

Odontoameloblastoma: Report of a Case

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Abstract

Odontoameloblastoma is a very rare odontogenic tumour that is similar to ameloblastoma in its locally aggressive behaviour. Its clinical presentation, however, often mimics the more innocuous odontoma, and hence the recognition of its aggressive nature is commonly only ascertained after its histopathologic diagnosis following enucleation. This paper presents a case of odontoameloblastoma. Some of the difficulties that may be encountered in the diagnosis and treatment planning of odontoameloblastomas are discussed.

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Key words: Aggressive, Difficulties in diagnosis and treatment planning, Very rare

Introduction

Ameloblastoma is a benign but locally invasive tumour that affects the jaws.¹ Although it is commonly regarded as a homogeneous group of tumours, detailed investigations have proven its clinicopathologic diversity.² Infrequent cases of dental hard tissue induction by the ameloblastic epithelium have been reported. This simultaneous occurrence of an ameloblastoma and an odontome in the same tumour mass has been named odontoameloblastoma. Odontoameloblastoma is defined by the WHO³ as "a very rare neoplasm characterised by the presence of enamel, dentine and an odontogenic epithelium resembling that of an ameloblastoma, both in structure and behaviour". Although more than 40 cases have so far been reported in the literature, only a few have truly met the histologic and clinical criteria of an odontoameloblastoma.

Case Report

The patient was a 12-year-old Chinese girl whose mother noted a painless progressively enlarging swelling over her left paranasal region of about 2 months duration. Clinical examination revealed a non-tender diffuse swelling at the left paranasal region which lifted the left alar of the nose. The right submandibular lymph node was palpable, mobile and non-tender. Intraorally, there was a fluctuant, non-tender swelling in the buccal sulcus from the upper right central incisor to the upper left canine region. Neighbouring teeth were all responsive to electric pulp testing. The upper left central and lateral incisors had increased mobility. Needle aspiration of the swelling yielded a straw-coloured fluid with

shimmering crystals.

The orthopantomogram (Fig. 1) and anterior maxillary occlusal film (Fig. 2) showed a well-defined radiolucency with scalloped borders of about 3 cm diameter with a radiopaque mass at the left anterior maxilla, crossing the midline. A buried upper left canine and an impacted upper left first premolar were also noted adjacent to the lesion.

A preoperative diagnosis of an odontogenic cyst associated with an odontome was made.

Under general anaesthesia, a buccal mucoperiosteal flap was raised. Thin buccal bone overlying the lesion was removed with rongeurs. The lesion with a hard whitish mass within was enucleated *in toto*. The upper left canine and first premolar were elevated. The bony cavity walls were scored with a rosehead bur. The operation site was irrigated with copious saline, haemostasis was achieved and Carnoy's solution was applied to the defect. The wound was closed using 3.0 black silk sutures. The patient was discharged on the same day.

The gross specimen measured 3 x 1.5 x 1 cm. It consisted of a soft tissue lining enclosing an ovoid-shaped hard tissue mass. On microscopic examination, the hard tissue of the specimen was composed of haphazardly arranged masses of dentine, enamel, cementum and woven bone with little or no intervening stroma (Figs. 3 & 4). This was encased in fibrous connective tissue free of inflammation. Lining the connective tissue was an odontogenic epithelium which was, in areas, keratinised. The epithelium was made up of a polarised basal layer surmounted by polyhedral and stellate cells which, in

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Fig. 1. Orthopantomogram showing the mixed radiolucent and radiopaque lesion in the left anterior maxilla and the adjacent buried upper left canine and impacted upper left first premolar.

