ISSN No:-2456-2165

2 Case Reports of Maxillofacial Osteomyelitis with Unique Presentations and Clinical Challenges

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Abstract:- This case series presents two distinct cases of maxillofacial osteomyelitis with unique clinical presentations and diagnostic challenges. Despite variations in age, medical history, and initial symptoms, both cases highlight the importance of early diagnosis, interdisciplinary collaboration, and patient follow-up for optimal management. These cases underscore the complexity of maxillofacial osteomyelitis and its potential impact on adjacent anatomical structures, necessitating thorough evaluation and comprehensive treatment strategies.

Keywords:- *Maxillofacial Osteomyelitis, Interdisciplinary Collaboration, Clinical Presentation, Diagnostic Challenges.*

I. INTRODUCTION

Maxillofacial osteomyelitis is a condition characterized by inflammation in the bones. When it affects the bones in the maxillofacial area, it typically involves both the inner marrow and the outer cortical bone. Therefore, this term is commonly used to describe inflammation in the basal and alveolar bones of the maxillofacial skeleton, it is an inflammatory condition affecting the bone and its marrow within the facial region and presents a myriad of clinical challenges due to its varied manifestations and potential complications(1). This article delves into two distinct cases of maxillofacial osteomyelitis, each characterized by unique clinical presentations and diagnostic complexities. Despite differences in patient age, medical histories, and initial symptoms, these cases underscore the critical importance of early diagnosis, interdisciplinary collaboration, and meticulous patient follow-up for effective management.

Maxillofacial osteomyelitis is multifaceted, and its accurate diagnosis demands a comprehensive approach that combines clinical acumen with advanced radiological imaging techniques. Both cases underscore the significance of orthopantomography (OPG) and cone-beam computed tomography (CBCT) in unravelling the extent and nature of the lesions. The clinical presentations in these cases demonstrate the complex interplay between patient history, systemic factors, and the intricate anatomical structures of the maxillofacial region.

As the cases reveal, the impact of maxillofacial osteomyelitis extends beyond the local site of infection, often involving adjacent anatomical structures such as the maxillary sinus and the skull base. The collaboration of multiple medical disciplines, such as Oral Maxillofacial Surgery and Neurology, becomes pivotal in comprehensively evaluating the disease's scope and devising an effective management strategy.

In this dynamic landscape of maxillofacial osteomyelitis, these cases serve as poignant reminders of the ever-evolving nature of clinical challenges. By sharing these cases and their intricate diagnostic journeys, we contribute to the collective understanding of this condition and emphasize the necessity for continuous research, multidisciplinary collaboration, and tailored patient care. Through these cases, we shed light on the complexities of maxillofacial osteomyelitis and underscore the significance of a holistic and well-coordinated approach to diagnosis and management.

➢ *Case 1*

An 82-year-old male, with a history of prostate cancer and hypertension, presented with a year-long, unrelenting pain in the upper left back tooth region, exacerbated during meals, and sporadically self-relieved. Remarkably, he also complained of recent-onset numbness in the lower left back of the tooth region for the past week. A history of tooth extraction in the same region a year back.

Clinical examination revealed mandibular paresis extending from the chin region to the left mandibular border. Intraorally, inspection revealed denuded necrotic bone in the alveolar region, extending from 36 region to retromolar region (fig 1), and from the 26 region to the 28 region (fig 2) which appeared discolored and devitalized. The surface of the necrotic bone appeared to be rough, porous, and irregular. On palpation, it was non-tender with no pus discharge. On extra oral examination no changes were noted.

