CASE REPORT

An unusual presentation of garre's osteomyelitis of mandible: A case report

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ABSTRACT

Chronic non-suppurative osteomyelitis, also known as Garre's osteomyelitis is a local thickening of periosteum caused by slight irritation or infection. Here, we present the case report of a unilateral bony swelling in the left side of the lower jaw associated with an infection in an 8-year-old child. The radiograph revealed the pathognomic feature of 'onion skin' appearance. Surgical recontouring of the lower jaw was performed since there was no evidence of bone remodelling after the removal of granulation tissues.

Key words: Orthopantomograph, cone beam tomography, onion skin, histopathological examination

Garre's osteomyelitis, which was first described by Carl Garre in 1893, is a chronic non-suppurative sclerotic bone inflammation characterized by a focal gross thickening of periosteum with peripheral reactive bone formation resulting from infection [1]. The first cases affecting the jaw were reported in 1948 by Berger, and Pell described them in 1955. In general, osteomyelitis has a bimodal age distribution. Acute hematogenous osteomyelitis is primarily a disease in children. Direct trauma and contiguous focus osteomyelitis are more common among adults and adolescents than in children [2].

Vertebral osteomyelitis is more common in persons older than 45 years. Garre's osteomyelitis is due to long-standing periapical infection, recent infection of soft tissue, and unhealed infected extraction areas [3]. The mandible is more often affected than the maxilla in men aged below 30 years, and it is most generally seen at the lower margin of the mandible in the mandibular first molar region because the maxilla has a rich vascular supply and thin cortical plates while the blood supply to the mandible is only limited to the inferior alveolar artery. There is typically a non-tender swelling on the mesial and lateral sides of the jaw, with the size of swelling from 1-2 cm to the involvement of the entire length of the jaw on the affected side by the thickness of the cortex can reach 2-3 cm, caused by persistent low-grade infection in children and young adults in the presence of high osteoblastic activity triggers the periosteum to lay down new bone [4]. Garre's osteomyelitis presents as facial asymmetry on the lower part of the face with a solitary swelling with normal to inflamed overlying skin and on palpation, the swelling is bony hard and non-tender. The causative factors associated with this lesion are grossly carious tooth, periodontal defects, persistent infection, and trauma [5,6]. There is no macroscopically suppurative lithic area in cases of Garre's osteomyelitis, histopathological examination has detected microabscesses and microsequesters. Orthopantomograph (OPG) reveals a periosteal thickening of the inferior border of the mandible due to an increase in osteoblastic activity resulting in an "onion skin" appearance [6,7].

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An 8-year-old female patient reported to our department with a chief complaint of facial asymmetry in the lower portion of her face for six months. The swelling was initially small and gradually increased to its final dimensions. On general examination, the patient was mesomorphic, having a normal gait with stable vitals. The patient was previously healthy with no known systemic illnesses/allergies and had not received any medications apart from frequent antibiotics and NSAIDs for painful episodes of jaw swelling. No trauma/ dental history was reported. There was no significant family history.

On extraoral examination, the facial asymmetry with a solitary localized oval-shaped swelling measuring 30 mm x30 mm was seen on the left side of the face near the body of the mandible (**Figures 1A & 1B).** On palpation, the swelling was

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noted to be well-defined, hard in consistency, non-tender, nonpulsatile, and non-fluctuant. Localized ipsilateral submandibular lymphadenopathy was also noticed. Intraorally, obliteration of the lower buccal vestibule on the left side was seen **(Figure 1C).**

Figure 1(A & B): Pre-operative clinical photographs showing facial asymmetry on the left side of the mandibular region as seen in front and worm's view; (b) Clinical photograph showing mild obliteration of the lower buccal vestibule on the left side

The radiographic examination revealed an 'onion skin' appearance at the inferior border of the mandibular body suggestive of a periosteal reaction **(Figure 2)** and a radiolucent area in the apical region of the left mandibular first molar tooth. When the axial and cross sections were evaluated during the examination with cone-beam computed tomography (CBCT), a tunnel-like defect was identified in the cortical bone in the cortical bone in the vestibule surface of inflamed bone, starting from the apical region of the left mandibular first molar tooth **(Figure 3).** Based on the clinical examination, radiographic findings, and histopathological examination, the lesion was diagnosed as Garre's osteomyelitis, and surgical recontouring under general anesthesia was planned **(Figure 4).** We exposed the lesion by intraoral approach, and excess bony mass was excised using a surgical bur and handpiece.

Figure 2: Orthopantomograph reveals expansion of bone along with radiolucency in the outer cortex of the inferior aspect of the mandible on the left side

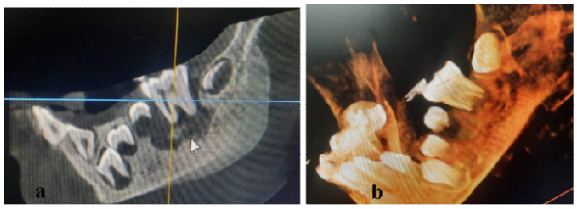

Figure 3: a) and b) Axial and cross sections in CBCT showing new bone formation and a tunnel-like defect in the vestibule cortical surface of the inflamed bone starting from the apical region of tooth number 36.

