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Periostitis ossificans (Garrè's osteomyelitis) radiographic study of two cases

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Summary. *Background.* Periostitis Ossificans (PO) is a non-suppurative type of Osteomyelitis, commonly occurring in children and young adults, in mandible. The most common cause for PO is periapical infection of mandibular first molar. Radio graphically PO is characterized by the presence of lamellae of newly formed periosteal bone outside the cortex, giving the characteristic appearance of 'onion skin'.

Case reports. Two male children 11 years of age reported to the Department of Oral Medicine with a painless and persistent bony hard swelling in the mandible, with a short duration (Figs 1, 5). Both the patients had grossly decayed mandibular permanent first molar tooth with periapical infection and buccal cortical plate expansion (Figs 2, 6). The radiographic study revealed different appearances, the Orthopantomograph of case I showed a single radiopaque lamella outside the lower cortical border, without altering original mandibular contour (Fig. 3) and in case II showed a newly formed bony enlargement on the outer aspect of the lower cortical border without altering the original madibular contour (Fig. 7). Occlusal radiograph of both the patients showed two distinct radiopaque lamellae of periosteal bone outside the buccal cortex (Figs 4, 8).

Kawai *et al.* classified PO of mandible into type I and type II, based on whether the original contour of mandible is preserved or not. Each type is further classified into two sub types (Table 1). In case I, the orthopantomographic appearance is characteristic of type I-1 (Fig. 3), but the appearance in occlusal radiograph is characteristic of type I-2 (Fig. 4). In case II, the appearances in both the radiographs are characteristic of type I-2 (Figs 7, 8).

Conclusions. Apart from the typical onion skin appearance, PO shows various other radiographic appearances.

The radiographic appearance of Periostitis Ossificans may reflect the duration, progression and the mode of healing of the disease process.

The radiographic classification of PO depends on the type of radiographs taken for evaluation.

Review of literature

Periostitis ossificans (PO) is a type of chronic osteomyelitis, more popularly known as Garrè's osteomyelitis. Other names attributed to PO in the literature are nonsuppurative ossifying periostitis [1], osteomyelitis with proliferative periostitis [2,3], nonsuppurative sclerosing osteomyelitis [4], and chronic sclerosing inflammation of the jaw [5]. Garrè was not responsible for the description of the disease that now bears his name. In his original publication there is no mention of periostitis, periosteal duplication, or onion-skin appearance. Garrè himself had no access to pathologic specimens for microscopic examination. The term 'Garrè's osteomyelitis' as synonymous to PO may therefore be an improper designation and the term periostitis ossificans may be preferred [5,6].

PO is most commonly reported in body of mandible [7]. Most reported cases are unifocal and unilateral, but at least one case involving four quadrants has been reported [8]. Apart from the mandible, the most common bone to be involved in PO is the femur [5].

The cause of inflammatory subperiosteal bone production in PO affecting mandible or maxilla is spread of infection from the nonvital teeth perforating the cortex and becoming attenuated, which in turn stimulates bone formation by the periosteum.

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Table 1. Classification of PO based on the radiographic appearance [10].

	TYPE I (Original contour of mandible preserved)	TYPE II (original contour of mandible is lost)
Subtype 1	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.
Subtype 2	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 homogeneously 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

Table 2. Classification of Gross Periostitis Ossificans – GPO [11].

Type A	Showing an onion skin appearance, resulting from a nonvital tooth or following extraction of a tooth.
Type B	Consolidation form shows fine bony spicules perpendicular to bone surface.
Type C	Consolidation form shows coarse trabeculation with wide marrow spaces.
Type D	Shows more osteosclerotic and osteolytic changes in the affected medullary bone and disappearance of original cortex or loss of the original bone contour.

The periosteal reaction may be in single or multiple layers. The apparent duplication is understood to be a result of periodic exacerbation and remission of the infection, which causes repeated perforation of the outermost new bone with restripping of the periosteum resulting in repetitive layering of bone [9].