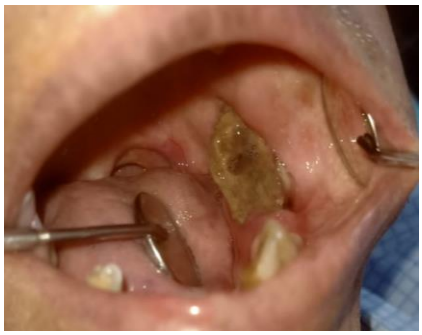

Fig 1 Denuded Necrotic Bone in the Alveolar Region, Extending from 36 Region to Retromolar Region

ISSN No:-2456-2165

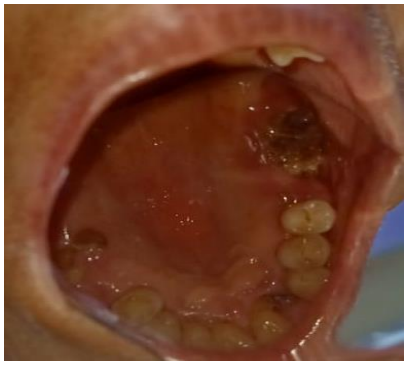

Fig 2 Denuded Necrotic Bone Noted Extending from 26 Region to 28 Region

Based on these clinical observations, the provisional diagnosis indicated the presence of osteomyelitis affecting both the maxilla and mandible. To obtain a clearer visual assessment, the patient was advised to undergo an orthopantomogram (OPG). The OPG imaging divulged a distinctive lesion that combined radiopaque and radiolucent elements. This lesion, measuring 5x3 cm, occupied the left mandibular body, spanning from the 36 region till ramus. Impressively, this lesion extended its reach into the left maxilla extending from 24 to 26 region, while concurrently disrupting the floor of the maxillary sinus. (Fig 3)

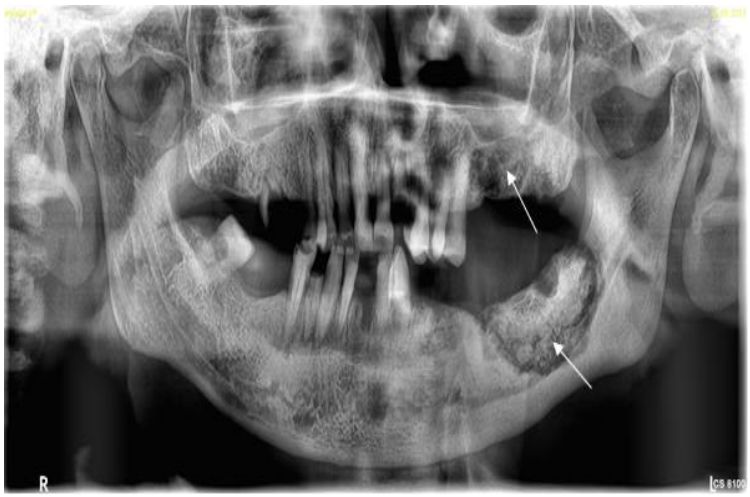

Fig 3 Panoramic Radiograph Reveals Mixed Radiopaque and Radiolucent Lesion on Left Body of Mandible Involving the Inferior Alveolar Nerve and Ill Defined Radiolucency Noted in Left Maxilla Extending from 24 to 26 Region

To further enhance the clarity and precision of the imaging, the patient was subsequently recommended to undergo cone-beam computed tomography (CBCT). This advanced imaging modality solidified the diagnosis of osteomyelitis, which revealed a non-homogeneous mass of tissue is noted extending from the distal aspect of tooth 17 to the retromolar region in the right maxilla, and a similar mixed radiopaque and radiolucent structure is observed on the left maxilla, extending from the distal aspect of tooth 25 to the retromolar region, with mild expansion and disruption of cortical plates. Both lesions involve the buccal and lingual cortical plates. Additionally, a mixed radiopaque and radiolucent structure involves the left mandible and the inferior alveolar nerve. The overall radiographic diagnosis was given as chronic osteomyelitis affecting both the right and left maxilla as well as the left mandible which effectively confirming its presence in both the maxillary and mandibular regions. (Fig 5 and 6)

