better, the patient was discharged in a satisfactory state with oral antibiotics, emergency analgesics for five days, and a strict routine for oral hypoglycemics and anti-hypertensives. Subsequent follow-ups revealed satisfactory healing without any long-term complications.

## **DISCUSSION**

While rare overall, maxillofacial osteomyelitis is extremely uncommon in the maxilla. Treatment for osteomyelitis should begin as soon as feasible if symptoms are identified early. If treatment is not received, the disease will quickly spread to new locations lowering immunity and the host's defense system.<sup>2</sup> Because the internal

maxillary artery's branches create anastomosing loops that limit widespread involvement, osteomyelitis of the maxilla is uncommon. In immunocompromised people with predisposing conditions, odontogenic infections originating from the pulp are the most common cause of osteomyelitis.<sup>4</sup> According to the literature, sinusitis and tooth infections are the two main causes of maxillofacial osteomyelitis. The frontal bone is typically the most damaged by sinusitis, but the upper jaw is less affected because of inadequate blood flow. In this instance, it is determined that the primary risk factor for maxilla involvement is uncontrolled diabetes. Diabetes is associated with changes in the pattern of blood flow to the jaw and a weakened immune system.<sup>5</sup>

Pain, swelling, and a foul-smelling discharge with sinus development are typical clinical symptoms.<sup>3</sup> Gram-positive and Gram-negative bacteria, such as *Staphylococcus aureus*, *Staphylococcus epidermidis*, *Peptostreptococcus*, *Pneumococci*, *Hemolytic streptococci*, *Escherichia coli*, and *Bacteroides*, are the agents responsible for the microbiological etiology. Mycotic infections, like *Mucormycosis*, can also be the secondary cause.<sup>6</sup> There are two main types of osteomyelitis: suppurative and nonsuppurative.<sup>7</sup> While nonsuppurative osteomyelitis is chronic and has an unclear origin, it does not rule out the presence of the pathogen, suppurative osteomyelitis is primarily caused by odontogenic infections and is defined by the presence of pus, fistula, and sequestrations.<sup>8</sup> As on Mac Beth's classification, there are three types of maxillary osteomyelitis: odontogenic (root sepsis), rhinogenic (spontaneous spread of infection from antrum, and postoperative rhinogenic instances), and traumatic (after surgery or damage).<sup>9</sup> Trauma and hematogenous or contiguous spread may be the cause of the pathogenesis. In children, hematogenous spread is frequently observed, while in adults, post-traumatic spread is frequently noted.<sup>8-9</sup>

The clinical presentation, radiographic characteristics, culture, and histopathologic testing are used to make the diagnosis. Conventional radiography, CT, PET-CT, CBCT, doppler flowmetry, magnetic resonance imaging, and nuclear scans are examples of imaging modalities.<sup>6</sup> In chronic osteomyelitis, radiographic characteristics mostly consist of a "Moth-eaten" look resulting from the widening of Volkman's canals and enlargement of medullary gaps caused by bone lysis and replacement with granulation tissue.<sup>7</sup> A reactive subperiosteal deposition of new bone forms an involucrum when bone is destroyed, creating islands of dead bone known as sequestra. Osteomyelitis is defined histopathologically by necrotic bone, irregular clusters of bony trabeculae, and empty lacunae because of the lack of osteoblastic lining, osteocytes, and chronic inflammatory cells like lymphocytes.<sup>9</sup>

Up to 90% of patients can receive successful treatment for osteomyelitis; however, this depends greatly on the severity of the condition, the kind of interventions used (surgical debridement vs. antibiotics), and the vascular supply to the affected area.<sup>10</sup> Because of the region's architecture and aesthetic concerns, treating osteomyelitis of the head and neck might come with its own set of challenges. Osteomyelitis is usually treated with appropriate antibiotic therapy after surgical debridement of necrotic material and biopsies for the culture of damaged tissue/bone (if the patient is a suitable candidate).<sup>4</sup>

