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Ameloblastic Fibroma of the Anterior Maxilla Presenting as a Complication of Tooth Eruption: a Case Report

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Abstract. *Ameloblastic fibroma is a rare mixed odontogenic tumour, which is extremely uncommon in the anterior maxillary region. A case report is presented where failure of eruption of an upper central incisor was the presenting feature.*

Index words: Ameloblastic Fibroma, Incisor, Maxilla.

Introduction

Ameloblastic fibroma is a tumour of mixed connective and odontogenic tissue origin, most commonly found in younger age groups, between 15 and 25 years of age, with males being more affected than females. It presents more frequently in the mandible than the maxilla, usually in the canine to molar region. Clinically, the tumour grows slowly and painlessly, expanding the jaw. Radiographically, it appears as a unilocular area of radiolucency with a smooth outline. It may be difficult to distinguish from a unilocular ameloblastoma or dentigerous cyst, especially if unerupted teeth are involved (Lucas, 1984).

Histologically, the tumour consists of strands and groups of epithelial cells in a connective tissue background and does not invade bone. The epithelial cells are generally cuboidal or low columnar in type, and are similar to the cells that form the peripheral layer of the follicle in an ameloblastoma. They are arranged in irregularly branching strands that have some resemblance to the dental lamina. The connective tissue resembles cellular fibroblastic tissue similar to the dental papilla in the developing tooth, with occasional collagen bands present and sometimes hyaline-like tissue has been seen adjacent to the epithelial strands. Occasional myxomatous areas and foci of predentine have been observed. The tumour is distinguished from ameloblastomas by the fact that both the epithelial and mesenchymal components are neoplastic, while in ameloblastomas only the epithelium is neoplastic (Soames and Southam, 1998).

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Clinical management

Ameloblastic fibroma is a benign lesion and simple excision should be adequate in most cases. Gorlin *et al*. (1961) reported on 23 cases and found only two recurrences. Trodahl (1972), however, followed up a series over longer period and found a recurrence rate of 43·5 per cent, which suggested that the tumour might be more difficult to excise than originally thought. Overall, however, wide resection is not usually required. Other forms include the ameloblastic fibro-odontoma, which occurs more often in the upper jaw. Malignant variations are rare, and include the ameloblastic fibrosarcoma and the extremely rare ameloblastic odontofibrosarcoma.

Case report

A 9-year-old caucasian female was referred by the community dental service for an orthodontic assessment. The presenting problem was failure of eruption of the upper right central incisor (1|). Medical and dental histories were non-contributory. Treatment 2 months previously had involved removal of the retained upper right central and lateral deciduous incisors (BA|) in an attempt to encourage eruption of the upper right central incisor. Observation for 6 months was suggested and the patient was placed on review. As the 1| had not erupted by that stage, further examination was undertaken.

Clinical examination showed that the following teeth were present in the mouth:

6 E D C | 1 2 C D 5 6

6 E D C 2 1 | 1 2 C D E 6

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with a firm swelling on the buccal aspect of the gingiva in the 1| region. No discharge, redness, or fluctuation could be elicited from this area. The initial orthopantomogram confirmed the presence of the following unerupted teeth:

7 5 4 3 2 1 | 3 4 7

7 5 4 3 | 3 4 5 7

Radiographically, the upper right central incisor (1|) was displaced vertically, with a diffuse area of radiolucency around the crown (Figure 1). The intra-oral occlusal radiograph showed the area in greater detail, with the lesion showing what appeared to be a fairly well circumscribed border (Figure 2). The provisional diagnosis was of a dentigerous cyst, and the patient was referred for a surgical opinion and treatment by a maxillofacial surgeon.

The area was investigated under general anaesthetic and found to consist of firm tissue, greyish-white in colour. Samples sent for histopathology were reported as follows.

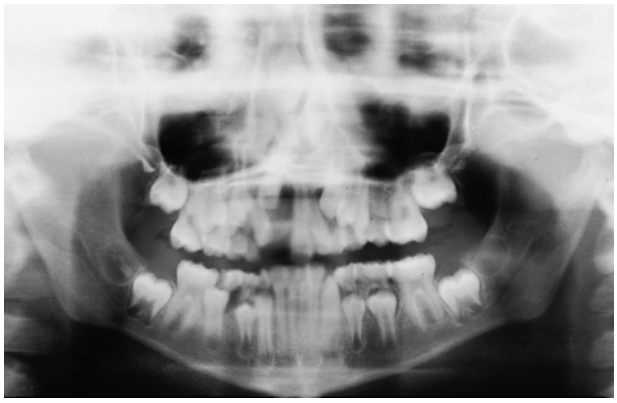

FIG. 1 Orthopantomogram of patient on initial presentation showing displacement of the upper right central incisor.

