Peripheral ameloblastic fibroma: a rare case report

S. Leuci[1], N. Coppola[1*], M. D. Mignogna[1], A. E. Di Lauro[1]

[1]*Department of Neurosciences, Reproductive and Odontostomatological Sciences, Oral Medicine Unit, University of Naples Federico II, Via Pansini no. 5, Naples, 80131, Italy*

**Corresponding Author*

Email: noemi.coppola@unina.it

DOI 10.23804/ejpd.2023.1572

Abstract

Ameloblastic fibroma is a rare mixed odontogenic tumour which involves mostly the posterior mandible. Its peripheral variant is very rare. Only eight cases have been reported worldwide. In this report, we described a case of peripheral ameloblastic fibroma occurring in the maxillary gingiva in a 10 year-old child. The lesion was excised with a conservative surgical approach and no recurrence has occurred. Peripheral ameloblastic fibroma should be considered in the differential diagnosis of a slow growing lesion involving the gingiva.

KEYWORDS Ameloblastic fibroma, peripheral ameloblastic fibroma, odontogenic tumor, case report

Introduction

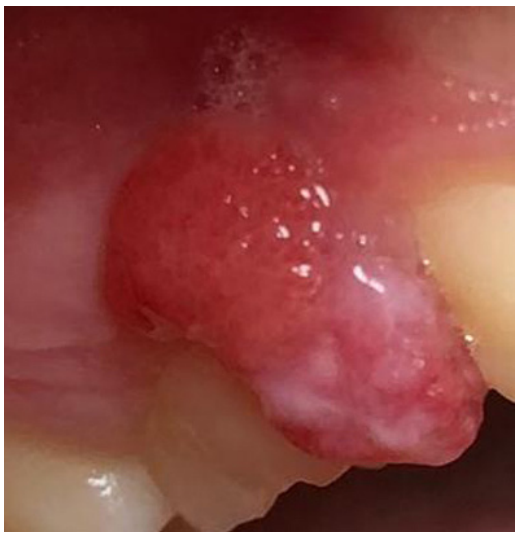
Peripheral odontogenic tumours are rarer than their central variant and histologically similar to these. Clinically, they may have morphology similar to other soft tissues malignancies [Bajpai et al., 2018]. The extraosseous form of ameloblastic fibroma is an exceedingly rare lesion. Only 8 cases were reported worldwide, two of which are in the Japanese literature [Bajpai et al., 2018]. We described a case of peripheral ameloblastic fibroma occurring in 10-year-old children.

Case description

A 10-year-old child complaining of an asymptomatic swelling in the right upper molar region was referred to our Oral Medicine Unit. The patient reported that the lesion had been present for 6 months with a progressive increase in size. His medical history and family history were unremarkable and laboratory tests were within standard limits. On clinical oral examination a sessile polypoid lesion with papillary erythematous surface on the marginal gingiva was observed in the right maxillary between first and second deciduous molar. The lesion measured about 0,8 mm and was tender to palpation, without cervical lymphadenopathy (Fig. 1). Radiographic examination revealed no bone involvement (Fig. 2). Based on clinical and instrumental findings, the main diagnostic hypotheses were pyogenic granuloma, hemangioma, papilloma, peripheral odontogenic fibroma and malignancies. An incisional biopsy was made, and the specimen was sent for histopathological evaluation. Microscopically sections showed an inactive-looking odontogenic epithelium in the form of nests/islands associated with connective tissue of variable cellularity and no inductive changes were present (100x magnification) (Fig. 3). Based on clinical and microscopic findings, the final diagnosis was peripheral ameloblastic fibroma, a benign odontogenic tumour. A radical surgical excision was made, histopathological examination again confirmed the diagnosis of peripheral ameloblastic fibroma, and all margins were free of neoplasm. No recurrences were observed after 12 months, and the patient will continue the follow up every six months.

Discussion

Ameloblastic fibroma is a rare, benign, mixed odontogenic tumour with odontogenic epithelium in an ectomesenchymal connective tissue similar to dental papilla, without dentin or enamel [Darling and Daley, 2006]. It represents 2% of all odontogenic tumors. The mean age of occurance is 15 years, ranging between (0.5-62 years) and a slight male predominance was reported [Kalantari et al., 2016]. In more than 80% of cases the mandible is involved, mostly in the posterior area [McGuinness et al., 2001]. Its extraosseous (peripheral) variant is very rare and was not described in the odontogenic tumour literature until 1991. Peripheral ameloblastic fibroma share the same histopathologic features of its central (intraosseous) counterpart consisting of both odontogenic epithelial and ectomesenchymal components without the formation of dentin or enamel [Darling



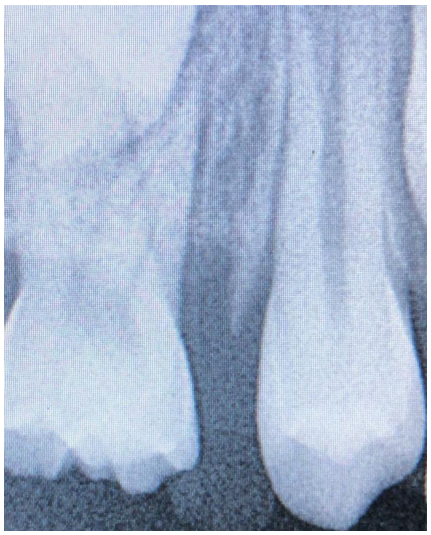


FIG. 1 Clinical pictures showing intraoral soft tissue growth.

