An unconventional ameloblastic fibro-odontoma with compound odontoma like features

Radhika Manoj Bavle, Soumya Makarla, Paremala K., Sreelatha S. Hosthor, Sudhakara M., Reshma Venugopal

ABSTRACT

Introduction: Odontogenic tumors cover a wide and diverse spectrum of tumors. Some odontogenic tumors have a varied etiopathogenesis and share a presentation very similar to hybrid odontogenic tumors. Ameloblastic fibro-odontoma is one such tumor. Though the WHO 2017 Classification of Head and Neck tumors continues to support this hypothesis, it is clear, with more than 215 case reports, that it is a tumor that does not feature in the continuum of the spectrum of ameloblastic fibroma ending with ameloblastic fibro-odontoma and odontoma. It, therefore, deems recognition as an independent entity. The ameloblastic fibro-odontoma irrespective of central or peripheral location will show an ameloblastic fibroma like presentation along with a compound or complex composite odontome histologically. It has also been conjectured whether it is a true neoplasm or hamartoma.

Case Report: We report a case of an ameloblastic fibro-odontoma in an eight-year-old boy with a diffuse swelling extraorally in the infraorbitalzygomatic area. Intraoral examination revealed a large 4.5x3 cm soft tissue mass on the right maxillary alveolar area involving primary molars and permanent 1st and 2nd molar region. Radiologic examination revealed a lesion with 32 denticles and destruction of alveolar bone, which showed a histopathologic picture of ameloblastic fibro-odontoma with a composite compound odontoma like pattern.

Conclusion: Our case describes an ameloblastic fibro-odontoma, which presented as a painful mass of the maxilla with compound composite odontome like features supporting the tumor concept and as an independent entity not belonging to the continuum spectrum.
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Keywords: Ameloblastic fibro-odontoma, Central, Composite compound odontoma, Dentin, Enamel, Induction, Maxillary posterior quadrant

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INTRODUCTION

Odontogenic tumors cover a wide spectrum and comprise an array of tumors. These have been classified into epithelial, mesenchymal and mixed tumors by the 2017 WHO classification of odontogenic tumors [1]. Ameloblastic fibro-odontoma (AF0) previously was listed under the category of mixed odontogenic tumors formerly described under names like immature ameloblastic odontoma (Slootweg) and ameloblastic odontoma (Hooker). This group of tumors shows the evidence of odontogenic induction along with mesenchymal tissue proliferation. The present tumor morphologically has resemblance of ameloblastic fibroma along with odontome like areas. It strikes ahead of ameloblastic fibro dentinoma by showing the presence of induction in formation of enamel or enamel matrix [2].

Ameloblastic fibro-odontoma is a rare tumor with approximately 215 well documented reports (211 – central, 3 peripheral lesions) [3]. Irrespective of its location, the tumor presentation is similar histologically. It is a biologically non-aggressive tumor, responding well to conservative treatment. It is unfortunate that this distinct lesion has not been recognized by the recent classification of odontogenic tumors and has been grouped under the umbrella term developing odontomas or as a variant of ameloblastic fibroma [1, 4].

We present a case of ameloblastic fibro-odontoma with a very classic histopathologic presentation supporting the tumor concept and as an independent entity not belonging to the continuum spectrum.

CASE REPORT

An eight-year-old boy reported to the clinical department with a complaint of painful mass in the upper jaw since a month. The child presented with a diffuse swelling of the right cheek, extraorally, measuring approximately 5x4.5 cm and extending anterior-posteriorly from the nasolabial fold to the pre-auricular area and superior-inferiorly from the infraorbital region to 2 cm below the ala-tragal line. Intraoral examination revealed a large 4.5x3 cm soft tissue mass on the right maxillary alveolar area involving primary molars and permanent 1st and 2nd molar region. Mucosa covering the lesion appeared erythematous on the superior surface and the mass was obstructing the occlusion (Figure 1A–B).

On palpation, a soft tissue mass with well-defined borders was noted, with expansion of both buccal and palatal plates. A provisional diagnosis of odontome was given. On radiologic examination, the orthopantomogram revealed a mixed dentition state of the child. The upper right maxillary quadrant showed an ill-defined radiolucent lesion with multiple small pebble size radio-opaque masses. Approximately, 32 such denticle like structures were noted in the radiograph. Destruction of the alveolar bone in the right maxillary molar region and retromolar area was observed. Developing tooth follicles of permanent molars 16 and 17 were pushed superiorly into the infraorbital area. The incompletely formed teeth 14 and 15 were mesially displaced (Figure 1C).

The lesion was provisionally diagnosed as compound odontome based on radiographic features and a conservative excision was planned. The differential diagnoses included complex composite odontome and odontoameloblastoma.

Surgical note described a well-delineated mass, soft in nature, which shelled out of the bone. It was irregular in shape with multiple hard tooth crown like structures within the tumor mass.