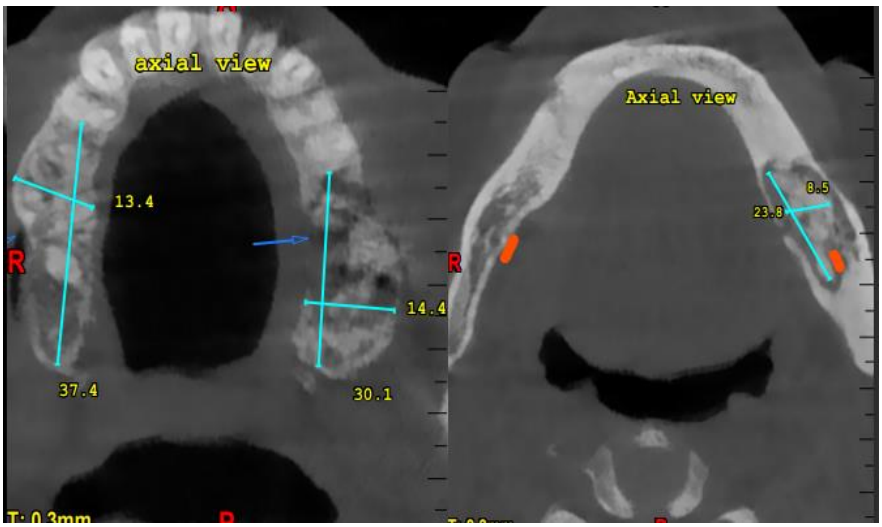

Fig 4 (A) Axial View Showing Involvement of Right and Left Maxilla (B)Left Mandibular Region

ISSN No:-2456-2165

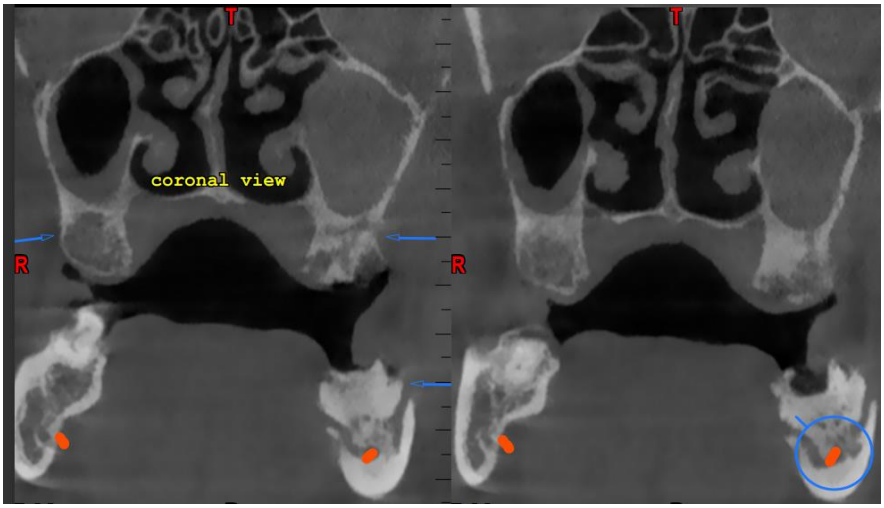

Fig 5 (A) Coronal View Showing the Involvement of Right and Left Maxilla (B) Left Mandibular Region

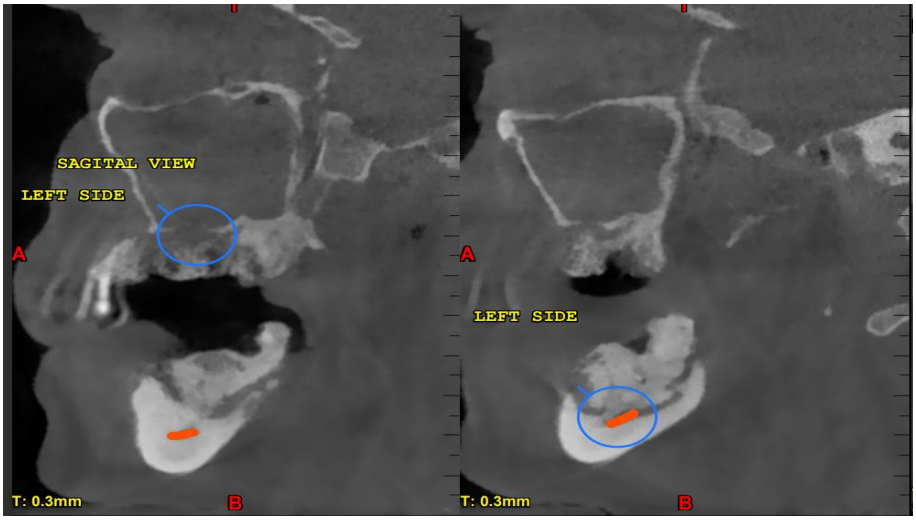

Fig 6 Sagittal View Showing the Involvement of Left Maxilla and Mandibular Region

