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Case Report

Dentistry Section

Case Report on Proliferative Periostitis of the Left Mandible: A Radiographic Diagnostic Dilemma and Work-up

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ABSTRACT

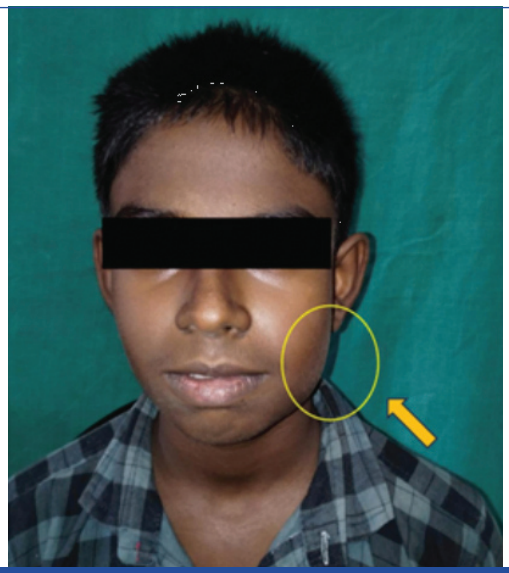
Proliferative periostitis, often referred to as Garre's osteomyelitis, is a distinct type of chronic osteomyelitis marked by the formation of new bone along the periosteal surface. This condition typically occurs in response to a low-grade, persistent infection or irritation, most commonly associated with dental infections or trauma. Early detection and addressing the underlying cause of the infection are crucial for successful management and positive outcomes. Hereby, the authors present a case of a 13-year-old male diagnosed with proliferative periostitis, in whom no evident source of infection could be identified, presenting a diagnostic challenge. Authors highlight the importance of using various radiographic techniques to facilitate a prompt diagnosis, which prevented unnecessary treatment delays and invasive procedures.

Keywords: Contrast-enhanced computed tomography, Garre's osteomyelitis, Periosteal reaction, Persistent infection

CASE REPORT

A 13-year-old male presented with complaints of pain and intermittent swelling on the left lower third of his face, persisting for the past six months. According to the patient's mother, the swelling occurred in episodes and subsided with medication during the initial phase of the disease (antibiotics and analgesics were prescribed). It was associated with prodromal symptoms such as difficulty in opening the mouth. The patient had been treated with antibiotics previously, which provided temporary relief from the swelling. There was no history of trauma, allergies, or significant weight loss. Additionally, the patient's father had passed away from multiple myeloma, and his vaccination records were noted to be incomplete.

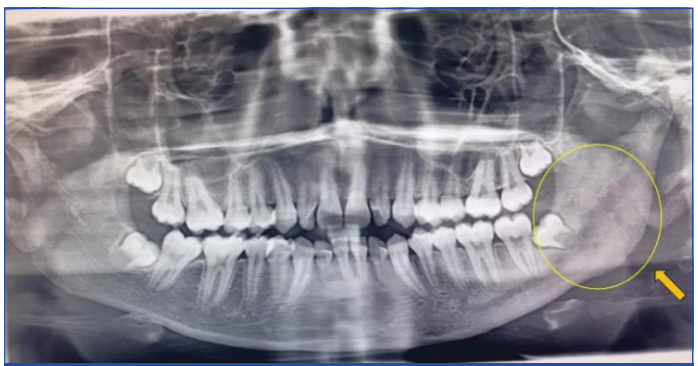
The extraoral examination of the area of chief complaint revealed a firm, diffuse swelling in the left parotid region, which initially developed into the present size later. The swelling was mildly tender to palpation and warm to the touch; however, no sinus tract was observed [Table/Fig-1]. Upon physical examination, a solitary, non tender, freely mobile submandibular lymph node, approximately 0.5 cm in size, was identified, along with a similarly sized middle cervical lymph node on the left-side.



[Table/Fig-1]: Extraoral swelling in the left one-third of the face.

Intraoral examination showed normal dentition for the patient's age and good oral hygiene. Based on the clinical presentation and the absence of a clear odontogenic cause, a provisional diagnosis of recurrent left parotitis was made, characterised by intermittent swelling and palpable lymph nodes. Radiological and haematological investigations were conducted to rule out odontogenic or infectious causes; however, these investigations did not yield conclusive results.

A panoramic radiograph [Table/Fig-2] showed changes in the trabecular pattern, including widening of the margins of the left inferior alveolar nerve canal and a ground-glass appearance. Additionally, an unerupted right lower third molar was observed.