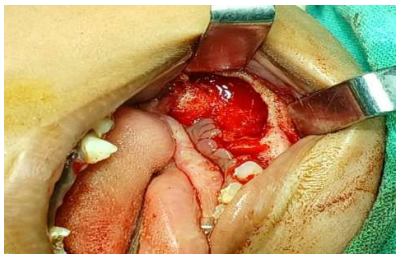

Figure 4: Exposed bony defect where sequestrum and necrotic tissue was present

The excised bone fragments were sent for histopathological examination which revealed trabeculae of bone with osteoblastic rimming and reversal lines together with a chronic inflammatory infiltrate and that microscopic picture confirmed the diagnosis of Garre's osteomyelitis **(Figure 5c).** The patient was followed up 3 months and 6 months post-surgery respectively with no recurrence of the lesion **(Figures 5 a & b).**

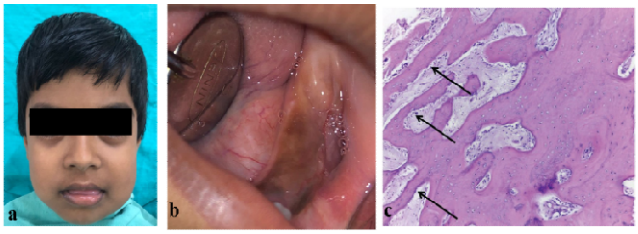

Figure 5: (A) Post-operative clinical photograph at 1 month reveals gross improvement in facial symmetry as seen in frontal view. (B) Follow up of the patient after 6 months showing healing socket area and good vestibular depth. (C) Histopathological analysis shows parallel rows of richly cellularized reactive trabecular bone extending from the cortical surface (thin arrows)

DISCUSSION

Garre's osteomyelitis is a localized periosteal thickening caused by mild irritation or infection. It is known that a moderate infection (such as dental decay, periodontal disease, or soft tissue disease), starting from the spongiosa layer of the jaw and extending into the periosteum is the result of stimulating bone formation. Its development depends on the interplay of chronic infection, the activity of osteoblasts, the virulence of infectious agents, and host resistance [7]. A possible way in which bone formation takes place is illustrated in **(Figure 6).** Normal bone surrounds a source of irritation, such as a dental abscess (A) (stage 1). The bone at this stage has an unaffected and normal periosteum. Sinus formation (S) begins and pus tracks toward the surface of the bone (stage 2).

The pus (X) reaches the surface of the bone and causes a subperiosteal bone reaction (R) which initiates new bone formation. Subperiosteal bone formation continues as long as the mild chronic stimulus persists (stage 4). Successive layers of new bone are deposited, leading to the "onion-peel"

appearance seen on radiographic examination. This may continue (stage 5) or the source of irritation may be removed (stage 6). After a period of time the bone contour will return to normal when the cause of the reaction has been removed. This probably conforms to Wolff's law [8]. Bennett suggests that the increase in the mass of bone found in this condition may be due to a mild toxic stimulation of the periosteal osteoblasts by a low-grade infection. The excessive bone deposition may represent an exuberant attempt at repair [9,10].

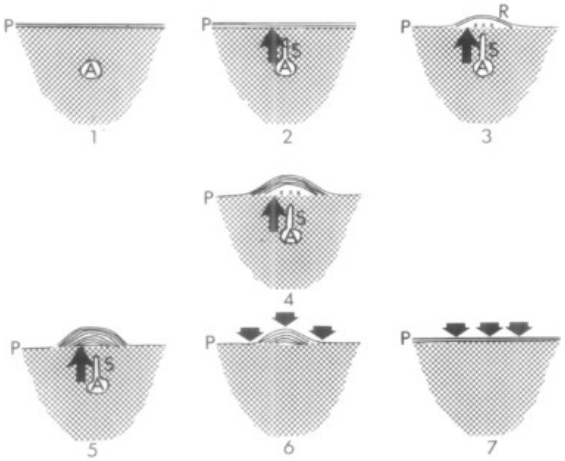

Figure 6: Probable pathogenesis of Garre's osteomyelitis. 1, Dental abscess; 2, pus tract; 5, sinus formation; 4, subperiosteal bone deposition; 5, onion-peel formation; 6, irritation removal; 7, bone remodelling.

There is no need for biopsy during diagnosis of Garre's osteomyelitis, except the cause is unknown and conventional radiograph methods or CBCT images are sufficient for the diagnosis. In addition to Garre's osteomyelitis, new bone formation can occur in many pathological conditions. It should be distinguished from other pathologies that cause new bone formation, including Ewing's sarcoma, Caffey disease, Fibrous dysplasia, Paget's disease, or osteosarcoma [11]. Ewing's sarcoma is similar to Garre's osteomyelitis in terms of the subperiosteal bone formation and appearance in young people but it can also be distinguished from Garre's osteomyelitis due to producing osteophytes with a "sun ray" appearance, causing bone enlargement rapidly and causing more osteolytic reactions in the bone, as well as the occurrence of frequent complications such as facial neuralgia and lip paraesthesia [12].