The radiographic appearance of PO includes radiopaque laminations that are roughly parallel to each other and to the underlying cortical surface, giving an onion-skin appearance. Laminations vary from 1 to 12 in numbers. Radiolucent separation is present between new bone and original cortex. Within the new bone, areas of small sequestra or osteolytic radiolucencies may also be found [6,9]. There will be patchy increased radiopacity overlying the lesion in cases involving the buccal cortex. This is a result of attenuation of X-ray image by the reactive bone [9].

The classification 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 I lesions are of shorter duration than type II. Both the subtypes of type I PO occurs in the early stages of mandibular osteomyelitis. With adequate treatment there can be complete resolution of PO type I cases; however, if the disease continues, type I-1 may progress to type II-2 and type I-2 to type II-1, followed by type II-2. In type II cases where there has been loss of mandibular contour, deformity remains even when normal bony architecture has been restored and the disease process has been resolved [10].

Mandibular osteomyelitis with bulbous bony enlargement in young patients is referred as gross periostitis ossificans (GPO). Kawai *et al*.'s classification of this more severe form of the disease is shown in Table 2. Type A is associated with carious tooth or followed extraction of tooth, showing onion-skin appearance. In 36·8% of types B and C, no infectious source could be identified; it was suspected that it could be caused by a developing unerupted tooth or a dental follicle. Type D was seen in the chronic stage [11].

When other imaging modalities such as scintigraphy are used in cases of PO, these have shown intense uptake of Tc 99 m methylene diphosphonate at regions of interest. A computerized tomographic study has demonstrated cortical thickening, which appeared as sclerosis and periosteal new bone formation perpendicular to the cortex, which assumed a sunburst appearance [12].

The following case reports describe the occurrence of PO in two paediatric patients, with contrasting radiographic findings.

Case report

Case I

An 11-year-old boy presented with a complaint of swelling on the right side of mandible present for the previous 45 days. Swelling had started following an episode of pain in the carious right lower first molar tooth. Even after treatment using antibiotics and anti-inflammatory agents there had been no

Fig. 1. An 11-year old boy (case 1) with a diffuse swelling of the right side of lower jaw.

Fig. 2. Intraoral view in case 1 showing carious right mandibular first molar with buccal expansion.

significant reduction in the size of the swelling. On examination there was a diffuse swelling of the right side of the body of the mandible. This was bony hard in consistency with mild tenderness. Skin and mucosa over the swelling was normal (Fig. 1).

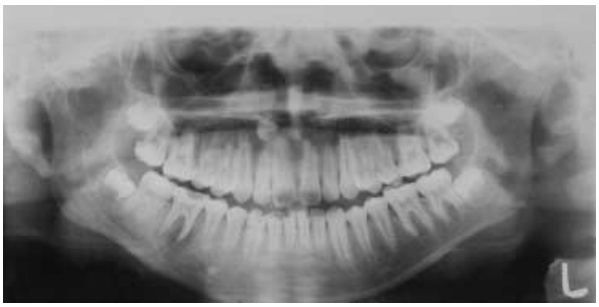

Fig. 3. Orthopantomograph in case 1 showing an ill-defined area of osteosclerosis with a periapical radiolucency of 0·5 cm in right mandibular first molar. Note a single radiopaque lamella below the lower border of mandible, separated by a fine radiolucent line.

Intraoral 46 was grossly carious and there was buccal cortical expansion adjacent to the tooth (Fig. 2). Routine haematological investigations gave normal results. An orthopantomograph of the patient revealed an area of diffuse radiopacity suggestive of osteosclerosis extending from 44 to 47 and associated with a periapical radiolucency of 0·5 cm involving the mesial root of 46. It also revealed a single radiopaque lamella below the lower border of the mandible and separated from the cortex by a fine radiolucent line in the 46 region (Fig. 3). A mandibular lateral occlusal view was taken, which showed a fusiform bony expansion of the buccal aspect of the mandible extending from 46 to 48 regions. Two distinct radiopaque lamellae were visible outside the buccal cortex, with an osteolytic lesion evident on the buccal aspect in the 46 region (Fig. 4). The case was diagnosed as periostitis ossificans. The carious 46 was extracted and the patient was prescribed amoxycillin, metronidazole, and ibuprofen for five days. Review at 1 month showed marked reduction in the size of the swelling.