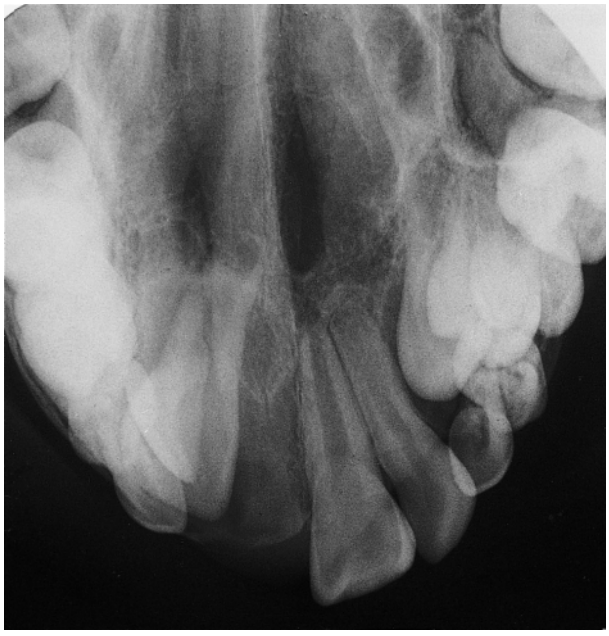

FIG. 2 Maxillary occlusal radiographic view of anterior maxilla at initial presentation showing radiolucency in the region of the upper right central incisor.

'Sections show tumour consisting of a mass of immature mesenchymal tissue with myxoid pattern in which is interspersed islands of benign glandular type epithelium of odontogenic type (Figure 3). The findings are those of ameloblastic fibroma. The tumour is fragmented and incompletely removed. Complete surgical removal is necessary. No fibrous capsule is included.'

Since incomplete removal of the tumour had taken place on the first occasion, the patient underwent further surgery and excision of the remaining tumour tissue was performed. Radiographs (Figures 4 and 5) were taken subsequent to this. After a further 6 months to allow bone regeneration in the region, the upper right central incisor was re-exposed under general anaesthetic and the labial surface bonded with gold chain in order to bring the tooth into the line of the arch using orthodontic traction. The tooth erupted spontaneously through the buccal mucosa and was aligned with a sectional fixed orthodontic appliance (Figure 6).

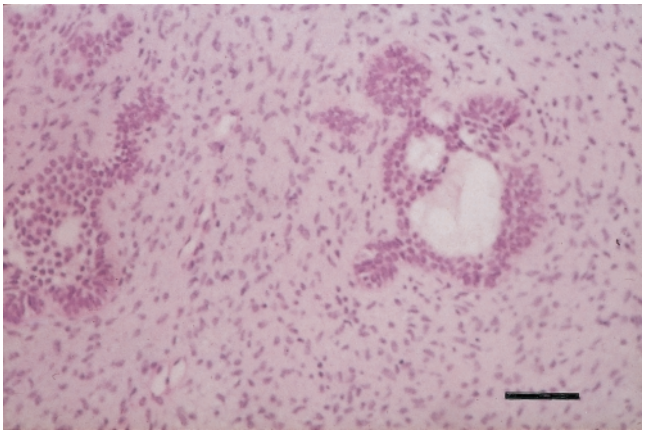

FIG. 3 The stroma shows fibromyxoid appearance with clusters of epithelial cells forming solid nests and glands (high power field, magnification ×40; stain: haemotoxylin and eosin; the horizontal bar represents 100 μ).