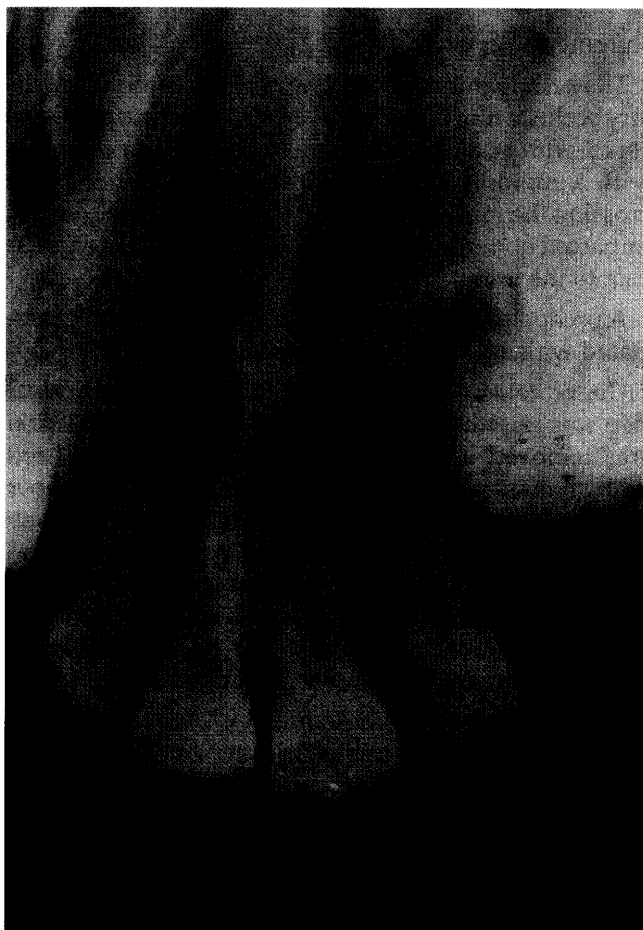


Fig. 2. Anterior maxillary occlusal film showing part of the lesion.

areas, were arranged in whorls and eddies (Fig. 5). The lumen contained aggregations of ghost cells (Fig. 6). The histopathologic diagnosis was an odontoameloblastoma.

Postoperatively, the upper left lateral incisor was noted to be non-vital and it was subsequently root-canal treated. The postoperative recovery was otherwise uneventful.

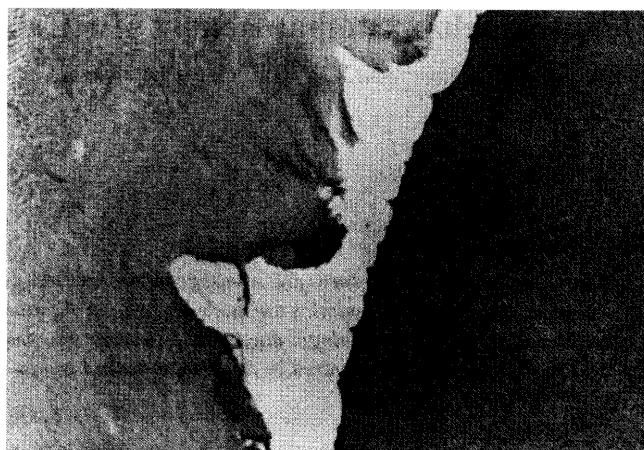


Fig. 3. Enamel and dentine from the hard tissue specimen (H&E, original magnification x100).



Fig. 4. Dentine and cementum from the hard tissue specimen (H&E, original magnification x100).

There were no signs of recurrence at 3 months, 6 months and 1 year post-surgery and orthopantomograms taken during these follow-up visits showed satisfactory bone filling (Fig. 7).

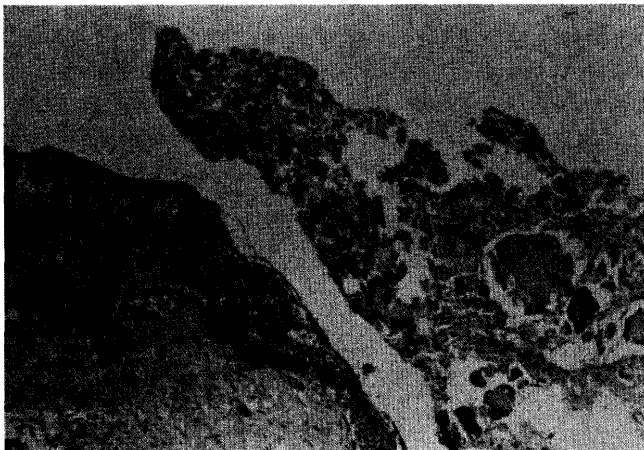
Discussion

Odontoameloblastoma was previously classified as an ameloblastic odontoma along with ameloblastic fibro-odontoma. However, as the names suggest, odontoameloblastoma resembles ameloblastoma in its clinical and histologic behaviour, while ameloblastic fibro-odontoma is more like odontoma (Table I). This difference was later recognised and Hooker⁴ (1967) separated the 2 entities at the annual meeting of the American Academy of Oral Pathology. The WHO then deleted the term ameloblastic odontoma from its Histological Typing of Odontogenic Tumours, Jaw Cysts and Allied Lesions³ and subdivided this category into ameloblastic fibro-odontoma and odontoameloblastoma.

