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Peripheral calcifying odontogenic cyst with multinucleated giant cell formation



KEYWORDS

Peripheral calcifying odontogenic cyst; Gingiva; Gorlin cyst

Calcifying odontogenic cyst (COC, also known as Gorlin cyst), which was firstly described by Gorlin in 1962, is a rare odontogenic lesion. COC represents less than 1.0% of all odontogenic tumors and cysts. Most COCs are present as an intraosseous lesion. The extraosseous counterpart accounts for about 10% of total COC cases. We herein reported a rare peripheral COC in a 48-year-old female patient.

The patient complained of a swelling over the buccal gingiva of teeth 37 and 38 for 10 months. Intraoral examination revealed a pinkish, oval-shaped, firm, smoothsurfaced nodule at the buccal gingiva of teeth 37 and 38 (Fig. 1A). Mild tenderness was noted, but no bony expansion was identified. Panoramic and periapical radiographies showed a well-defined oval-shaped radiolucent lesion with a corticated margin over the alveolar crest of teeth 37 and 38 (Fig. 1B, C). No bony expansion was observed in the lower occlusal radiograph (Fig. 1D). Cone beam computed tomography displayed a well-defined bowl shaped bony defect over the buccal side of alveolar crest of teeth 37 and 38 (Fig. 1E). The clinical differential diagnoses included peripheral ossifying fibroma, peripheral odontogenic fibroma, and peripheral ameloblastoma. The lesion was subsequently totally excised under local anesthesia.

Histopathological examination showed a cystic lesion in the lamina propria of the bisected gingival mucosa (Fig. 1F). The cystic lesion was lined by nonkeratinized epithelium with variable thickness and a well-defined basal cell layer of palisading columnar cells (Fig. 1G). The overlying epithelial cells were loosely cohesive, resembling the stellate reticulum of enamel organ with abundant ghost cells in the cystic lumen and the fibrous wall (Fig. 1H). The ghost cells presented in the fibrous wall were surrounded by multinucleated giant cells (Fig. 1I). Some dentinoid calcified materials in the fibrous wall were also discovered. Therefore, the histopathological diagnosis of a peripheral COC was rendered.

The finding of multinucleated giant cells surrounding the ghost cells in the current case, to the best of our knowledge, has possibly been firstly described in the literature. This interesting phenomenon is perhaps due to the foreign body reaction to the ghost cells. Review of English literature, peripheral COC shows an equal distribution in the maxilla and in the mandible with the predilection of anterior part of the jaw bone.² Furthermore, a slight female predilection with the mean age of 48.4-53.8 years is documented.²⁻⁴ This mean age is higher than that in patients with intraosseous COC and is similar to that in patients with gingival cyst or peripheral ameloblastoma.4 On the other hand, despite identical histopathological findings, the origins of the intraosseous and extraosseous COCs are considered to be different. The peripheral counterpart is favored to be originated from epithelial rests of Serre.⁴ Total excision, as shown in this case, is the treatment of choice, and the recurrence of peripheral COC after surgical excision is rare.1

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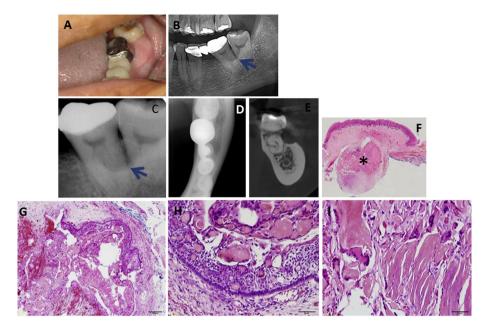


Figure 1 Clinical (A), radiographic (B-E), and microscopic (F-I) pictures of the current case of peripheral calcifying odontogenic cyst. A pink-colored swelling with smooth surface over the buccal side of teeth 37 and 38 (A) was found. Periapical (B) and panoramic (C) radiographs of the lesion revealed a well-defined bowl-shaped bony defect of alveolar crest of teeth 37 and 38 (arrow). No bony expansion was noted in the lower occlusal radiograph (D). Cone beam computed tomography displayed a well-defined bowl shaped bony defect over the buccal side of alveolar crest of teeth 37 and 38 (E). The low-power view showed a cystic lesion (asterisk) in the lamina propria of gingival mucosa (F, magnification, $2\times$). Histopathological microphotographs of the lesion exhibited a cystic lesion lined by the epithelium with the basal cell layer consisting of palisading columnar cells (G, magnification, $40\times$) and the overlying epithelial cells resembling the stellate reticulum of the enamel organ (H, magnification, $200\times$). Abundant ghost cells are present in the lumen and epithelium (H, magnification, $200\times$). The ghost cells presented in the fibrous wall were surrounded by multinucleated giant cells (I, magnification, $200\times$).

Conflicts of interest

The authors have no conflicts of interest relevant to this article.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.jds.2019.01.003.

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