The comparatively avascular and ischemic character of the infected area, together with the low oxygen content and antibiotic resistance of isolated areas, are thought to be the pathophysiology of the disease.<sup>10</sup> The polymorphic white blood cells' bactericidal action is diminished by the decreased oxygen pressure, which restores the former aerobic state. No matter what concentration is used, the dead bone's supply of antibiotics is so sluggish that it is typically impossible to reach the desired location. An inadequate antibiotic concentration at the infection site may arise from therapy recommendations based on serum level monitoring.<sup>11</sup>

Therapeutic interventions can be as basic as various non-invasive techniques or as drastic as more intrusive procedures. Antibiotics, non-steroidal anti-inflammatory medications, hyperbaric oxygen therapy, and bisphosphonate therapy are examples of non-surgical techniques.<sup>4</sup> One of the cornerstones of treatment for the final management of craniofacial osteomyelitis is surgery. Its objectives are to restore fresh blood flow to the affected area, drain the infected area, and remove the sequestrum and other foreign objects.<sup>9</sup> Simple sequestrectomy to segmental resection and reconstruction in patients who refuse to cooperate are among the options. Depending on the kind of osteomyelitis, skin grafting, skin excision, and partial (marginal) resection may also be considered.<sup>12</sup> Regrettably, active treatment might cause a serious recurrence of comorbidities, necessitating additional reconstruction surgery or the use of different types of obturators. Conservative treatment also invariably results in multiple recurrences of the disease. Because maxillary osteomyelitis is so difficult to treat, it presents a challenge to both clinicians and patients. It appears to be challenging even with the major advancements in diagnosis and treatment.<sup>10</sup>

Although various treatment modalities are available to control the infection, care to remove predisposing factors in immunocompromised patients with long-term follow-up is necessary to prevent future recurrence.<sup>12</sup>

## CONCLUSION

Because of its proximity to important structures and the intricate craniofacial anatomy and skeletal system, osteomyelitis of the jaws is a true bone infection that is usually not properly treated. This article outlines the methodical methodology of several investigations to illustrate the importance of diagnosis, with a prompt insight into the management options. Future doctors will definitely benefit from having their young minds stimulated by the prospect of seeing such rarities reflected in the current situation. Remembering that these anomalies still happen, each case needs to be handled with extreme caution.

### **Ethical Approval:**

As per international standards or university standards written ethical approval has been collected and preserved by the author(s).

### **Consent**

As per international standards or university standards, patient(s) written consent has been collected and preserved by the author(s).

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- 3.

## REFERENCES

1. Topazian RG, Goldberg MH, Hupp JR. Oral and maxillofacial infections. (No Title). 2002 Mar.
2. Macbeth R. Osteomyelitis of the maxilla. *The Journal of Laryngology & Otology*. 1952 Jan;66(1):18-28.
3. Koobusch GF, Fotos P, Goll KT. Retrospective assessment of osteomyelitis: etiology, demographics, risk factors, and management in 35 cases. *Oral surgery, oral medicine, oral pathology*. 1992 Aug 1;74(2):149-54.
4. Pattnaik B, Padmavathi BN, Kumari M, Sharma SS. Osteomyelitis of maxilla: A rarity. *Ann Int Med Den Res*. 2017;3:DE07-10.
5. Gupta V, Singh I, Goyal S, Kumar M, Singh A, Dwivedi G. Osteomyelitis of maxilla-a rare presentation: case report and review of literature. *Int J Otorhinolaryngol Head Neck Surg*. 2017 Jul;3(03):771-6.
6. Poonia M, Sidhu SK, Solkhe M, Sihmar SS. Chronic osteomyelitis of maxilla: A rare case report. *J Oral Med Oral Surg Oral Pathol Oral Radiol*. 2016;2:88-90.
7. Habib A, Sivaji N, Ashraf T. Maxillary osteomyelitis: a rare entity. Case reports in otolaryngology. 2016 Aug 25;2016.
8. Spellberg B, Lipsky BA. Systemic antibiotic therapy for chronic osteomyelitis in adults. *Clinical infectious diseases*. 2012 Feb 1;54(3):393-407.
9. Chandra Prasad K, Chandra Prasad S, Mouli N, Agarwal S. Osteomyelitis in the head and neck. *Acta oto-laryngologica*. 2007 Jan 1;127(2):194-205.
10. Manimaran K, Suresh Kannan P, Kannan R. Osteomyelitis of maxilla bilateral involvement: a case report. *JIADS*. 2011 Apr;2(2):57-8.
11. Pincus DJ, Armstrong MB, Thaller SR. Osteomyelitis of the craniofacial skeleton. In *Seminars in plastic surgery* 2009 May (Vol. 23, No. 02, pp. 073-079). © Thieme Medical Publishers.
12. Patel V, Harwood A, McGurk M. Osteomyelitis presenting in two patients: a challenging disease to manage. *British Dental Journal*. 2010 Oct 23;209(8):393-6.