Caffey disease presents in a similar view to Garre's osteomyelitis due to onion skin appearance in the bone but it is distinguished from Garre's osteomyelitis due to early age of onset, it is more common in the ramus and angle region of the mandible with bilateral involvement [13]. Fibrous dysplasia is seen at younger ages, which is similar to Garre's osteomyelitis, and the resulting bone mass is similar in both shape and volume but it is distinguished from Garre's osteomyelitis due to the "ground glass appearance" as well as thinning seen in the cortex [14]. Treatment options include antibiotics, anti-inflammatory drugs, colchicine, steroids, conservative therapy, and surgery.Management varies from conservative to surgical approach including endodontic therapy, extraction of tooth, and recontouring the bone [9].

The latest treatment modality for Garre's osteomyelitis involves resection of bone followed by transport distraction osteogenesis. Antibiotic therapy alone is ineffective, independent of the administration route, because the "bone sequestration" found in the chronic disease comprises fragments of necrotic bone and thus does not present a blood supply that would allow antibiotics to arrive at the infected tissue. Suma R et al [7] advocate the use of metronidazole and gentamicin as an irrigating system after the systemic use of analgesics and antibiotics. A sequestrectomy is a surgical procedure involving the removal of a sequestrum which is a fragment of dead bone or other tissue that has separated from healthy tissue as a result of injury or disease. Such fragments (the plural form is sequestrum) often end up in a wound or abscess (a collection of pus) [15]

CONCLUSION

Garre's osteomyelitis is associated with low-grade infection usually arising from an infected tooth that results in stimulation of bone formation. We all agree that, in the presence of an infected tooth, the microorganisms are responsible for the irritation causing the host's proliferative response. In the present case, granulation tissue was found and no such bony tissue was found.

Declaration of patient consent: The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

REFERENCES:

1. Cierny Iii G, Mader JT, Penninck JJ. The classic: a clinical staging system for adult osteomyelitis. Clini Orthopae Related Res. 2003; 414:7-24.
2. Topazian RG, Goldberg MH, Hupp JR. Oral Maxillofac Infect. 2002; 3.
3. Schwartz S, Pham H. Garre's osteomyelitis: A case report. Pediatr Dent. 1981; 3(3):283-6.
4. Chang YC, Shieh YS, Lee SP, et al. Chronic osteomyelitis with proliferative periostitis in the lower jaw. J Dent Sci. 2015; 10(4):450-5.
5. Akgül HM, Çağlayan F, Yılmaz SG, et al.Osteomyelitis of the mandible caused by infected tooth. Case Reports in Dent. 2018; 1:1409539.
6. Nikomarov D, Zaidman M, Katzman A, et al. New treatment option for scleros osteomyel of Garré. J Pedia Orthopae B. 2013; 22(6):582.

7. Suma R, Vinay C, Shashikanth MC, Reddy VS et al. J Society of Pedodont and Preven Dentis. 2007; 25:S30-3.
8. Betts NJ, Abaza NA, Kazemi A. An expansile bony lesion of the posterior mandible in a 12-year-old girl. J oral and maxillofac surg. 1996; 54(2):203-9.
9. Athikesavan Jayasenthil AJ, Aparna PV, et al. Non-surg endodont mgmt. of Garre's osteomyelit: A case report. J Adv in Medi Med Res. 2015; 9(3):1-4.
10. Bennett GE, Hopkins JV. Report of a case of scleros osteomyelit associated with Trichinosis. J Bone Joint Surg. 1928; 10(4):834-9.
11. Nortje CJ, Wood RE, Grotepass F. Periostitis ossificans versus Garrè's osteomyelitis: Part II: Radiologic analysis of 93 cases in the jaws. Oral Surgery, Oral Med, Oral Pathology. 1988; 66(2):249-60.
12. Smith SN, Farman AG. Osteomyelit with proliferative periostit (Garre's osteomyelitis): Report of a case affecting the mandible. Oral Surgery, Oral Med, Oral Patho. 1977; 43(2):315-8.
13. Kannan SK, Sandhya G, Selvarani R. Periostitis ossificans (Garrè's osteomyelitis) radiograph study of two cases. International J Paediatric Dent. 2006; 16(1):59-64.
14. Eswar N. Garre's osteomyelitis: A case report. J Indian Society of Pedodont Prevent Dent. 2001; 19(4):157-9.
15. Ahmed KA, Khonglah TG, Borgohain B et al. An Uncomm Presentat of Chronic Osteomyelit of the Fibula in an 11-Year-Old Child: A Case Report. J Clini Diagno Res. 2019; 13(6):1-3.

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