CASE REPORT

Diagnostic Challenge Involving Diffuse Mandibular Swelling in Mixed Dentition: Use of Cone-beam Radiography

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Abstract

Aim and objective: This case report aims to discuss a case of chronic osteomyelitis with proliferative periostitis (COPP) in which cone-beam computed tomography (CBCT) played a key role in the diagnosis and identification of the disease.

Background: Osteomyelitis is frequently seen in the maxillofacial region, with the mandible being more affected as compared to the maxilla. Chronic osteomyelitis with proliferative periostitis is often seen in children and young adults, which occurs due to the spread of infection from a bacterial focus.

Case description: A 12-year-old boy presented with a swelling on the right side of the mandible. Extraorally, the swelling appeared diffuse, non-tender, hard on palpation, and was associated with right submandibular lymphadenopathy. Intraorally, a hard bony protrusion was found on the buccal aspect of the edentulous space of the first premolar region obliterating the vestibular depth. It was provisionally diagnosed to be a case of chronic alveolar abscess with respect to deciduous molar, with a suspicion of some underlying bony pathology. Blood investigations showed a raised value of erythrocyte sedimentation rate and alkaline phosphatase. Cone-beam computed tomography examinations showed bony laminations parallel to each other and the underlying cortical bone in the axial and sagittal images and these were present both buccally and lingually. The CBCT descriptions led to the diagnosis of Garre's osteomyelitis in this case which was later confirmed by histopathological examination.

Conclusion: The detailed imaging as well as the three-dimensional reconstruction of the images has helped to reach out to a differential diagnosis in this case.

Clinical significance: This case report emphasizes the importance of newer techniques such as CBCT in reaching out at a diagnostic conclusion in challenging clinical cases.

Keywords: Garre's osteomyelitis, Mandibular osteomyelitis, Osteomyelitis, Proliferative periostitis.

International Journal of Clinical Pediatric Dentistry (2021): 10.5005/jp-journals-10005-1947

Background

Osteomyelitis of the jaws is a serious and challenging disease despite many advances in diagnosis and treatment. The word "Osteomyelitis" is derived from the Greek words "osteon" and "muelinos" meaning bone and marrow, respectively. Thus, osteomyelitis is understood as inflammation of the medullary portion of the bone. According to Lew and Waldvogel, osteomyelitis is classified as suppurative and non-suppurative related to the focus of infection.[1]

Chronic non-suppurative sclerosing osteomyelitis is also known as proliferative ossificans, chronic sclerosing inflammation of the jaw, Garre's sclerosing osteomyelitis, juvenile chronic mandibular osteomyelitis, or chronic osteomyelitis with proliferative periostitis (COPP).[2,3] Carl Garre first described this disease or condition in 1893 as "an irritation induced focal thickening of the periosteum and cortical bone of the tibia". Clinically, it is characterized by distention and thickening of the bone, but there is a lack of suppuration, sequestration, or fistularization. Gorman coined the term, periostitis ossificans, to specifically identify the formation of new bone overlying existing cortical bone.[4] The condition is seen exclusively in children or young adults. The mandible is generally more often affected than the maxilla.[5]

Here, we report a case of COPP in a 12-year-old child diagnosed by using cone-beam computed tomography (CBCT). Cone-beam computed tomography provides high-resolution images, with added advantages of shorter scanning durations and reduced radiation exposure when compared with conventional computed tomogram (CT), but the soft tissue changes are difficult to detect.

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How to cite this article: Passi S, Gupta J, Mehta A, *et al.* Diagnostic Challenge Involving Diffuse Mandibular Swelling in Mixed Dentition: Use of Cone-beam Radiography. Int J Clin Pediatr Dent 2021;14(3): 430–433.

Source of support: Nil

Conflict of interest: None

Case Description

A 12-year-old male child patient visited the Department of Pediatric Dentistry with the presenting complaint of swelling and pain on the right side of the mandible for 6 months. The patient presented with a history of extraction of a carious tooth from the region 4

months back. On extraoral examination, it was found that a diffuse non-tender swelling was present on the right side of the mandible. The swelling was hard on palpation and was associated with right submandibular lymphadenopathy. The overlying skin color was normal. On intraoral examination, a hard bony swelling was found on the buccal aspect of the edentulous space of the first premolar region obliterating the vestibular depth (Fig. 1A) and was extending till the first permanent molar region.

