J Oral Pathol Med (2006) 35: 190–2

www.blackwellmunksgaard.com/jopm

CASE REPORT

Peripheral ameloblastic fibroma

M. R. Darling, T. D. Daley

Division of Oral Pathology, Department of Pathology, Faculty of Medicine and Dentistry, University of Western Ontario, London, ON, Canada

Peripheral ameloblastic fibroma is an exceedingly rare lesion. Only three reports could be found, two of which appeared in the Japanese literature. Here, we report a case of peripheral ameloblastic fibroma occurring in a 5-year-old girl. The diagnosis was made after careful microscopic examination, to exclude other lesions. The lesion was excised and has not recurred 1 year after removal.
J Oral Pathol Med (2006) **35**: 190–2

Keywords: ameloblastic fibroma; gingiva; peripheral

An otherwise healthy 5-year-old female presented with a localized, ulcerated enlargement of the attached gingiva in the right maxillary second deciduous molar region. The second molar was erupted through the alveolar bone crest but not through soft tissue, and was tilted slightly mesially, causing some resorption of the distal root of the first primary molar (Fig. 1). No intrabony radiolucency was present. On the radiograph, small focal radiopacities could be seen overlying the pulp chamber of the adjacent first primary molar (Fig. 1), but these were interpreted to be artefact because they did not correspond to the site of the lesion. The lesion had been present for approximately 2 months.

The lesion was excised and examined microscopically. It was composed of oedematous fibrous connective tissue, with foci of more primitive appearing myxoid connective tissue resembling dental papilla. The tissue contained cords and islands of odontogenic epithelium with central stellate reticulum-like areas. The cords demonstrated thickening of their terminal ends. Focal fusion of the odontogenic epithelial islands with the overlying surface epithelium was seen. Areas of eosinophilic, less cell-rich hyalinization without calcifications were seen in the lesional connective tissue, mostly away from areas of epithelial proliferation. No inductive change was present. A diagnosis of peripheral ameloblastic fibroma was reached (Figs 2 and 3). No recurrence has been reported 1 year after the excision.

Correspondence: Mark R. Darling, Assistant Professor, Division of Oral Pathology, Department of Pathology, Faculty of Medicine and Dentistry, University of Western Ontario, London, ON N6A 5C1, Canada. Tel.: (091 519) 661 211 (ext 86399). Fax: (091 519) 850 2926. E-mail: mark.darling@fmd.uwo.ca
Accepted for publication June 8, 2005

Comments

Ameloblastic fibroma is a mixed intraosseous tumour of odontogenic origin; a true biphasic tumour consisting of proliferating odontogenic epithelium in ectomesenchymal tissue that resembles the dental papilla. Both epithelial and mesenchymal tissues are neoplastic (1–3). It is rare; the most recently published figures indicate that it represents 1.5–4.5% of odontogenic tumours with a slight male predilection in the ratio of 1.4:1 (4). The mean age of occurrence is 14 years. More than 80% are found in the mandible, and 88% within the posterior regions (1, 2). Areas of hyalinization within the sparsely cellular regions of the connective tissue have been described, which are not necessarily in close association with the epithelium (1, 5). The hyalinization could represent abortive dentinoid. However, in their study, Sapp and Jensvold (5) found that this type of hyalinization was commonly seen in ameloblastic fibromas and was insufficient to alter the diagnosis (5). The epithelial component is composed of islands of cells with preameloblasts and stellate reticulum and resembles early enamel organ development. It would not be unusual for the epithelial component in a peripheral ameloblastic fibroma to fuse with the overlying surface epithelium, a phenomenon that may be seen in peripheral odontogenic tumours (1).

