Peripheral Ameloblastoma in a 7 Year old child: A rare case report

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Citation

ABSTRACT
Peripheral ameloblastoma (PA) is a rare, benign, soft tissue odontogenic tumor that is confined to gingiva or alveolar mucosa. Peripheral ameloblastoma signifies the similar histological characteristics as of intraosseous ameloblastoma, though it is less aggressive than this typical subtype. Although the recurrence rate is low, it needs a close and a longer follow-up. Following case report of PA in the right posterior mandible in a 7 years old child, highlighting the importance of histological examination to the diagnosis.

Keywords: Peripheral ameloblastoma, Posterior mandible, extraosseous.

1. INTRODUCTION
Peripheral ameloblastoma (PA) is an odontogenic extraosseous, rare, benign, and soft tissue tumour that was first reported within the literature by Stanley and Krogh in 1959 (Curran, 2004). It comprises 1- 4% of all ameloblastomas. The clinical appearance of PA
may vary, but most of the time, it presents clinically as a slow growing, firm, painless mass with a sessile or pedunculated base with a smooth surface and a normal mucosa colour (Curran, 2004; Riechart & philipsen, 2004). Although PA is limited to the gingiva or the alveolar mucosa, it may be a reason for depression of the underlying bone or reveal a “cupping” effect due to the pressure resorption (Curran, 2004). This case report aims to show the clinical case of peripheral ameloblastoma that occurred in the mandibular right posterior gingiva in a paediatric age group.

2. CASE REPORT

A 7 years old male patient presented to the department of Oral Medicine and Radiology, Sharad Pawar Dental College Wardha, with the complain of painless growth on the gingiva in the right posterior region of jaw since around one and half months. On extra-oral examination, a single right submandibular lymphnode was palpable of size 1x1cm approx., firm in consistency, mobile and tender in nature. The intraoral examination revealed a nodular, sessile growth which was pale pink in colour, with granular surface and indentations of maxillary tooth and soft in consistency which was located in right posterior region of mandible in relation with 44, 45 region measuring approximately 2x2cm in size (fig. 1).

![Figure 1](image1.jpg)

**Figure 1** Nodular, sessile growth with granular surface in relation with 84, 45 region

There was grade I mobility with 74 and occlusal caries with 46 and the clinical provisional diagnosis was given as pyogenic granuloma with the clinical differential diagnosis of peripheral giant cell granuloma and peripheral ossifying fibroma. After clinical examination, radiological investigation was done in which panoramic projection revealed developing 45 with loss of follicular crypt (fig. 2).

![Figure 2](image2.jpg)

**Figure 2** Panoramic projection reveals soft tissue shadow of the lesion with developing 45 and loss of follicular crypt of 45.

The clinico-radiological provisional diagnosis was given as peripheral ossifying fibroma with differential diagnosis of peripheral giant cell granuloma and peripheral ossifying fibroma. On suspecting a soft tissue tumour or a peripheral odontogenic tumour, an excisional biopsy was performed (fig.3).

![Figure 3](image3.jpg)
Figure 3 Complete excision of the lesion

The specimen was fixed in 10% formalin buffer and it was sent to the Oral Pathology Department for histopathological correlation. Histopathological investigation of the specimen revealed the basal cell layer of odontogenic epithelium showed tall columnar cell with palisading hyperchromatic nuclei, subnucleolar vacuolization and scanty eosinophilic cytoplasm. The suprabasal cell layer showed interconnecting cells with centrally placed basophilic nuclei suggestive of stellate reticulum like cells (Fig 4).

Figure 4 H&E Tissue section shows odontogenic epithelium with stellate reticulum like cells

On correlating radiopathologic features the final diagnosis of peripheral plexiform ameloblastoma was given. Lingual bar extension was placed in the 45 region for space maintenance and the patient was kept on a regular follow-up for 1 year (Fig. 5). There was no recurrence and patient had no complaints.