Case II

An 11-year-old boy with a complaint of painless swelling in the left side of mandible, which had been present for the previous 3 months presented to the oral medicine department of Meenakshi Ammal Dental College & Hospital. On examination there was a localized bony hard swelling of the left side of the body of mandible with thickening of the lower border (Fig. 5). Intraoral examination revealed a temporary restoration in a grossly carious 36. There was also buccal cortical plate expansion, which was not tender (Fig. 6). An orthopantomograph revealed an osteosclerotic area in the body of mandible extending up to the lower border with expansion and

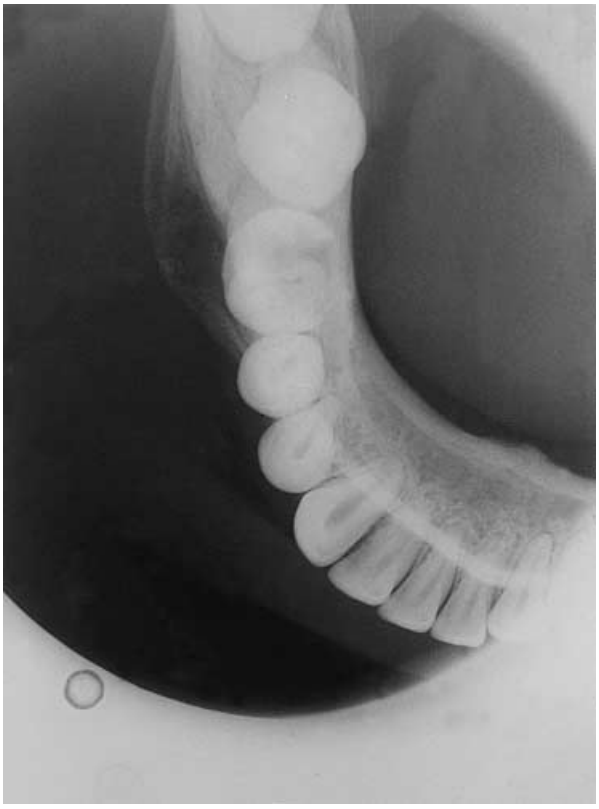

Fig. 4. Occlusal view of the right mandible in case 1 showing fusiform buccal expansion extending from 46 to 48 regions with two radiopaque lamellae. Note the osteolytic area in the new bone in 46 region.

Fig. 5. An 11-year-old boy (case 2) with a localized swelling in the left side of mandible.

bone enlargement on the outer aspect of the original cortex without evidence of alteration of the mandibular contour. There was also a periapical radiolucency in 36 region (Fig. 7). The mandibular lateral occlusal view revealed expansion of buccal cortex extending from 35 to 37 regions together with two distinct radiopaque lamellae. The bony trabecular patterns within the swelling were coarse and wide (Fig. 8). The offending tooth 36 was extracted and patient was prescribed amoxycillin, metronidazole, and ibuprofen for 5 days. As the patient was then lost to follow-up, the final result of treatment is not known for case II.

Fig. 6. Intraoral view in case 2 showing temporary filling in carious left mandibular first molar tooth.

Discussion

Periostitis ossificans (PO) is a type of chronic osteomyelitis, more popularly known as Garrè's osteomyelitis. Gorman coined the term PO in 1951. The mean age of patients with PO has been reported to be 13 years [6,9]. In our two cases the patients were both 11 years old when diagnosed as having PO. There is no sex predilection for PO, although slight predilection in males has been reported, with a M/F ratio of 1·4:1 [9]. Both of our cases were males.

Fig. 7. Orthopantomograph in case 2 showing intense area of osteosclerosis in 36 region extending to lower border and a newly formed bony enlargement on the outer aspect of original cortex without altering the mandibular contour.

