Ameloblastic fibro-odontoma—report of two cases with ultrastructural study of tumour dental hard structures

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Summary Two cases of ameloblastic fibro-odontoma (AFO) are reported. Both cases occurred in boys, aged 11 years (case 1) and 9 years (case 2). In case 1 a large mixed radiolucent and radiopaque lesion was observed in the angle of the left mandible. In case 2 with involvement of the left mandibular angle a well-defined translucency was seen without any noticeable calcifications. Both tumours were enucleated. Case 1 revealed the histologic characteristic of an AFO. One part of the tumour consisted of cementoid globules with Sharpey fibre-like structures at their periphery. Electron microscopy revealed areas representative of dentinal tubules. At the periphery connective fibres showed signs of initial calcification. Case 2 showed features of an immature AFO. Further ultrastructural studies are needed to define in more detail the mineralising structures of AFO.

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Introduction

The ameloblastic fibro-odontoma (AFO) has been defined as "a lesion similar to ameloblastic fibroma, but also showing inductive changes that lead to the formation of both dentine and enamel".\textsuperscript{1} In 1997 Philipsen et al. surveyed the literature and disclosed an additional 36 cases.\textsuperscript{2} The relative frequency of AFO among odontogenic tumours (OT) varies between 1% and 3.1% but among 759 Chinese cases of OT AFO was found only in 0.3\%\textsuperscript{3}. The average age was 9.0 years (range 1–21 years), and the male:female ratio was 1.4:1.\textsuperscript{2} Fifty-four percent of AFO are found in the posterior mandible.\textsuperscript{2} Clinically, the AFO appears as a painless slow growing and expanding tumour. In 83% of AFO unerupted teeth are found.\textsuperscript{2} The size varies from microscopically small\textsuperscript{4} to large calcified masses (present case 1). Radiologically, the AFO appears
as a uni- or multilocular radiolucency with a well-defined border.

Histopathologically, AFO is composed of strands, cords and islands of odontogenic epithelium in a primitive ectomesenchyme. Hard structures are composed of dentine-like material or osteodentine in addition to enamel matrix. Few ultrastructural studies (TEM) of AFO have been published. It is the purpose of this paper to report two cases of AFO with transmission electron microscopy (TEM) findings of dentine-cementoid-like structures in one (case 1).

Case reports

Case 1. On routine panoramic radiography, a 11-year old, healthy boy showed a well-circumscribed lesion, posterior to the first mandibular left molar. The second molar was displaced into the ascending ramus. The formation of the roots of this tooth was comparable to that of the second molar of the right mandible. It was, therefore, assumed that the displaced tooth was the second molar of the left mandible. The third molar was missing. The tumour measured 2.5–4 cm in diameter and was composed of a mixed radiolucency/radiopacity (Fig. 1). Computer tomography (CT) confirmed the findings of two large, inhomogeneous radiopacities. The provisional diagnosis of OT, most likely AFO or odontoma was made. Under general anaesthesia the tumour was removed by stepwise sectioning of the hard structures. Postoperative healing was uneventful and bony apposition occurred. Recurrence was not observed after a follow-up period of 3 years.

Figure 1 The panoramic radiograph of the left mandible shows a large well circumscribed lesion of mixed radiopacity and radiolucency. The second molar is displaced, the third molar is missing.

Figure 2 Cementoid globules in a fibrillar connective tissue stroma. At the periphery of these islands Sharpey fibre-like structure irradiate into the adjacent connective tissue (H&E, ×100).

Figure 3 Transmission electron microscopic view of material as shown in Fig. 2. The matrix is in the process of mineralisation and several irregular lumens of different diameters are seen which mimic dentinal tubules (TEM ×1000, bar representing 10 μ).

Parts of the surgical specimen were fixed in 4% buffered formalin and decalcified using EDTA. Sections were stained with H&E, van Gieson, Mallory and Giemsa. Particles of the dental hard structures (1×1×1 mm) were immersed in glutaraldehyde (2.5%) and prepared for routine TEM.