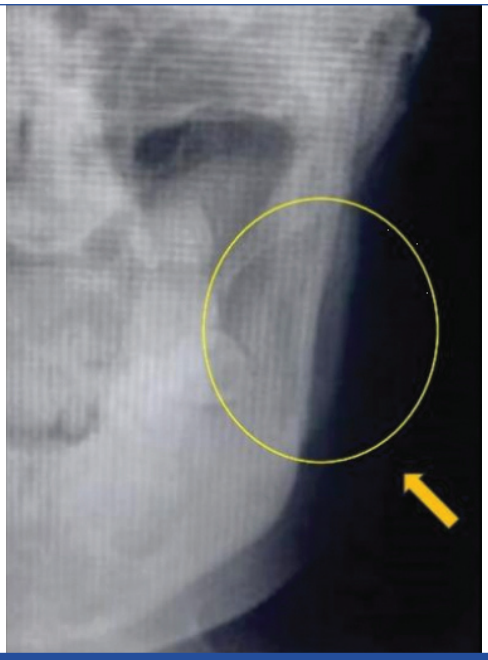


[Table/Fig-2]: Orthopantamograph showing changes in trabecular pattern.

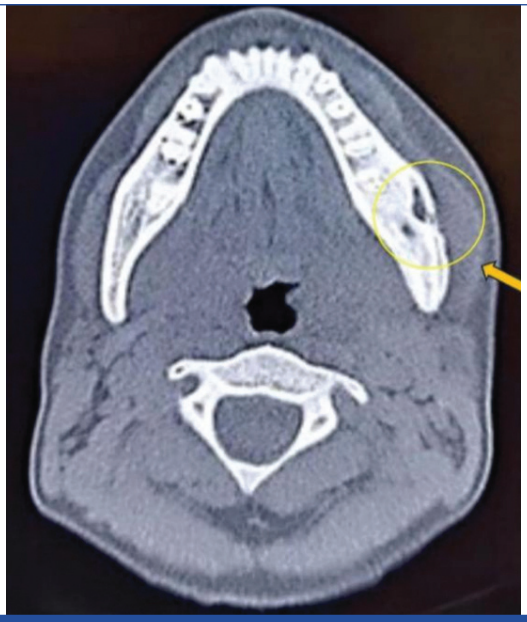
A Posteroanterior (PA) skull view [Table/Fig-3] revealed thickening of the cortical border in the left ramus, suggesting the possibility of a periosteal reaction.

However, due to the lack of clinical signs of oral infection or trauma, a differential diagnosis of proliferative periostitis or early-stage fibrous dysplasia was considered. The patient's family history of malignancy added complexity to the diagnosis, necessitating further investigations to rule out the possibility of malignancy.

Ultrasonography revealed prominent lymph nodes with a compressed fatty hilum in levels IA, II, III, and V, including a 9 mm node in the left IB region. Hyperechoic areas were also observed over the left masseter muscle. Contrast-enhanced Computed Tomography (CECT) axial section [Table/Fig-4] confirmed a significant periosteal reaction, along with a hyperdense area in the left mandibular ramus, supporting the diagnosis of proliferative periostitis.

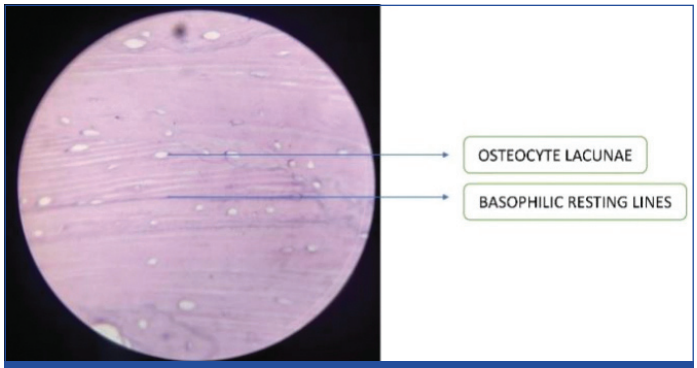


[Table/Fig-3]: The PA view of the skull showing thickened cortical border in left ramus.



[Table/Fig-4]: The CECT of skull showing significant periosteal reaction.

Fine Needle Aspiration Cytology (FNAC) did not provide definitive results, prompting an incisional biopsy. Histopathological examination of the Haematoxylin and Eosin (H&E)-stained tissue of the specimen revealed compact bone arranged in a lamellar pattern, containing numerous spindle-shaped osteocytes within osteocyte lacunae, along with basophilic resting lines suggestive of bone remodeling. These findings confirmed the diagnosis of proliferative periostitis [Table/Fig-5].