FIGURE 1: Clinical pre-operative details of the patient, depicting the necrotic area in the maxilla.

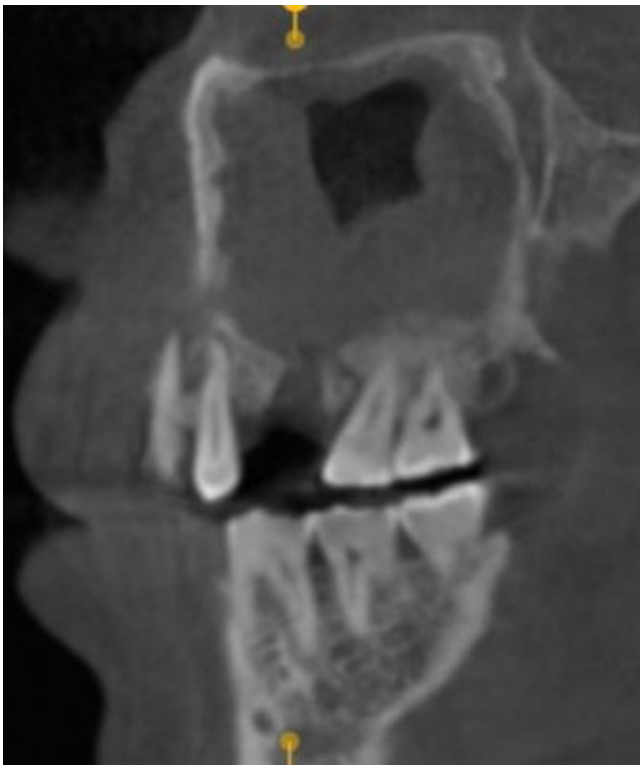


FIGURE 2: A sagittal cut confirming the case of osteomyelitis with a typical moth-eaten appearance.



FIGURE 3: A three-dimensional cut of the jaw explaining the involvement of the bone.

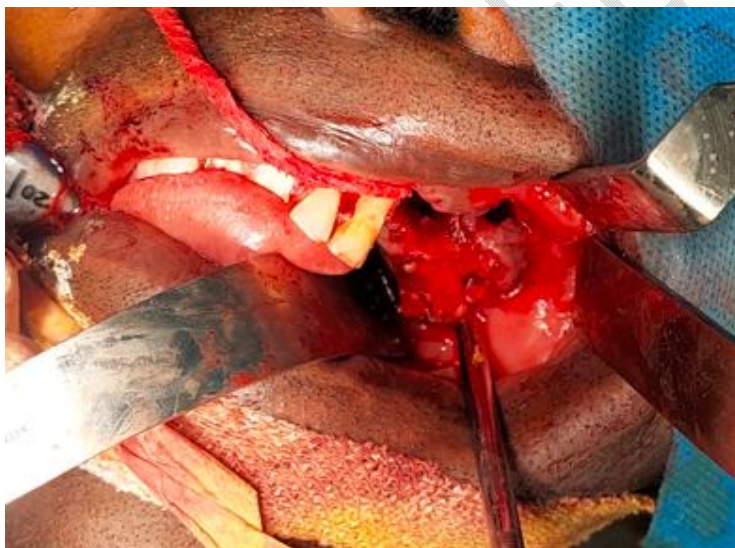


FIGURE 4: An intra-operative view highlighting the sequestrectomy being done, with fresh bleeding being noted.



FIGURE 5: Post-operative healthy healing and good slough being noted.

UNDER PEER REVIEW