Even more rare is the peripheral ameloblastic fibroma, of which only three reports could be traced, two of which appeared in the Japanese literature [cited by Ide et al. (6); Table 1] (6, 7). It presents as a localized gingival enlargement or epulis. One of these cases (7) is disputed, and is thought possibly to be a case of peripheral odontogenic fibroma (8). Too few cases have been reported to demonstrate trends in gender, site and age predilections. The lesion may be associated with delayed eruption of teeth as illustrated in the case presented here. A clinical differential diagnosis should include focal fibrous hyperplasia (irritation fibroma, fibroepithelial polyp), pyogenic granuloma, peripheral ossifying fibroma, peripheral giant cell granuloma,

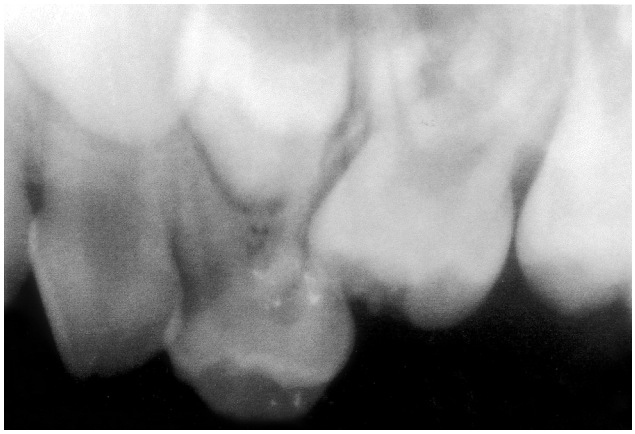

Figure 1 Radiograph of right maxilla, showing deciduous molars.

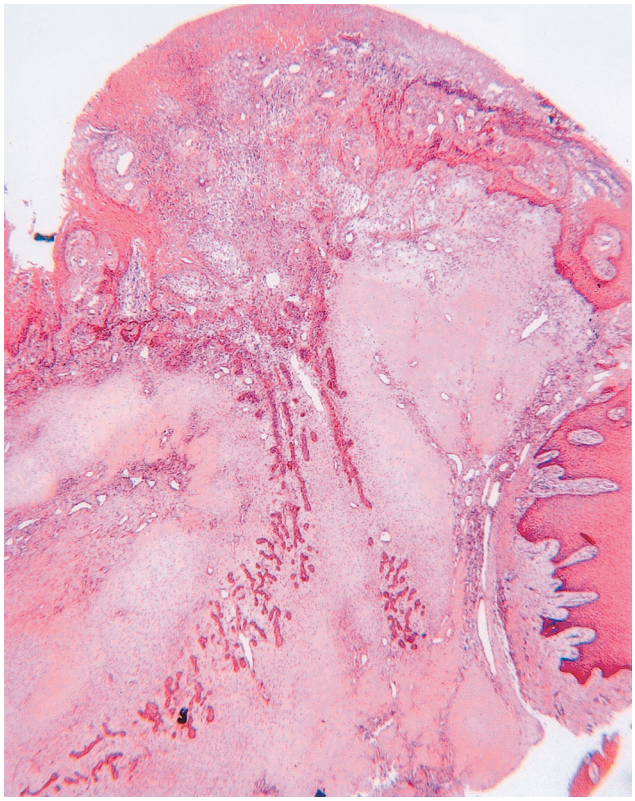

Figure 2 Peripheral ameloblastic fibroma, with proliferating odontogenic epithelium in a primitive appearing myxoid stroma, and showing gingival surface ulceration (original magnification ×25).