Based upon these preliminary findings, it was provisionally diagnosed to be a case of chronic alveolar abscess with respect to deciduous molar, with a suspicion of some underlying bony pathology. To differentially diagnose the case, the patient had been advised a series of blood investigations and radiographic investigations (orthopantomogram, cone-beam CT). Complete blood count, bone biochemistry blood tests including levels of calcium, phosphate, electrolytes, etc., all were well within the normal range. Increased values of erythrocyte sedimentation rate (32 mm/hour) and alkaline phosphatase (280 U/L) were present.

Orthopantomogram showed developing tooth buds of mandibular right canine, premolars, and molars (Fig. 1B). A radiolucency was seen in the periapical area of the developing right second premolar area and there was also seen the localized thinning of the lower border of the mandible.

Cone-beam computed tomography examinations showed bony laminations parallel to each other and the underlying cortical bone in the axial and sagittal images and these were present both buccally and lingually (Figs 2A and B). The lamellae were more prominent in the posterior sections of the mandible. However, the buccal cortex was destroyed in the anterior sections especially in the second premolar and molar region. Areas of small sequestrum and osteolytic radiolucencies were seen within the newly formed bone (Fig. 2C), which were alternating with each other. Enlargement of the affected mandibular canal was also evident. A three-dimensional reconstruction image showed an increase in the cortical bone and volume of the mandible on the right side. Based upon these findings in the CBCT, a provisional diagnosis of Garre's osteomyelitis was formed in this case.

The lesion was planned to be surgically curetted under 2% lignocaine with 1:200,000 adrenaline concentration. Informed verbal consent was taken from the child's parents to carry out the treatment. The surgical curettage of the area was followed by antibiotics along with anti-inflammatory drugs which were started 1 day before the surgery and continued for 8 weeks. Clinically, the patient's face was symmetrical and no swelling was present either extraorally or intraorally. An OPG was repeated which revealed the eruption of underlying mandibular first premolar but thinning around the lower border was still present (Fig. 3). The patient was recalled after 3 months but unfortunately, he did not report back. The tissue samples sent for histopathology showed irregular trabeculae of bone with empty appearing osteocytic lacunae in a background of collagenous tissue, areas of hemorrhage, and inflammatory cells (Fig. 4). These findings further supported the clinical diagnosis reached out through CBCT.

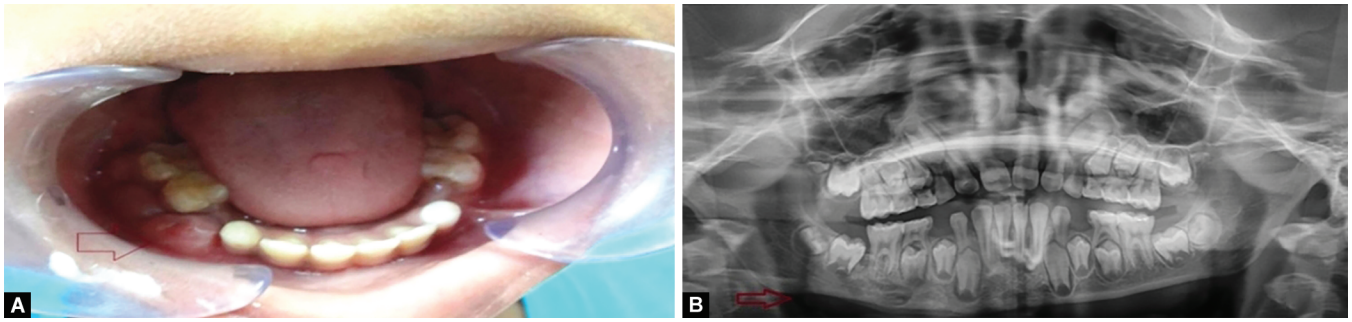
Discussion

Periapical infections arising from dental caries, pericoronal infection, dental extractions, or periodontal disease may be causative factors in COPP. However, keeping in consideration that a tooth follicle or the underlying tooth bud is usually non-infected, the finding of COPP in such cases may then be considered to be of unknown origin.[6] On occasion, no infectious cause is detectable. In the present case, a definitive locus of infection was not identifiable. It was speculated that the origin of COPP could be due to extraction of carious tooth in that region or could be due to the unerupted lower right second premolar as a periapical radiolucent area was present there.