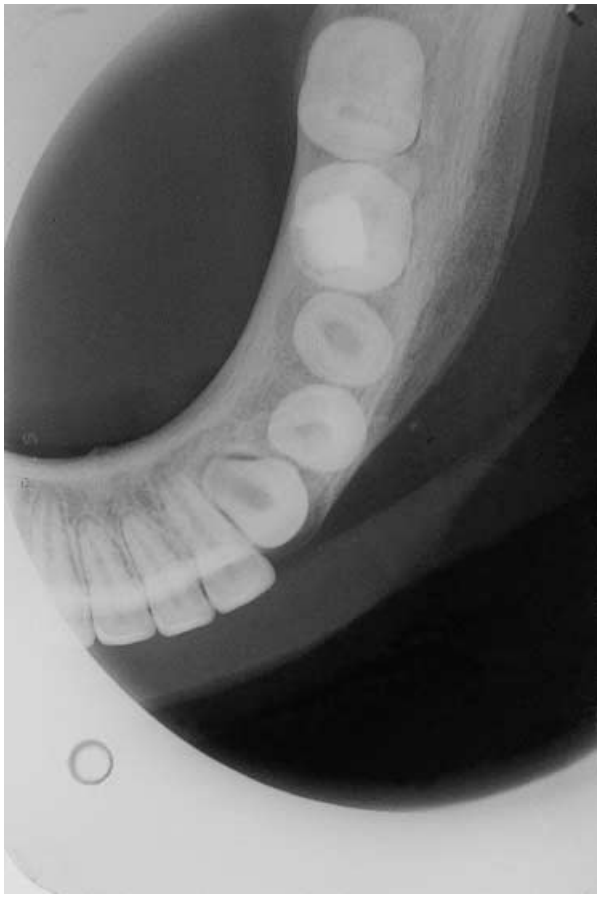

Fig. 8. Occlusal view of the left mandible in case 2 showing buccal expansion with homogenous fine trabecular pattern and with two distinct radiopaque lamellae.

The etiological factors for PO include periapical infection of mandibular molars, periodontal infection, untreated fractures, developing tooth follicle, unerupted teeth, previous extraction site, pericoronitis, buccal bifurcation cyst, lateral inflammatory odontogenic cyst, and nonodontogenic infection [6,7,9–11]. In some cases there may not be any demonstrable aetiological factor and these are termed idiopathic [7]. Of these, the most common aetiological factor is said to be the periapical infection of mandibular first molar [9]. In both of our cases the source of infection for PO was a nonvital carious mandibular first molar.

For unknown reasons PO is most common in the mandible, and has been especially related to the left side of the mandible [12]. A previous study showed 82·7% of periostitis occurs along the lower border of mandible, 43% in buccal cortex, and 6·5% in lingual cortex [9]. In our cases there was involvement of lower border of body of the mandible and the buccal cortex without involvement of the lingual aspect.

Radiographically, both our cases showed evidence of periapical infection in their first mandibular molar. The affected area showed increased radio density, which may be attributed to osteosclerosis. In case I, the orthopantomograph showed a single radiopaque lamella separated from the original lower cortical border of mandible by a fine radiolucent line. As the cortical border was not destroyed and the mandibular contour was unaffected, it may be categorized radiographically as type I-1 (Fig. 3). But occlusal radiographs taken for the same case at the same visit showed fusiform bony mass with two radiopaque lamellae outside the original buccal cortex, giving an onion-skin appearance more characteristic of type I-2 (Fig. 4). This clearly indicates that this radiographic classification of PO may be at least partly dependent on the type of radiograph taken. There may be radiopaque sequestra or osteolytic lesion within PO lesion [6,9]. In our case I, there was an area of osteolytic radiolucency in the periapical region and in the newly formed bone on the buccal aspect of 46 region (Figs 3 and 4).

In case II, the orthopantomograph showed an ill-defined area of uniform osteosclerosis and a bony enlargement on the outer aspect of the original cortex of lower border without alteration of the contour. So, radiographically, this may be considered as type I-2 (Fig. 7). In the occlusal view it showed duplication of cortical lining and homogenously dense osteosclerotic bone (Fig. 8).

Based on the radiographic classification of GPO, both our cases correspond to type A, both showing onion-skin appearance in the occlusal radiograph and being associated with nonvital carious mandibular first molars.

Both of our cases of PO were probably of relatively short duration (3 months and 45 days) and were in the early stages of the disease. The discrepancy in their radiographic appearances may be attributed to the difference in the duration, progression and mode of healing of the disease process in two individuals. Radiological differential diagnosis of PO includes infantile cortical hyperostosis, Ewing's sarcoma, osteosarcoma, etc., none of which fitted the clinical and radiographic features of the two cases reported here.