➢ *Case 2*

A 40-year-old male presented at our Oral Medicine and Radiology department, reporting missing upper arch teeth for two months. The patient recounted sudden tooth loss accompanied by pus discharge, mild radiating pain, bad breath, and a month-long nasal congestion. Despite a history of hemiparalysis a year prior, he was presently healthy without medication. No significant familial associations were noted. Noteworthy is the patient's 15-year history of substantial tobacco chewing 3-4 packets per day.

Extraoral examination revealed no facial asymmetry and Intraoral examination unveiled the completely edentulous maxillary arch. Denuded necrotic bone appeared discolored and devitalized. It had dulled yellow, and brownish hue compared to the surrounding healthy bone. The surface of the necrotic bone appeared rough, porous, and irregular. The texture of denuded necrotic bone was altered. Generalized attrition and an unpleasant odor were observed. Palpation revealed firm, swollen mucosa encircling bony fragments and erythematous alveolar mucosa. No pus discharge or sinus openings were detected, and the area remained non-tender without bleeding tendencies. (Fig 7)

Clinical presentation and history established a provisional diagnosis of chronic osteomyelitis involving the left maxilla. The indurated and inflamed alveolar mucosal periphery prompted the inclusion of malignancy in the differential diagnosis. OPG and CT scans were prior adviced and were conducted.

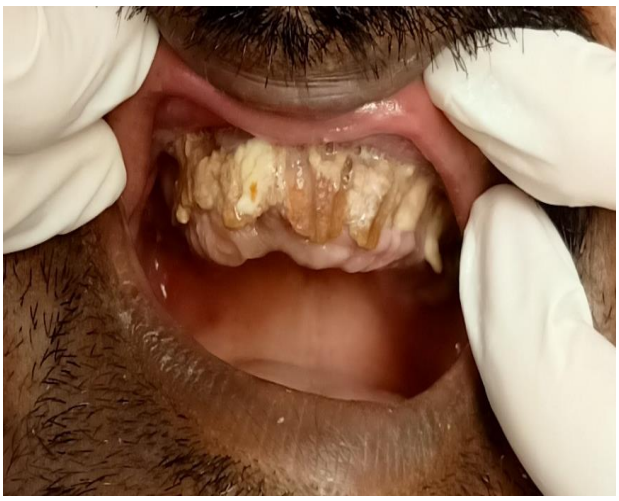
Fig 7 Showing Exposed Necrotic Maxillary Alveolar Bone

ISSN No:-2456-2165

The panoramic radiographs revealed the absence of multiple teeth. Additionally, conspicuous alveolar bony erosion was evident, particularly notable within the confines of the maxillary arch. These findings collectively contributed to the ongoing diagnostic evaluation. (Fig 8)

The initial CT scan provided a comprehensive insight into the structural alterations present. Notably, bilaterally, erosive changes were observed in the medial dental alveolus, bony palate, and pterygoid root. Moreover, erosion was also noted in the lesser wing of the sphenoid bone, as well as the right-sided base of the sphenoid. A leftward deviation of the nasal septum was also discernible. Importantly, this scan unveiled bilateral maxillary and left sphenoidal sinusitis, prompting a critical clinical correlation, and reinforcing the need for subsequent follow-up. (Fig 9)

