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### RESEARCH ARTICLE

#### PERIOSTITIS OSSIFICANS: REPORT OF TWO CASES

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#### Abstract

Periostitis ossificans, more popularly known as Garre's osteomyelitis is a distinctive type of chronic osteomyelitis occurring in children and young adults and is commonly associated with odontogenic infection. This paper describes two cases of Periostitis ossificans, one a nine-year-old girl and the other an eleven-year-old boy who presented with a bony hard swelling of the lower jaw. Both of them had an associated decayed permanent mandibular molar tooth. Occlusal radiograph revealed the pathognomonic "onion skin appearance". Extraction of the molar teeth was performed as the patients did not consent for endodontic management due to financial constraints. There was complete remission of the swelling when reviewed after 6 months and later after 7 yrs, showed complete bone remodelling.

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#### Introduction:-

Periostitis ossificans (PO) is a non-suppurative type of chronic osteomyelitis occurring in children and young adults. It is also known as Garre's osteomyelitis and is named after Carl Garre who first described the condition in 1893. He described a sclerosing form of osteomyelitis that caused distension and thickening of bone but no suppuration, sequestration or fistula formation.<sup>1</sup> Although this disease entity is rare in occurrence, it belongs to the group of primary chronic osteomyelitis that accounts for 10.3% of all osteomyelitis of jaws.<sup>2</sup> Mandible is often affected than maxilla.<sup>3</sup> The most common cause of PO is periapical infection of mandibular first molar. A fine balance is maintained between the resistance of the host and the number and virulence of the organisms present, such that infection can continue at a low, chronic stage, invasive enough to stimulate new periosteal bone formation but not severe enough to induce bone resorption.<sup>4,5</sup> Clinically it presents as a bony hard swelling of the jaw with facial asymmetry. Radiographically, PO is characterised by the presence of lamellae of newly formed periosteal bone outside the cortex, giving the characteristic appearance of "onion skin".<sup>6</sup>

#### Case Report 1

A nine-year-old girl reported to the dental outpatient department with a swelling and pain in relation to the left lower molar. Restoration was done on the tooth one year before and she had intermittent pain and swelling since the last 3 months. There was no relevant medical history. On extra oral examination, there was a diffuse swelling of size 1×2 cm on the left side of face. Fig 1. On palpation, it was tender and bony hard in the inferior aspect of the body of the mandible. Intra orally, a temporary restoration was noted on 36. A diffuse swelling was observed on the buccal aspect of gingiva in relation to 36, measuring 0.5×1 cm Fig 2. On palpation it was firm in consistency and mildly

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tender. Intraoral periapical radiograph (IOPAR) of 36 showed evidence of secondary caries. Mandibular lateral occlusal radiograph revealed alternating radiopaque and radiolucent bands on the buccal aspect of mandible extending from 75 to 37 region suggestive of “onion skin” periosteal reaction Fig 3. A final diagnosis of “Garre’s osteomyelitis” of the mandible was made based on the clinical and radiographic findings. The patient was administered a course of antibiotics and anti-inflammatory analgesics. Extraction of 36 was done as the patient did not consent for endodontic management. The swelling subsided after treatment and there was complete remission when reviewed after eight months. There was complete bone remodelling when reviewed after 7 years. Fig 4.

### Case Report 2

A eleven-year-old boy reported to the dental outpatient department with pain and swelling on the right side of face for two weeks. There was no relevant medical history. Clinical examination revealed the presence of a diffuse swelling measuring 1.5×3 cm on the right side of face. Fig 5. On palpation, it was tender and bony hard on the inferior aspect of the mandible. Intraorally deep caries was noted on 46. A diffuse swelling was present on the buccal aspect of the gingiva extending from 85 to 46 region. Fig 6. IOPAR in relation to 46 showed evidence of deep caries involving pulp. Mandibular lateral occlusal radiograph showed alternate radiopaque and radiolucent bands on the buccal aspect of the mandible from 85 to 47 region suggestive of “onion peel” periosteal reaction Fig 7. A final diagnosis of Garre’s osteomyelitis of the mandible was made. Extraction of 46 was done following a course of antibiotics and anti-inflammatory analgesics. The swelling subsided and there was complete remission when reviewed after six months. Complete bone remodelling was noted when reviewed after 7 years. Fig 8.