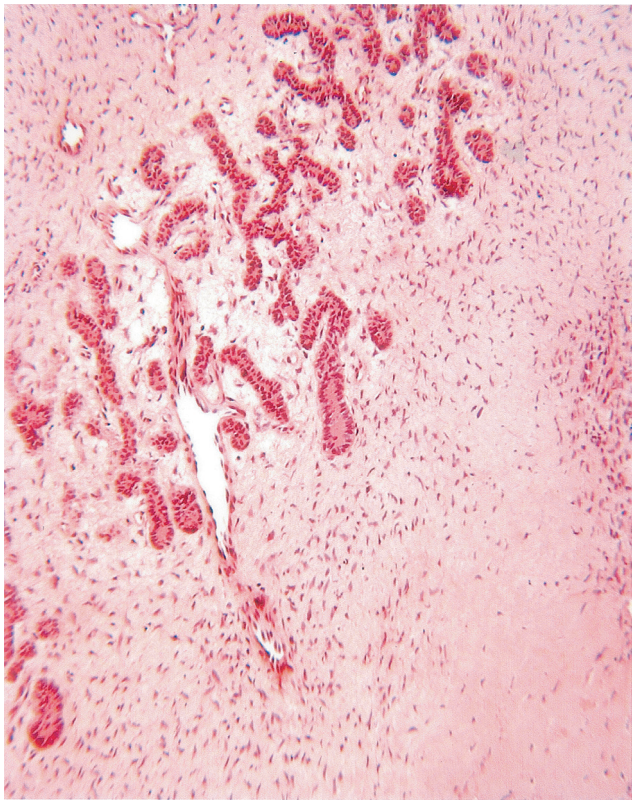

Figure 3 Peripheral ameloblastic fibroma, showing proliferating odontogenic epithelium in a primitive appearing ectomesenchymal stroma (original magnification ×100).

Table 1 Published reports of peripheral ameloblastic fibroma

Author	*Age (years)*	*Gender*	*Site*
Nakamura et al. (1980)[1]	2	M	Posterior mandibular alveolus
Harada et al. (1992)[1]	1	F	Mandibular molar gingiva
Kusama et al. (7)	40	F	Mandibular right premolar gingiva
Darling and Daley (2005)[2]	5	F	Maxillary attached gingiva (right premolar/ deciduous molar region)

[1]Cited by Ide *et al.* (2000).
[2]Darling MR, Daley TD. Peripheral ameloblastomic fibroma. *J Oral Pathol Med.*

peripheral ameloblastoma and other peripheral odontogenic tumours as well as numerous soft tissue tumours.

The peripheral ameloblastic fibroma must be distinguished histologically from the peripheral odontogenic fibroma (1, 3). The latter presents in a wide age range of occurrence, with a slight male predilection (1, 2). The attached gingivae of the mandible and maxilla are equally involved. Histologically, the lesion is identical to the central odontogenic fibroma, which presents with a range of features varying from myxoid, fibroblastic, to densely hyalinized. Odontogenic epithelium may be absent, scanty and inactive appearing, or easily discernible forming cords and nests throughout the lesion, osteoid may also be noted (1).

The peripheral ameloblastic fibroma exhibits prominent cords and islands of epithelium with central areas resembling stellate reticulum, against a pale staining myxoid stroma which resembles dental papilla, as opposed to the tissue morphology seen in odontogenic fibromas. The epithelial islands are reminiscent of the early stages of enamel organ development (1).

In conclusion, it is important to note that peripheral ameloblastic fibromas do occur, and must be separated

from other peripheral odontogenic tumours in a clinical and histological differential diagnosis. Excision results in a cure with no recurrence.

References

1. Sciubba JJ, Fantasia JE, Kahn LB. *Atlas of tumour pathology: tumours and cysts of the jaw*. Washington, DC, USA: Armed Forces Institute of Pathology, 2001; 108–17.
2. Sapp JP, Eversole LR, Wysocki GP. *Contemporary oral and maxillofacial pathology*, 2nd edn. St Louis, USA: Mosby, 2004; 155–6.
3. Takeda Y. Ameloblastic fibroma and related lesions: current pathologic concept. *Oral Oncol* 1999; **35**: 535–40.
4. Philipsen HP, Reichart PA, Praetorius F. Mixed odontogenic tumours and odontomas. Considerations on interrelationship. Review of the literature and presentation of 134 new cases of odontomas. *Oral Oncol* 1997; **33**: 86–99.
5. Sapp JP, Jensvold J. The distribution and morphologic variation of hyaline deposits in odontogenic lesions. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1983; **55**: 151–61.
6. Ide F, Shimoyama T, Horie N. Peripheral ameloblastic fibroma. *Oral Oncol* 2000; **36**: 308.
7. Kusama K, Miyake M, Moro I. Peripheral ameloblastic fibroma of the mandible: report of a case. *J Oral Maxillofac Surg* 1998; **56**: 399–401.
8. Shimoyama T, Horie N, Ide F. Clarification of diagnostic criteria for ameloblastic fibroma. *J Oral Maxillofac Surg* 1999; **57**: 219.