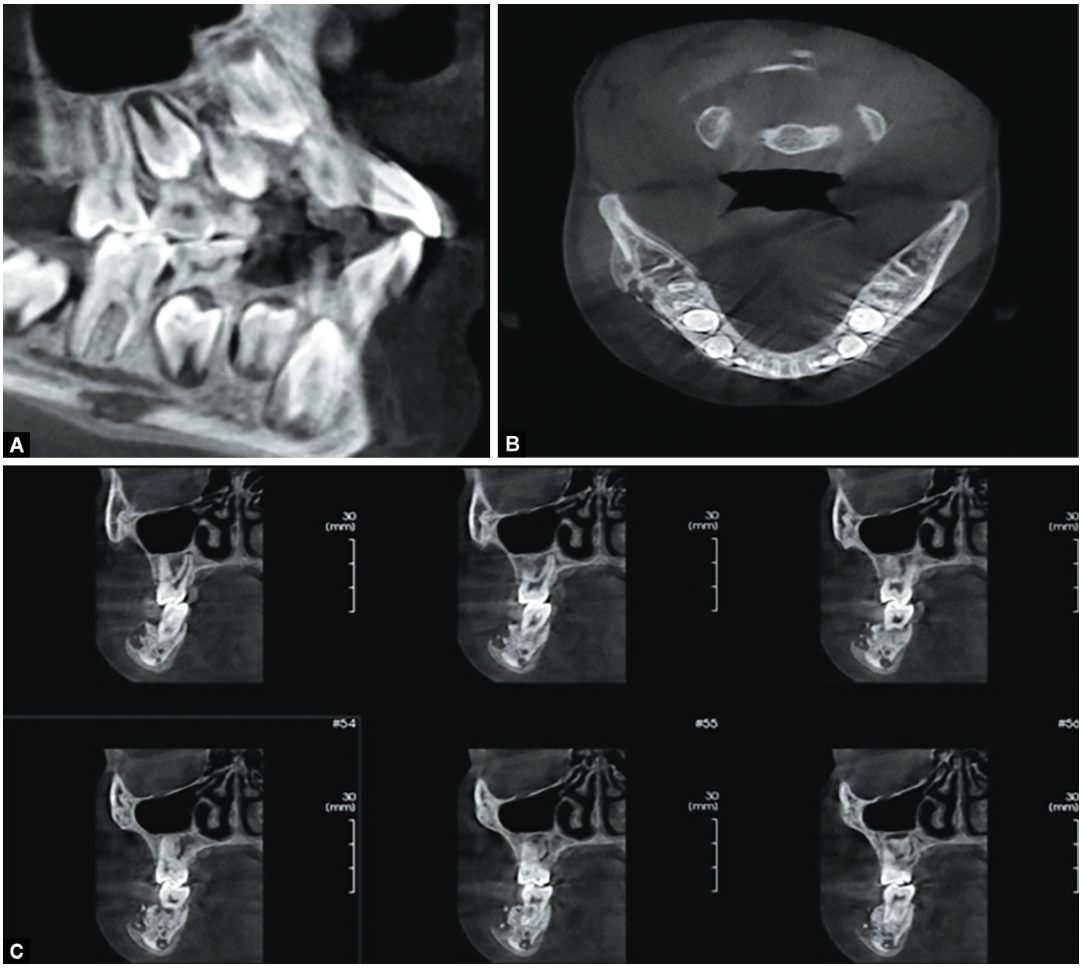
The basic pathogenesis given for the development of COPP is that in very young patients especially those who are below 15 years of age, the periosteum has more osteogenic potential in response to stimulation by an inflammatory process than that of an older patient, during the phase of jaw bone growth. It tends to be stripped with ease. The severity and extent of the symptoms depend on various factors such as the immunity of the host, virulence of the microbe, and the underlying systemic condition of the individual.[4]

Most cases (76%) of COPP arise in the premolar-molar area of the mandible.[7] This is because of poorer blood supply of the mandible when compared with the maxilla and increased thickness of mandibular cortical bone than the maxillary bone. The buccal cortex is commonly involved.[8]