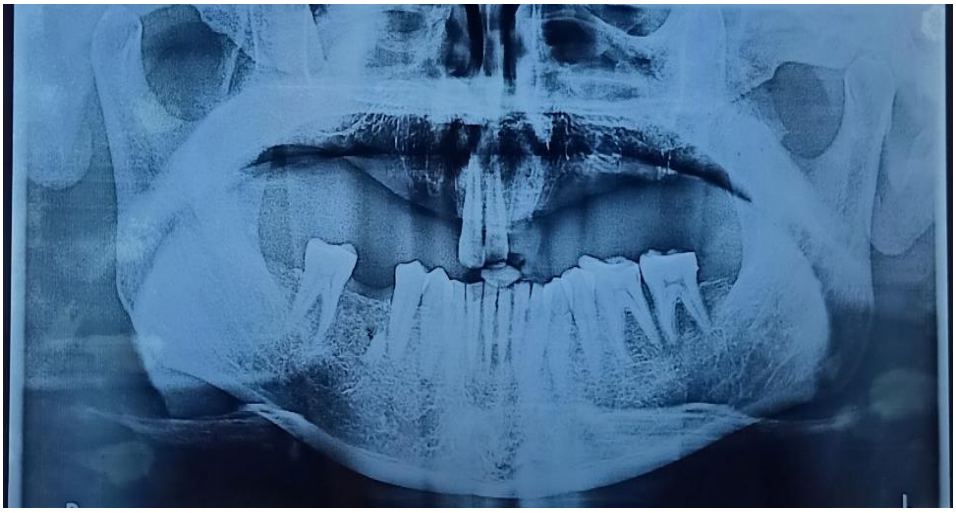

Fig 8 Panoramic Radiograph Reveals Absence of Multiple Teeth. Alveolar Bony Erosion was Evident, Within the Confines of the Maxillary Arch

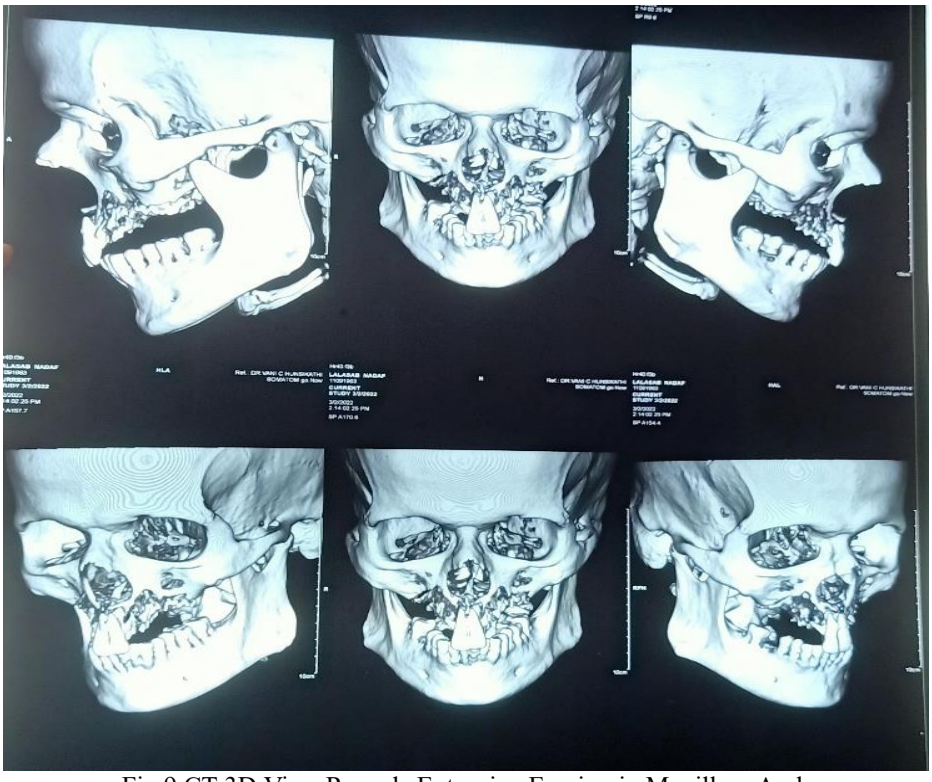

Fig 9 CT 3D View Reveals Extensive Erosion in Maxillary Arch

Sequentially, a CT scan was taken which offered additional layers of information. Specifically, erosion was evident within the hard palate and the posterior wall of the sphenoid sinus. Further scrutiny revealed bilateral maxillary sinus involvement. Additionally, adjacent sclerosis was detected in the Celestica cleavers and the greater wing of the sphenoid bone. A distinctive hyperdense content was identified within the sphenoid bone, accompanied by paracavernal enhancement, which collectively evoked strong indications of chronic osteomyelitis. (fig 10)