[Table/Fig-5]: Compact bone arranged in lamellar pattern with basophilic resting lines (H&E, 40X).

The source of the infection was identified as an unerupted wisdom tooth. Due to the parents' apprehension regarding early tooth extraction and the patient's reluctance to proceed with treatment, a recommendation was made to monitor the condition. The patient was advised to undergo tooth extraction and jaw recontouring in the future. Since, the patient's parents were hesitant about the extraction of the tooth at an earlier age, the patient was advised to monitor the situation, as they were not willing to proceed with the treatment. The patient was placed on a routine empirical therapy regimen with antibiotics (Tab. Amoxicillin clavulanic acid- 625 mg for five days) and T. Hifenac-P, which was advised for five days after meals. Regular follow-up was recommended as interim measures. The patient was followed-up for three months and presented with non palpable lymph nodes, with no recurrence noted; after this period, the patient lost to follow-up.

DISCUSSION

Proliferative periostitis predominantly occurs in children and adolescents and represents a defensive reaction by the periosteum, the dense layer of vascular connective tissue surrounding bones, except at joint surfaces. The condition was first described by Carl Garre in 1893, as mentioned by Zand V et al., and the term "periostitis ossificans" was introduced by Gorman in 1951 [1,2]. Typically, the mandible, especially the lower border is the most frequently affected site, which aligns with the findings in the current case [3].

Chronic low-grade infections, often stemming from dental problems such as caries, periodontal disease, or unerupted teeth, are the primary causes of proliferative periostitis. Tong ACK et al., described this condition as initiating from a persistent inflammatory response, leading to new bone formation as a protective reaction [4]. While typically painless, the affected area may present as unilateral swelling that can be detected by palpation. According to Passi S et al., differentiating proliferative periostitis from conditions like fibrous dysplasia, Ewing's sarcoma, or osteosarcoma is important, as these conditions may present similarly but are more aggressive [5]. Imaging, such as cone beam Computed Tomography (CT), can help confirm the benign nature of proliferative periostitis.

Treatment involves removing the source of infection, such as through tooth extraction or endodontic therapy. Antibiotics may be helpful, and surgery is only needed in rare cases. With proper care, the prognosis is excellent, as the bone usually remodels to restore a normal appearance [5]. Regular follow-up is essential to ensure full recovery and prevent recurrence [6].

According to Chang YC et al., radiographic differential diagnoses for proliferative periostitis include fibrous dysplasia, osteosarcoma, and Ewing's sarcoma [7]. Timely diagnosis and treatment, generally involving the elimination of the infection source and subsequent bone remodeling, are crucial for achieving favourable outcomes. According to Neville BW, White SC et al., and Whaites E et al., in contrast to osteomyelitis, Ewing's sarcoma and osteosarcoma exhibit distinct radiographic features and can lead to more severe complications, such as rapid bone enlargement and neuralgia [8-10]. In the present case, due to the presence of a diffuse ground-glass appearance in the left ramus region, it was radiographically considered fibrous dysplasia of the left ramus region. On CECT, there was a hyperdense area in the left ramus region, and histopathological examination revealed compact bone arranged in a lamellar pattern instead of fibrous connective tissue with a normal blood profile. The following features were considered to rule out fibrous dysplasia.

The current case of proliferative periostitis offers valuable insights into its connection with Garre's osteomyelitis, particularly concerning the pathophysiology linked to chronic low-grade infections. The present case highlights the diagnostic and therapeutic challenges associated with proliferative periostitis, a rare condition predominantly affecting children and adolescents. Park J and Myoung H reported similar

cases of chronic suppurative osteomyelitis with proliferative periostitis in paediatric patients, emphasising the rarity of such presentations, particularly those linked to unerupted or impacted third molars [11]. Their concise summary of these cases underlines the diverse clinical manifestations and management strategies required for effective treatment. In their report, surgical intervention- comprising germectomy, debridement, and saucerisation- was pivotal in achieving resolution, complemented by appropriate antibiotic therapy.

In comparison, the current case employed advanced imaging and histopathological confirmation to overcome diagnostic challenges. These findings underscore the importance of early and accurate diagnosis to prevent complications and optimise outcomes [Table/Fig-6] [4,11-16]. Additionally, incorporating the below documented cases into the present discussion emphasises the clinical uniqueness of this report and highlights the need for a comprehensive approach to differential diagnoses, including conditions like fibrous dysplasia and other periosteal reactions.