### Discussion:-

Periostitis ossificans is associated with gross thickening of the periosteum of bones and peripheral reactive bone formation resulting from mild irritation or infection. The term was coined by Gorman in 1951.

Mandible is most often affected than maxilla and it is generally seen at the lower margin of the mandible in the mandibular first molar region. In both of our cases, mandibular first molar was affected. The etiological factors for PO include periapical infection of mandibular molars, periodontal infection, untreated fractures, developing tooth follicle, previous extraction site, pericoronitis, buccal bifurcation cyst, lateral inflammatory odontogenic cyst, non-odontogenic infections and rarely no causative factors could be found.<sup>7</sup> In our cases there was clear evidence of periapical infection from mandibular molars. A previous study showed 82.7% of PO occurring along the lower border of mandible, 43% in buccal cortex and 6.5% in lingual cortex.<sup>6,8</sup> The buccal cortex and the lower border of the body of the mandible was involved in both of our cases.

Radiographically PO presents with the typical ‘onion skin’ appearance appreciated well on occlusal radiographs. It is seen as radiopaque bone laminations that are arranged roughly parallel to each other and to the underlying cortical surface. These laminations vary from one to twelve in number and radiolucent separations are often present between the new bone and the original cortex. Periapical, occlusal and panoramic radiographs are often used for diagnosis and have a high diagnostic value. Computed tomography may also be used and it is accurate for detecting not only typical alterations inside the bone, but also periosteal reactions and soft tissue involvement.<sup>6,9</sup> The classification of radiographic appearance of PO of the mandible by Kawai et al is shown in Table 1. In this system of classification PO has been classified into two types, each with two subtypes based on whether the original contour of mandible is preserved or not. Type 1 lesions are of shorter duration and the original contour of mandible is preserved. Type 2 lesions are of long duration and the original contour of mandible is lost. Both the subtypes of Type 1 PO occur in the early stages of mandibular osteomyelitis. With adequate treatment there can be complete resolution of PO Type 1 cases, however, if the disease continues type 1 may progress to type 2 cases.

Our cases can be grouped in Type 1 subtype 2, both presenting with newly formed bony lamellae on the outer aspect of the cortex with preservation of the original contour of the mandible

There is no need for a biopsy during diagnosis of Garre’s osteomyelitis, except when the cause is unknown.<sup>5,6</sup> As our cases exhibited obvious clinical and radiographic features, a biopsy was not required which is supported by the fact that there was complete resolution of the swelling when the source of infection was eliminated. Other diseases which present with similar features include infantile cortical hyperostosis, hypervitaminosis A, syphilis, leukemia, Ewing’s sarcoma and metastatic neuroblastoma and must be considered in the differential diagnosis.<sup>7</sup>

The treatment procedure involves removal of the source of infection either by extraction or root canal treatment or periodontal treatment as the case may be. Surgical recontouring of the cortical expansion is usually not required as the jaws are gradually remodelled during the growth period.

**Table 1:-** Classification of radiographic appearance of PO of mandible by Kawai etal.

	<b>TYPE I (original contour of mandible is preserved)</b>	<b>TYPE II (original contour of mandible is lost)</b>
<b>Subtype I</b>	Single lamella seen as a radiopaque line of periosteal new bone overlying the original cortex separated by a radiolucent line	Newly formed bony enlargement with resorption of original cortex and osteolytic areas usually visible
<b>Subtype 2</b>	Visible hemi-elliptical newly formed bony enlargement, well outlined with a thin cortical surface located on the outer aspect of original cortex producing an onion skin appearance	Deformation with a homogenously dense osteosclerotic bone that made original cortex discernible. This subtype occasionally showed duplication of newly formed periosteal bone on the outer aspect of the deformed mandible

**Figures**

**Fig.1:-**Extra oral Swelling on left side of face.



Fig.2:- Intraoral swelling in relation to restored 36.



Fig.3:- Onion skin radiographic appearance.



**Fig.4:-** Post treatment complete bone remodeling.



Case No 2

**Fig 5:-** Swelling on the right side of face.



**Fig 6:-** Intraoral swelling in relation to 46.



**Fig.7:-** Onion skin appearance in relation to 46.



**Fig.8:-** Post treatment complete bone remodeling.**Conclusion:-**

This paper emphasizes the importance of recognition and early diagnosis of PO in children and young adults among dentists. PO in young people is curable with early diagnosis and adequate treatment. Successful resolution may be more difficult in severe and long-standing cases of PO.

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