On grossing, a 4.5x4 cm tumor mass, irregular in shape was seen. Cut section revealed off white, pale myxoid area and chalk white area intervened with numerous small tooth like structures. Some of the tooth like structures separated and shelled out from the tumor mass. A radiograph of the gross specimen reconfirmed multiple such tooth like structures, approximately 32 in number (Figure 2).

On histopathologic examination with hematoxylin and eosin staining, the lesional tissue showed a predominance of immature cellular cementoblast cell – made up of a myxoid stroma with plump spindle cells, which are monotonous. The odontogenic epithelium is seen in the form of strands and small nests. The strands were similar to the dental lamina (Figure 3).

At multiple areas the dental lamina like strands gave rise to dental follicles very similar to advanced bell stage, showing the presence of ameloblasts, odontoblasts along with enamel matrix and dentin formation. Enamel was partially mineralized or was seen as an un-mineralized matrix. Mineralized enamel showed the presence of well-formed enamel prisms (Figure 4). Dental follicles showed the presence of thick tubular predentin like areas lined by odontoblasts. The area of dentin next to the enamel is well mineralized with globular dentin. Different areas showed dental follicles in various stages of tooth development. The enamel organs bore resemblance to cap, bell and advanced bell stages very frequently. No evidence of cementum like tissue was seen, but primitive pulp in the form of dental papilla was observed (Figure 5). These areas appeared similar to a composite compound odontome. Based on these findings, a final diagnosis of ameloblastic fibro-odontoma was given.

DISCUSSION

Ameloblastic fibro-odontoma is defined as a lesion similar to ameloblastic fibroma but also shows inductive changes that lead to the formation of enamel and dentin [5]. It is a rare tumor accounting for 1–3% of all odontogenic tumors [3]. Ameloblastic fibro-odontoma bears resemblance to ameloblastic fibroma, ameloblastic fibrodentinoma.
and odontomes because of which investigators have hypothesized that ameloblastic fibro-odontoma could be one entity in different stages of development, in which: ameloblastic fibroma evolves to ameloblastic fibrodentinoma (AFD); ameloblastic fibrodentinoma evolves to ameloblastic fibro-odontoma and ameloblastic fibro-odontoma matures to an odontoma (Figure 6). However, the clinical age association, site of occurrence, histopathology, gender and evidence clearly shows a disagreeable point in the continuous differentiation hypothesis proposed by Cahn and Blum [5, 6].
Another hypothesis proposed is on the two lines of development of ameloblastic fibro-odontoma. A neoplastic line comprised ameloblastic fibroma and ameloblastic fibrodentinoma and a hamartomatous line comprising of ameloblastic fibro-odontoma as a stage of odontoma [3, 5] (Figure 7). Some authors suggest that AFO should be considered as an immature complex odontoma [7]. Trodahl suggested that AFO exists in a stage in between the two schools of thought [8]. However, ameloblastic fibro-odontoma cannot be unanimously considered as a hamartoma, as it can show neoplastic biological behavior, cause bone destruction, deformity and instances of malignant transformation and variants have also been noted [9–11].

The present case definitely shows neoplastic biological behavior as it presented as a large tumor mass with bone destruction and cortical plate expansion. It also emerges as an independent entity and not as a part of continuum spectrum based on the clinical data.

Ameloblastic fibro-odontoma arises in patients between 8–12 years of age, [4] with a mean age of 9 years [5]. A slight male predilection is seen with more than half of the cases arising in the posterior mandible region intrasosseously. It presents as a well-delineated slow growing painless mass; as a central lesion, generally associated with unerupted teeth, which is a common hallmark presentation of ameloblastic fibro-odontoma. On radiography, it presents as a unilocular or multilocular radiolucency with radio-opacities of different shapes and density. Displaced unerupted teeth show the presence of the tumor coronally [6, 12, 13].

The present case sits in the classical age group with eight years at the time of presentation in a boy, which was a typical feature. But the tumor presented in the maxillary posterior quadrant as a large tumor with cortical plate expansion and displaced unerupted teeth in a unilocular radiolucency. Multiple, as many as 32–40, tooth like structures were also seen on the radiograph. Approximately 80% of the lesions are associated with a tooth and 87% show radio-opacities as seen in our case, which showed the displacement of incompletely formed 14, 15, 16 and 17 [3, 5, 6].

Histopathological findings speak of immature complex odontome with enamel, dentin, cementum and pulp-like ectomesenchyme with odontogenic epithelium. Dentin can be in the form of tubular dentin or dentinoid [2].

Some authors describe it as a biphasic tumor with odontogenic epithelium proliferating in a highly cellular ectomesenchyme with primitive apparatus. It also contains tooth like structures – enamel and dentin with
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Conflict of Interest
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