Radiographs revealed the presence of radiopaque bone laminations that are parallel not only to each other but also to the underlying cortical bone. The presence of these laminations suggests a typical radiographic feature such as an "onion skin" appearance. Intraoral periapical, occlusal, and panoramic radiographs are often used for preliminary diagnosis. Computed tomograms show new periosteal proliferation located in successive layers parallel to the condensed cortical bone.[2] Computed tomogram is also helpful in identifying the extent of bone involvement and its relationship with the adjacent anatomical structures. The soft tissue changes seen on the CT scan are swelling of masseter over the affected bone, thickening of overlying skin, stranding of subcutaneous fat, and lymphadenopathy. Gadolinium-enhanced scans show enhancement in the surrounding soft tissues. Radionuclide scan with Tc^{99m} shows uptake of the tracer.[9]



Figs 1A and B: (A) Intraoral view shows obliteration of the right vestibule due to swelling; (B) Orthopantomogram shows localized thinning of the lower border of the mandible



Figs 2A to C: (A and B) Axial and sagittal views show bony lamellae both buccally and lingually; (C) Areas of small sequestra and osteolytic radiolucencies seen within the new bone

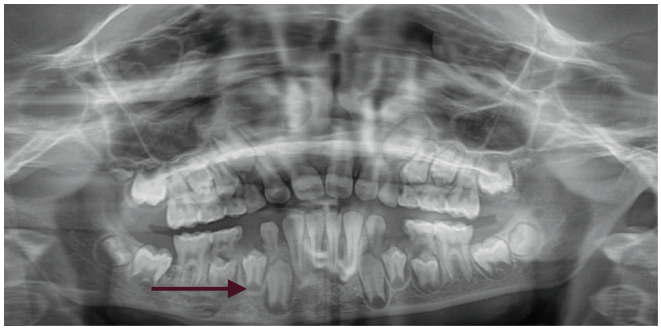


Fig. 3: OPG shows eruption of underlying mandibular right first premolar

In our case, laminations were present in the posterior aspect of the mandible but anteriorly the lamellae were destroyed along with the development of small sequestrate. The gradual dilution of the toxic byproducts and balanced interplay between the host resistance and microbial virulence in the posterior region led to new bone formation. The presence of both destruction and bone formation has made this case unusual.

The absence of fever and suppuration, less response to monotherapy with antibiotics, gradually increasing the size of the lesion, and radiographic features of a loss of the mandibular border and onion skin appearance are consistent with malignancy.

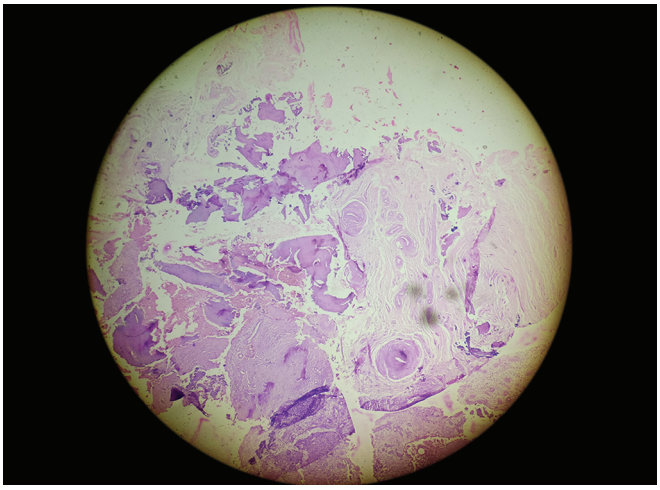


Fig. 4: Photomicrograph shows irregular trabeculae of bone with empty-appearing osteocytic lacunae in a background of collagenous tissue, areas of hemorrhage, and inflammatory cells (H&E, 10×)

Ewing sarcoma, osteosarcoma, Caffey's disease, fibrous dysplasia, hypertrophic osteoarthropathy, syphilitic osteomyelitis, healing fracture callus, calcifying hematoma, and osteoma should be considered in differential diagnosis.[10]

Once the definitive diagnosis is established, the mainstay of treatment in COPP cases is to eliminate the primary source of infection. If an infected tooth is a cause, then extraction of the tooth followed by curettage of the socket is recommended. The surgical debridement is followed by suitable antibiotics and non-steroidal anti-inflammatory drugs. Clinically, resolution of pain and swelling combined with the regain of normal facial appearance is a sign of resolution of the lesion. Radiographically, the return of normal bone morphology determines the complete success of the treatment.

Conclusion

The detailed imaging as well as the three-dimensional reconstruction of the images has helped to reach out to a differential diagnosis in this case.

Clinical Significance

This case report emphasizes the importance of newer techniques such as CBCT in reaching out at a diagnostic conclusion in challenging clinical cases.

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