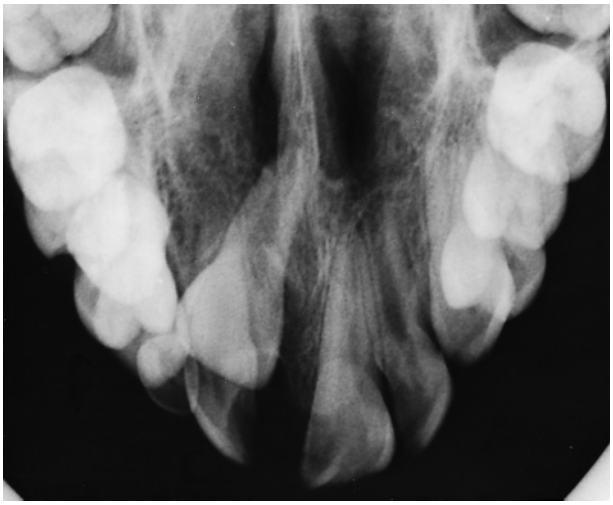

FIG. 4 The maxillary occlusal radiographic view of the anterior maxilla after excision of the tumour.

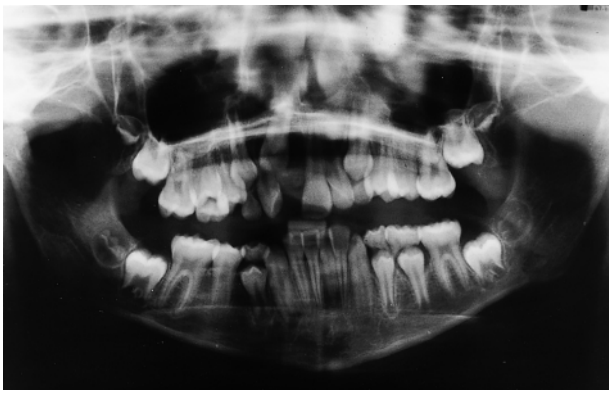

FIG. 5 The orthopantomogram taken after surgical excision of the tumour.

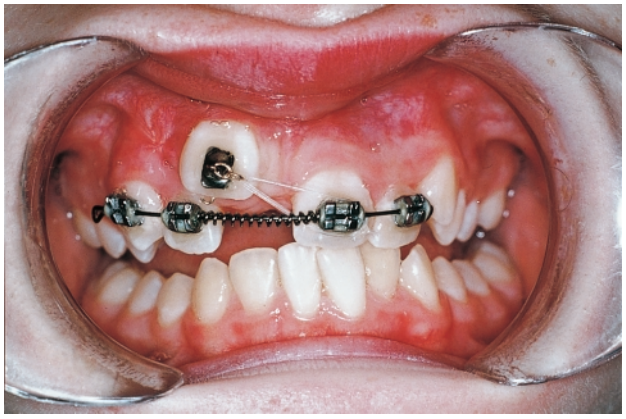

FIG. 6 Clinical view of the anterior teeth with fixed orthodontic appliance in place post-surgery.

Discussion

One of the most frequent causes of failure of tooth eruption is the presence of supernumerary teeth in the line of eruption (Jones and Oliver, 1994). Other causes of failure of individual tooth eruption may be impaction, displacement, or retained deciduous teeth; dilaceration of the root secondary to trauma to the primary incisors; or ankylosis, which may result also from trauma (Mills, 1987). Generalized failure of eruption is very rare (Ireland, 1991) and implies an abnormality of the eruption mechanism.

Most recent reports of tumours causing failure of eruption of teeth in the orthodontic literature have been of mandibular molars (Banks, 1990). The aetiology in these cases has been due to the development of post-inflammatory cysts, odontogenic keratocyst, dentigerous cysts, or ameloblastomas.

The majority of ameloblastic fibromas have been reported in the mandible (Schmidt-Westhausen *et al*., 1991; Junquera *et al*., 1995; Akal *et al*. 1997; Kusama *et al*. 1998). Unusual presentations include an ameloblastoma in association with an ameloblastic fibroma in a 5-year-old boy (Chen *et al*., 1991) and a cystic ameloblastic fibroma in a 7-year-old boy (Meyers *et al*., 1991). Kusama *et al*. (1998) state that a distinction must be made between ameloblastic fibroma and a peripheral ameloblastic fibroma: the latter is a true mixed tumour and in that case report it was found that the lesion had occurred within the gingival tissue, rather than the bony tissues.

In the case reported here, the main presenting feature was the failure of eruption of an upper central incisor tooth. Radiographic investigation showed a radiolucency associated with the crown of the unerupted central incisor and with the clinical feature of enlargement of the gingiva, a provisional diagnosis of a follicular or dentigerous cyst was made. Surgical investigation and histolopathology revealed the lesion to be a benign tumour of odontogenic origin. The case illustrates the importance of careful differential diagnosis in dentistry.

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