Histologically, the tumour was composed of a fibrous capsule and of strands and islands of odontogenic epithelium embedded in a cell rich...
primitive ectomesenchyme resembling the dental papilla. One part of the dental hard structure component was composed of dentine-like material and enamel matrix embedded in the ectomesenchymal component. The other major part of the dental hard structure revealed a completely

Figure 4 Ultrathin section showing longitudinal and cross sections of connective fibres of different diameter which undergo calcification at the periphery. Crystalloid structures are seen in the adjacent amorphous material (TEM ×25,000, bar representing 1 μ).

Figure 5 Ultrathin section showing part of a mineralised area with crystal needle-like formations and more dense crystalloid structures in the lower right corner of the print (TEM ×25,000, bar representing 1 μ).

Figure 6 Ultrathin section showing a fibrocyte amidst bundles of densely packed collagen fibrils (TEM ×8000, bar representing 10 μ).
different aspect. Globules and islands of different sizes were composed of a cementum-like material without any evidence of dentine-like material or enamel matrix being produced. The borderline between the cement-like globules and the connective tissue component was characterized by collagen fibres irradiating from the surface resembling Sharpey fibres (Fig. 2). The surrounding stroma was that of a mature connective tissue without the characteristic ectomesenchymal appearance.

Electron microscopically, the cementoid-like material revealed a mineralised matrix with the formation of tubules reminiscent of dentine tubules (Fig. 3). At the borderline between calcified and noncalcified connective tissue initial calcification of connective tissue fibres was observed (Figs. 4 and 5). Occasionally, fibrocytes with round electron dense bodies with electron lucent centres were seen surrounded by a densely packed collagen matrix (Fig. 6).

**Case 2.** Panoramic radiography of a 9-year old boy revealed a cyst-like radiolucency without signs of calcification in the area of the left mandibular third molar measuring $1.3 \times 1.5$ cm (Fig. 7). The lesion was enucleated under general anesthesia. Routine histology revealed the characteristic features of an immature AFO. Besides the typical ameloblastic epithelial strands and islands in an ectomesenchymal stroma enamel matrix and some small dystrophic calcifications were observed (Fig. 8).

**Discussion**

The AFO is a rare OT of childhood and adolescence. Histologically, it shows the characteristics of an immature complex odontoma with irregularly arranged enamel, dentinoid, cementoid-like structures and ectomesenchymal tissue. Dentine-like material may vary structurally from dentinoid to tubular dentine. The amount of ectomesenchymal tissue gradually decreases as hard tissue is forming in the central part of the tumour.

There are few ultrastructural studies of AFO in the literature. Reich et al. observed that the ameloblastic component was similar to that of the developing odontogenic tissue, except that the ectomesenchymal cells have not developed into columnar odontoblasts. This finding was taken as an explanation while osteodentin or dentinoid material is produced in AFO instead of tubular

Figure 7 Part of panoramic radiograph showing a well-circumscribed translucency distal to the left second mandibular molar (case 2).

Figure 8 Islands of ameloblastic epithelium in an ectomesenchymal stroma. Small island of initial calcification are seen (H&E, ×50).
dentin.\textsuperscript{10} A TEM analysis of the dental hard structures, however, was not performed.

The present case 1 is of interest, because parts of the dental hard tissues consisted of typical immature complex odontoma-like structures, while the other part represented a more cementoid-dentinoid-like material with Sharpey fibre-like structures at the periphery. The differentiation between osteodentine-like material and cementoid material has been the focus of academic discussion for years. The present TEM findings indicated a mineralised matrix of some of the hard structure components with tubular formations which may be representative of a dentinoid-like material. On the other hand, at the periphery irradiating Sharpey fibre-like structures have been observed which may be indicating a cementoid origin. Further electron microscopic studies of such hard structure are warranted to clarify the origin of such materials.

References