FIG. 2 Radiograph of the right maxilla showing no bone involvement.

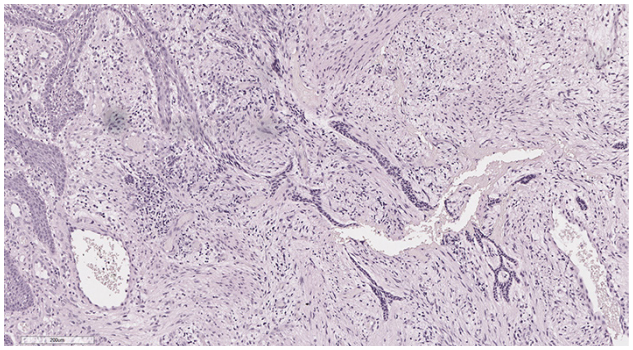


FIG. 3 Photomicrograph with hematoxylin and eosin staining, 100x magnification, showing nests/islands of odontogenic epithelium with connective tissue.

and Daley, 2006]. The main clinical differential diagnoses are represented by epulis, pyogenic granuloma, peripheral giant cell granuloma, localised juvenile spongiotic gingival hyperplasia, focal fibrous hyperplasia, peripheral odontogenic fibroma and other peripheral odontogenic tumors [Bajpai et al., 2018; Decani et al., 2021]. For a correct diagnosis it is necessary to distinguish histologically the peripheral ameloblastic fibroma from the peripheral odontogenic fibroma. In fact, on histological examination peripheral odontogenic fibroma shows a myxoid, fibroblastic or hyalinised stroma and odontogenic epithelium may be absent or inactive. Instead in the peripheral ameloblastic fibroma epithelial component with cords and islands of odontogenic epithelium with central stellate reticulum- like areas is prominent and myxoid stroma is present to a lesser extent [Langer et al., 2015].

Conservative surgical excision is the treatment of choice for peripheral ameloblastic fibroma and no recurrences were reported.

Other intraosseous odontogenic tumours, such as odontomas, or enamel defects are more common in the juvenile population compared to peripheral tumours such as the one described here (Marra et al., 2021; Borrego-Martí et al., 2021).

Therefore, the clinicians should be aware that peripheral ameloblastic fibroma clinically may mimic a wide range of entities and should be considered when a patient showed a slow growing lesion involving the gingiva.

Funding Source
This article has no funding source.

Conflicts of Interest
The authors have no conflict of interest to declare.

Acknowledgements
All authors gave final approval and agreed to be accountable for all aspects of the work in ensuring that questions relating to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

References

› Bajpai M, Pardhe N, Vijay P. Peripheral Ameloblastic Fibroma of Anterior Maxillary Gingiva Simulating Pyogenic Granuloma. J Coll Physicians Surg Pak. 2018;28(7):575. doi:10.29271/jcpsp.2018.07.575
› Darling MR, Daley TD. Peripheral ameloblastic fibroma. J Oral Pathol Med. 2006;35(3):190-192. doi:10.1111/j.1600-0714.2006.00353.x
› Kalantari M, Samieirad S, Kalantari P. Peripheral Ameloblastic Fibroma: Report of a Rare Case. J Dent (Shiraz). 2016;17(4):367-369.
› McGuinness NJ, Faughnan T, Bennani F, Connolly CE. Ameloblastic fibroma of the anterior maxilla presenting as a complication of tooth eruption: a case report. J Orthod. 2001;28(2):115-118. doi:10.1093/ortho/28.2.115
› Decani S, Lodi G, Sardella A, Varoni EM. Localised juvenile spongiotic gingival hyperplasia: A case of spontaneous resolution and a literature review. Eur J Paediatr Dent. 2021 Jun;22(2):159-162. doi: 10.23804/ejpd.2021.22.02.14. PMID: 34238009.
› Langer S, Choudhury M, Agarwal S, Mehra P. Congenital peripheral ameloblastic fibroma with intraosseous involvement in a 2-week-old infant: A case report with review of literature. J Indian Soc Pedod Prev Dent. 2015;33(4):351-354,. doi:10.4103/0970-4388.165721
› Marra PM, Nucci L, Itro A, Santoro R, Marra A, Perillo L, Grassia V. Prevalence of retained/transmigrated permanent and persistence of primary teeth associated with odontomas in young children. Eur J Paediatr Dent. 2021 Sep;22(3):215-218. doi: 10.23804/ejpd.2021.22.03.7. PMID: 34544250.
› Borrego-Martí N, Peris-Corominas R, Maura-Solivellas I, Ferrés-Padró E, Ferrés-Amat E. Hypomineralisation of second primary molars and primary canines: Prevalence and description of lesions in a population of 153 patients visited at a hospital paediatric dentistry service. Eur J Paediatr Dent. 2021 Sep;22(3):237-242. doi: 10.23804/ejpd.2021.22.03.11. PMID: 34544254.