PO in young persons is, in general, curable with early diagnosis and adequate treatment; however, if the correct diagnosis is delayed by more than 6 months, it may progress into a persistent and deforming stage. Elimination of the source of infection either by extraction or endodontic therapy of the offending tooth may be accompanied by antibiotic therapy in the early stages. Once the cause is removed the bone will remodel itself gradually and the original facial symmetry will be restored. This remodeling of the bone may be helped by the overlying muscle pull, which is attached to it [6]. Successful resolution may be more difficult in severe and long standing cases of PO.

What this paper adds

- Two cases of Periostitis Ossificans (PO) with different radiographic appearances are presented in this article to emphasize that, apart from the typical 'onion skin' appearance, PO may show various other radiographic pattern also.
- The difference in the duration, progression and the mode of healing of the disease process between individuals accounts for the various radiographic appearances of PO.

Why this paper is important for paediatric dentists

- Early diagnosis and elimination of source of infection in cases of PO lead to a complete resolution of the disease process. Successful resolution may be more difficult, if delayed.

Conclusion

1 Apart from the typical and consistent onion-skin appearance, PO shows a variety of other radiographic appearances.

2 The radiographic appearance of periostitis ossificans may reflect the duration, progression and the mode of healing of the disease process.

3 The radiographic classification of PO may depend at least partly on the type of radiographs taken for evaluation.

References

1 Rowe NL, Heslop IH. Periostitis and osteomyelitis of the mandible in childhood. *British Dental Journal* 1957; **103**: 67–78.

2 Garre C. Über besondere formen and folgezustände der akuten infektiösen osteomyelitis. *Beiträge zur Klinischen Chirurgie* 1893; **10**: 241–298.

3 Smith SN, Farman AG. Osteomyelitis with Proliferative Periostitis (Garrè's Osteomyelitis). *Oral Surgery* 1977; **43**: 315–318,.

4 Perriman A, Uthman A. Periostitis Ossificans. *British Journal of Oral Surgery* 1972; **10**: 211–216.

5 Wood RE, Nortjé CJ, Grotepass F, Schmidt S, Harris AMP. Periostitis ossificans versus Garrè's osteomyelitis Part-1. What did Garrè really say? *Oral Surgery Oral Medical Oral Pathology* 1988; **65**: 773–777.

6 Neville BW, Damm DD, Allen CM, Bouquot JE. Pulp and periapical diseases. In: Neville BW (ed.). *Oral and Maxillofacial Pathology*, 1st edn. Pennsylvania: W.B. Saunders and Co., 1995: 118–119.

7 Benca PG, Mostofi R, Kuo PC. Roentgeno-oddities. *Oral Surgery, Oral Medicine, Oral Pathology* 1987; **63**: 258–260.

8 Eisenbud L, Miller J, Roberts IL. Garrè's proliferative periostitis occurring simultaneously in four quadrants of the jaws. *Oral Surgery, Oral Medicine, Oral Pathology* 1981; **51**: 172–178.

9 Nortjé CJ, Wood RE, Grotepass F. Periostitis ossificans versus Garrè's osteomyelitis-part II: radiologic analysis of 93 cases in the jaws. *Oral Surgery, Oral Medicine, Oral Pathology* 1988; **66**: 249–260.

10 Kawai T, Murakami S, Sakuda M, Fuchihata H. Radiographic investigations of mandibular periostitis ossificans in 55 cases. *Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology and Endodontics* 1996; **82**: 704–712.

11 Kawai T, Hiranuma H, Kishino M, Murakami S, Sakuda M, Fachihata H. Gross periostitis ossificans in mandibular osteomyelitis. Review of English literature and radiographic variation. *Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology and Endodontics* 1998; **86**: 376–381.

12 Felsberg GJ, Gore RL, Schweitzer ME, Jui V. Sclerosing osteomyelitis of Garrè (periostitis ossificans). *Oral Surgery, Oral Medicine, Oral Pathology* 1990; **70**: 117–120.