Figure 5 6 months clinical follow-up
3. DISCUSSION

Peripheral ameloblastoma was primarily reported in 1949 whereas the first completely documented case was reported by Stanley and Krough in 1959 (Curran, 2004). Peripheral ameloblastoma is an odontogenic tumor that holds the same histologic characteristics as that of an intraosseous ameloblastoma occurring solely in the soft tissues covering the tooth-bearing areas of the jaws (Curran, 2004; Riechart & Philipsen, 2004; Gardner, 1977). Most likely it arises from the rests of Serres which are fragmentations of the dental lamina that occur during the morpho-differentiation stage of the tooth development (Riechart & Philipsen, 2004; Gardner, 1977; Worth, 1963).

Alternately, the tumor may arise from the surface epithelium that has the potential for production of odontogenic epithelium. The endurance between the tumor and the surface epithelium is incidental and simply represents union of the underlying tumor with the surface epithelium (Neville & Damm, 2002; Rajendran & Sivapathasundharam, 2012; Beena et al., 2012). Regardless of central or peripheral ameloblastoma, they lack CK7, CK8, CK10, CK18, CK20 and epithelial membrane antigen (Isomura et al., 2009; Molina et al., 2010; Rosario et al., 2001). Immunohistochemically, PA exhibited affirmative reactivity for AE1/AE3, KL1,34, E12, and MNF116 cytokeratin and negative staining for CK8, CK10, CK13, CK17, and CK18, which was in harmony with the interpretations seen in human enamel organ. In a review, Philipsen et al. reported 160 cases of PAs documented in literature that the age range varied from 9 to 92 years with an average of 52 years (Philipsen et al., 2001). It has been reported that although the most common sites of occurrence are the premolar or anterior regions of the mandible, but there is an evidence of unusual site like buccal mucosa (Isomura et al., 2012), base of tongue (Rajesh et al., 1998), and floor of the mouth (Ramnarayan et al., 1985) also being affected. PA generally occurs as a painless, sessile, firm, granular or pebbly, exophytic mass of the gingiva. Because of their location, peripheral ameloblastomas may be exposed to local trauma because of the occlusion between the mass and maxillary tooth which is responsible for ulceration or keratosis on the growth as it was present in this case (Curran, 2004). Reactive extraosseous lesions must be included in the differential diagnosis such as peripheral giant cell granuloma, peripheral odontogenic fibroma, peripheral ossifying fibroma, papilloma, pyogenic granuloma, epulis, and fibroma because of its benign and extraosseous nature (Beena et al., 2012). There is no radiographic or surgical evidence of bony involvement, while “cupping” or “saucerization” of the associated bone may be seen (Curran, 2004). PA and intraosseous ameloblastoma may exhibit identical various histological patterns (Beena et al., 2012). The histopathological features in our case showed odontogenic epithelium with stellate reticulum like cells which is a characteristic of plexiform ameloblastoma (Chauhan & Guruprasad, 2011). The current treatment of choice for the PA is complete surgical excision with attainment to complete tissue. Resection of the lesion and adjuvant cancer therapy cannot be advised because it might get considered as an exaggerated treatment (Ülker et al., 2020). Although the recurrence rate of PA is much lower (16% - 19%) than intraosseous ameloblastoma, a continuous and long-term follow up is required because it was stated that considering a non threatening looking peripheral ameloblastoma was recoated as an ameloblastic carcinoma (Baden et al., 1993). Furthermore PA that metastates and a recurrence of a peripheral ameloblastoma which showed dysplastic features was also reported (Lin et al., 1987; Wettan et al., 2001). This remains the importance and necessity of long term regular controls.

4. CONCLUSION

Peripheral plexiform ameloblastoma is the rare benign soft tissue tumour which morpho-differentiation stage of tooth development Clinical presentation could be deceptive. Common lesions should be kept on priority in differential diagnosis however the possibility of uncommon lesions cannot be ruled out. In every soft tissue lesion take an incisonal biopsy. Strict long term follow-up and periodic radiographic assessment should be carried out.

Conflict of Interest

The authors declare that they have no conflict of interest.

Informed Consent

Appropriate signed consent was taken from the patient before writing this case report. Identity of the patient was not revealed in this case report.

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Data and materials availability

All data associated with this study are present in the paper.
REFERENCES AND NOTES