ISSN No:-2456-2165

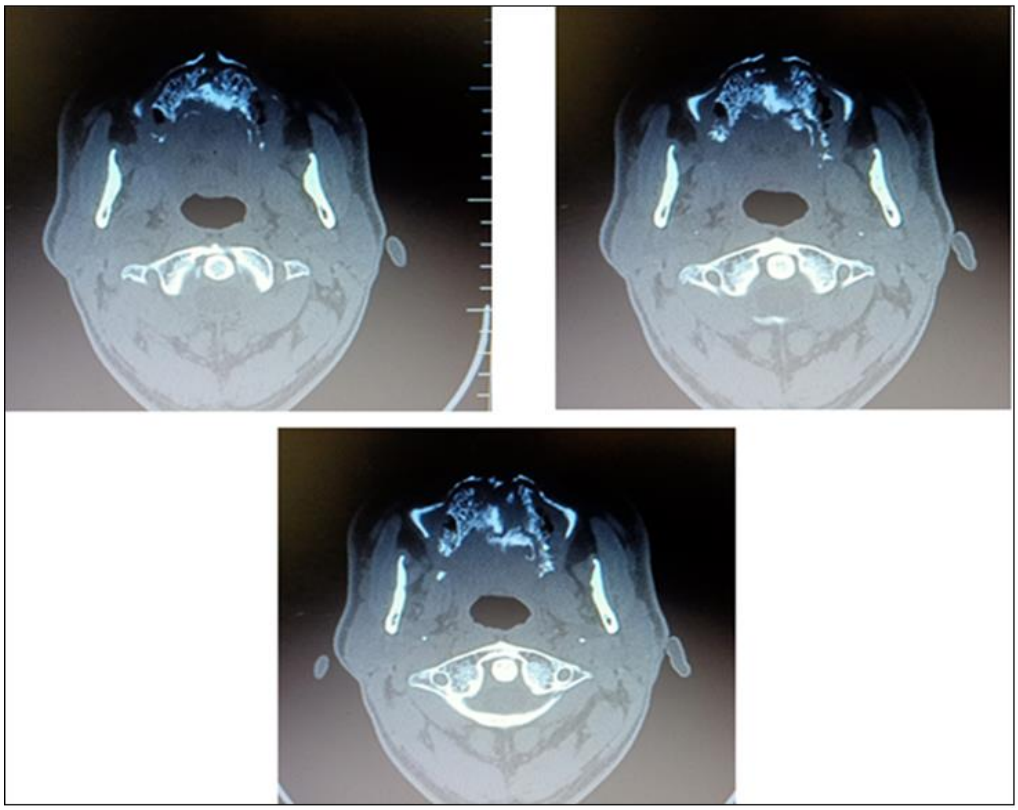

Fig 10 CT Axial View Reveals Osteolytic Changes.

II. CLINICAL PROGRESSION AND REFERRAL

Due to the extensive lesion involving the skull base, the patient was referred to the Department of Oral Maxillofacial Surgery for further evaluation. Given the intricate skull base involvement, the Neurology Department was also consulted. Here, the patient received an escalated antibiotic regimen.

III. DISCUSSION

Osteomyelitis denotes the inflammation involving both the bone and its marrow components. The term finds its origins in Greek mythology, where "osteon" signifies bone and "muelinos" pertains to the medullary aspect of bone. Recognizing the significance of this condition is imperative, as disregard could precipitate severe complications such as cavernous sinus thrombosis, meningitis, cerebral abscess, and septicemia.(2)

Waldegrel and Lew have broadly categorized osteomyelitis of the jawbones as either "Suppurative" or "Non-suppurative."

From a clinical standpoint, this classification can be further delineated into the following divisions:

- Acute suppurative
- Subacute
- Primary chronic (lacking an acute phase)
- Non-suppurative.

The occurrence of osteomyelitis in the jawbones is a rare and complex phenomenon, influenced by multiple factors. Mandibular osteomyelitis is observed 3%-19% more frequently compared to maxillary osteomyelitis.