Majority of the reported odontoameloblastoma occurs in children, within the first two decades. It affects males

TABLE 1: COMPARISON OF CLINICAL AND HISTOLOGIC FEATURES OF THE ODONTOAMELOBLASTOMA, AMELOBLASTOMA, AMELOBLASTIC FIBRO-ODONTOME AND ODONTOMA

Feature	Odontoameloblastoma	Ameloblastoma	Ameloblastic fibro-odontome	Odontoma
Age predilection	<20 years	20 to 40 years	<11 years	<20 years
Growth characteristic	locally invasive infiltrates between bony trabeculae	locally invasive infiltrates between bony trabeculae	non-invasive	non-invasive
Recurrence	relatively high with conservative treatment	relatively high with conservative treatment	not known	not known
Histology: epithelium	actively proliferating (ameloblastoma type)	actively proliferating	no active proliferation	nil
Histology: connective tissue stroma	mature connective tissue	mature connective tissue	immature dental papilla-like tissue	nil

Fig. 5. Proliferating odontogenic epithelium showing basal palisading, squamous metaplasia and presence of stellate reticulum-like cells (H&E, original magnification $\times 100$).Fig. 6. Aggregations of ghost cells undergoing dystrophic calcification (H&E, original magnification $\times 100$).

and females equally. The tumour can occur in the maxilla or mandible, most often in the molar-premolar region. Symptoms include dull pain, a slow progressive expansion of the alveolar plates, an altered occlusion, delayed eruption or impacted teeth.^{4,5}

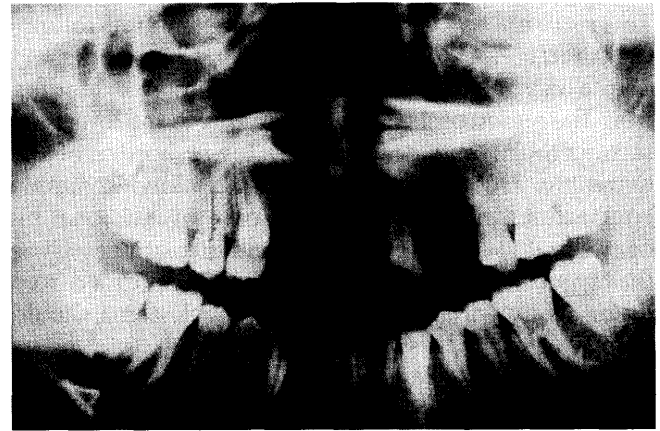


Fig. 7. Orthopantomogram taken 1 year after surgery showing good bone filling and no signs of recurrence.

Radiographically, odontoameloblastoma presents as a well-defined unilocular or multilocular radiolucency with varying amounts of radiopacities within it. The radiopaque material may be diffused particles or a large central mass, which may or may not bear resemblance to formed, miniature teeth.

Like ameloblastoma, odontoameloblastoma is a slow-growing but locally invasive tumour that infiltrates between bony trabeculae.^{1,3,5,6} Recurrences have previously been reported following simple curettage or enucleation⁷⁻⁹ and some authors⁵⁻¹⁰ have recommended radical surgery, as for ameloblastoma.

It is not the usual course to diagnose an odontoameloblastoma preoperatively. This is due to the fact that it is an extremely rare lesion and would not be the first diagnosis to come to mind; its clinical presentation, such as age of occurrence and its radiographic picture, often mimics the more innocuous odontoma (associated with a cyst) rather than the aggressive ameloblastoma. From the cases reported in the literature, preoperative provisional diagnoses mentioned included odontoma,¹¹ odontogenic cyst,⁶ dentigerous

cyst,⁸ fibro-osseous lesion⁵ and cementifying fibroma.⁹ These are all non-invasive lesions that are usually treated conservatively. Unless the lesion is extensive or shows signs of aggressive behaviour (such as resorption of adjacent tooth roots), an incisional biopsy prior to its removal is not routinely performed. In such cases, as in the present one, the diagnosis of odontoameloblastoma and hence the recognition of its aggressive nature is only ascertained after its enucleation. It is our opinion that in these smaller lesions, conservative treatment may in fact be inevitable and may be adequate although close post-operative follow-up is mandatory. The reported recurrences involved large lesions. Frissell and Shafer⁸ reported a case of odontoameloblastoma which extended from the region of the lower right first molar to the right mandibular ramus and condyle in an 11-year-old boy. This recurred twice at 6 months and 49 months following curettage. Another case was reported by Gupta and Gupta⁹ in a 50-year-old man with the tumour extending from the region of the lower left canine to the lower left second molar and down to the lower border of the mandible. This recurred 18 months following simple enucleation. In such sizeable lesions, an incisional biopsy should be done prior to definitive treatment. Once the diagnosis of odontoameloblastoma is confirmed from the histology, wide excision, as for ameloblastoma, should follow.

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