Author(s)	Case details	Treatment
Tong ACK et al., [4]	Chronic osteomyelitis with proliferative periostitis in a 12-year-old boy at the mandibular angle	Not specified. Secondary to an un-erupted third molar tooth germ
Park J and Myoung H [11]	Chronic suppurative osteomyelitis with proliferative periostitis in an 11-year-old female due to fully impacted third molar germ	Tooth extraction and saucerisation
Park J and Myoung H [11]	Chronic suppurative osteomyelitis with proliferative periostitis in a 12-year-old female due to fully impacted third molar germ	Tooth extraction with debridement and saucerisation
Reck SF et al., [12]	Osteomyelitis of the coronoid process in a 16-year-old boy due to third molar pericoronal infection.	Tooth extraction and partial coronoidectomy
Mohammed-Ali RI et al., [13]	Osteomyelitis of the mandibular ramus from the lower left third molar in a 22-year-old female	Tooth extraction and exploration/drainage from the left submasseteric space
Mohammed-Ali RI et al., [13]	Sclerosing osteomyelitis in the lower right third molar extraction space of a 21-year-old female	Extensive decortication
Lambade P et al., [14]	Condylar osteomyelitis due to an ectopic third molar in a 35-year-old female.	Extraction, curettage, and drainage
Wang R et al., [15]	Osteomyelitis linked to the lower left third molar in a 37-year-old male.	Extraction, curettage, and drainage.
Yoshida Y et al., [16]	Garre's osteomyelitis in a 12-year-old female due to germ infection in the right lower impacted wisdom tooth	Extraction
Present case	Proliferative periostitis was secondary to an unerupted third molar tooth germ, unlike others related to pericoronitis of a developed molar.	The patient was prescribed routine empirical therapy

[Table/Fig-6]: Comparision with cases from past literature [4,11-16].

Histopathological analysis is vital for distinguishing proliferative periostitis from other conditions. The histological characteristics observed in this case- featuring compact bone with spindle-shaped osteocytes- support a diagnosis of a benign reactive process. This finding corresponds with the work of Kim H et al., who emphasised the importance of histological evaluations in confirming non-suppurative osteomyelitis [17]. Yoshida Y et al., reported a similar case of Garre's osteomyelitis in a 12-year-old patient caused by infection in the dental germ of an impacted wisdom tooth. Their management strategy, which involved the surgical removal of the impacted tooth germ and hyperostotic bone, resulted in the complete resolution of swelling and normalisation of bone structure within nine months [16].

In the current case, the clinical presentation and imaging findings, including periosteal thickening and bone remodeling, parallel the observations described by Yoshida Y et al., [16]. Their study emphasises the importance of timely surgical intervention in eliminating the source of infection and achieving favourable outcomes. Advanced imaging techniques, such as CT and bone scintigraphy, also proved crucial in both cases to exclude differential diagnoses like malignant bone conditions and guide effective treatment.

Thus, the current case illustrates the effectiveness of a conservative management approach that addresses the condition without necessitating extensive surgical procedures. The patient was conservatively managed with routine empirical therapy, considering the age of the patient.

Furthermore, advances in imaging techniques have become essential in the diagnostic process. In this study, the use of contrast-enhanced CT facilitated the identification of the periosteal reaction and excluded malignancies. This method is consistent with findings from Ghazali FR and Samsudin AHZ highlighting that precise imaging can significantly lower the risk of misdiagnosis when distinguishing between benign and malignant processes [18].

Treatment for proliferative periostitis typically involves endodontic therapy, extraction of affected teeth, antibiotics, and, in certain cases, surgical intervention. Once the infection is managed, the prognosis is generally positive, with new bone formation typically stabilising without leading to functional impairment. In the present case, the causative factor was considered to be the unerupted third molar. Considering the age of the patient and the fact that the patient's mother was not willing to proceed with the extraction, the patient was placed under routine empirical therapy. In present case, the radiographic investigation confirmed osteomyelitis, which was subsequently validated by histopathological examination.

CONCLUSION(S)

The present case report underscores the diagnostic challenges of proliferative periostitis when no clear odontogenic infection is present. Utilising advanced radiographic techniques, such as CECT, along with histopathological examination, is essential for excluding malignancy and guiding appropriate management. Accurate diagnosis and treatment rely on the integration of clinical, radiographic, and histopathological findings to achieve successful outcomes.

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