Several factors contribute to the development of maxillary osteomyelitis, including:

- Traumatic causes (injuries to teeth, lacrimal apparatus, antrum, compound fractures, gingival ulcerations, furuncles, and periosteitis)
- Odontogenic origins (decayed teeth, primary tooth germ)
- Surgical sites and open soft tissue wounds
- Polymicrobial infections linked to diabetes, malignancies, malnutrition, bone metabolic disorders, and immune system suppression, all of which alter the host's response
- Osteoradionecrosis associated with chronic infections stemming from radiation therapy.

While chronic suppurative osteomyelitis most frequently manifests in the antral or odontogenic regions, our case could be attributed to either idiopathic/generalized immunosuppression or the presence of pre-existing bacteria in the dental biofilm of gingivitis and periodontitis. These oral conditions might serve as potential sources of infection, provided that other etiological factors are excluded.

The outlook for individuals with osteomyelitis hinges on various elements, including attributes and virulence of the microbial agents involved (such as Staphylococcus aureus and Staphylococcus epidermidis), host immune reactions, origin of infection, and local tissue blood

ISSN No:-2456-2165

flow.(3,4) The underlying pathophysiological process entails the buildup of inflammatory exudates consequent to microbial infection within the osseous cavities. This accumulation leads to the compression of both central and peripheral blood supplies to the bone, culminating in a compromised oxygen and nutrient delivery due to diminished blood circulation.(5)

Osteomyelitis predominantly affects males within the age range of fifty to sixty years.(6) This condition tends to manifest more frequently in the posterior region of the mandible, characterized by robust cortical plates and restricted blood supply. Clinical presentation includes symptoms like swelling, pain, purulent discharge, paresthesia, trismus, and regional lymphadenopathy. Notably, osteomyelitis often leads to complications such as pathological fractures and paresthesia. The hallmark features of this condition encompass progressive bone degradation and the presence of sequestra.(7)

Diagnosing, treating, and curing Osteomyelitis (OM) of the jaw has posed significant challenges. As mentioned, OM of the jaw can manifest in various ways, leading to multiple diagnoses, some of which are sub-classifications, while others may represent the same condition at different stages or with different expressions. Some diagnoses are primarily based on radiological appearance, the number of affected areas, the patient's age group, and whether there is the presence of pus or recurrence of the disease. Various diagnostic techniques have proven useful, but there is a consensus among many authors that the ultimate diagnosis should consider the following parameters:(1) the patient's clinical presentation and medical history,(8) imaging methods, (9) culturing, and (10) histologic analysis.(11,12)

Suppurative Osteomyelitis (OM) of the jaw presents with severe pain, fever, swelling, purulent discharge, fistulas, limited jaw movement, inferior dental nerve issues, and potential fractures. In milder cases, it can resemble alveolar osteitis.(1)

Non-suppurative OM, features recurrent pain, swelling, limited mouth opening, absence of pus, periostitis, occasional lymph node swelling, and reduced inferior alveolar nerve sensation(13–15).

Primary chronic mandibular OM radiographically shows a mix of sclerosis and osteolysis, leading to mandibular widening, periosteal bone reaction(14,16), and an unclear bone border (13). As it progresses, it tends to become primarily sclerotic, often displaying new bone on the periosteum and sequestra(17).

Primary chronic mandibular OM often exhibits a patchy pattern of osteosclerosis and osteolysis, with an "onion-skin appearance" due to subperiosteal bone formation (15).

Imaging methods for diagnosing jaw OM include conventional radiographs, CT scans, PET/CT scans, laser Doppler flowmetry, MRI scans, nuclear scans, and immunologic workup(10).

Differential diagnoses for bacterial jaw OM encompass Paget's disease, hypercementosis, fibrous dysplasia, and early malignant bone tumors. Radiographically, it can be confused with conditions like osteogenic sarcoma and fibrous dysplasia(17).

In non-suppurative OM, PCO may mimic malignancies like osteosarcoma, chondrosarcoma, Ewing's sarcoma, non-Hodgkin's lymphoma, metastatic disease, histiocytosis X, leukemia, and neuroblastomas, as well as benign conditions including fibrous dysplasia, ossifying and non-ossifying fibroma, juvenile parotitis, chronic sialadenitis, Paget's disease, cementoma, and nonspecific chronic lymphadenitis(10).

Histologically, suppurative OM shows bone necrosis with empty osteolytic lacunae, absent osteoblastic rimming, and empty Haversian canals. Inflammatory cells, active osteoclasts, and blood vessels are present. Microorganisms are detected in about a third of cases(10).

Non-suppurative OM, like PCO, involves chronic inflammation, bone changes, and may not always reveal bacterial factors(10).

Treatment of OM varies based on bacterial involvement, typically involving surgery to control infection and preserve viable bone. Treatments may include debridement, decortication, sequestrectomy, and saucerization. Tooth removal may be needed, and in severe cases, partial resection and reconstruction may be necessary(18).

Medications used for OM treatment include antibiotics, NSAIDs, steroids, and new antibiotics like Linezolid and Tigecycline. Local antibiotic delivery systems are under investigation. NSAIDs may relieve pain and inflammation in primary chronic mandibular OM, and hyperbaric oxygen therapy is effective, especially in refractory cases(10).

In both cases, we observed complex and diverse clinical presentations of maxillofacial osteomyelitis. In Case 1, the patient's history of prostate cancer and hypertension, which can compromise immunity, complicated the condition. The presence of persistent upper left back tooth pain, worsened during meals, along with lower left back tooth numbness, indicated an ongoing inflammatory process in the maxillofacial bones. Clinical examination revealed necrotic bone with characteristic features such as discoloration, roughness, and porosity. Loss of function and numbness suggested nerve involvement. In Case 2, the patient's extensive tobacco chewing history raised concerns about impaired blood supply and immune response, increasing the osteomyelitis risk. Sudden tooth loss, pus discharge, radiating pain, bad breath, and nasal congestion all pointed to severe inflammation. Advanced disease was

evident from the altered texture of denuded necrotic bone and erythematous alveolar mucosa. Notably, the absence of facial asymmetry and the lack of sinus openings during clinical examination helped differentiate osteomyelitis from more extensive facial pathologies.

Radiological imaging is crucial for osteomyelitis diagnosis. Initial orthopantomogram (OPG) scans provided insights into lesion extent and nature. OPG's panoramic view detected erosive changes, disrupted bone architecture, and radiopaque/radiolucent elements. These findings guided further evaluation. For precision, cone-beam computed tomography (CBCT) and CT scans were recommended. These techniques offer high-resolution 3D imaging, detailing affected areas. In Case 1, CBCT confirmed maxilla and mandible involvement, showing the lesion's extent and impact on structures like the maxillary sinus. In Case 2, CT highlighted erosive changes in the hard palate, sphenoid bone, and nearby structures, revealing intricate skull base involvement. CT and CBCT's ability to differentiate healthy and necrotic bone aided diagnosis.

Both cases exemplify the need for clinical correlation and multidisciplinary collaboration in managing maxillofacial osteomyelitis. In Case 1, the patient's history of cancer and hypertension necessitated a comprehensive evaluation to assess potential systemic factors contributing to the disease. The patient's referral to the Oral Maxillofacial Surgery and Neurology departments in Case 2 allowed for a more holistic assessment of the condition, considering both the extent of the osteomyelitis and its impact on adjacent structures.

IV. CONCLUSION

Maxillofacial osteomyelitis can manifest with varying clinical presentations and complications, necessitating careful assessment, timely referral, and collaborative management. These cases contribute to the growing body of knowledge surrounding this condition and emphasize the importance of patient follow-up to ensure optimal outcomes.

Maxillofacial osteomyelitis presents with a range of clinical manifestations and requires a combination of thorough clinical evaluation and advanced radiological imaging for accurate diagnosis and management. OPG and CBCT play pivotal roles in visualizing the extent and nature of the lesions. Multidisciplinary collaboration is essential, especially in cases involving complex skull base lesions, to ensure appropriate and effective management strategies. Continuous research and knowledge sharing are imperative to enhance our understanding of this rare yet significant condition.

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