

## CASE REPORT

# Ameloblastic fibroma in six-year-old male: Hamartoma or a true neoplasm

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**ABSTRACT**

Ameloblastic fibroma (AF) is an uncommon true mixed odontogenic tumor with a relative frequency between 1.5% and 4.5%. Recently, it has been proposed that two subtypes of AF exist: A neoplasm and a hamartomatous lesion. We report a case of AF in left mandibular posterior region in a 6-year-old male patient. The tumor showed unusual and aggressive features that suggested it was a true neoplasm.

**Key words:** Ameloblastic fibroma, hamartoma, neoplasm, odontogenic tumor

**INTRODUCTION**

Ameloblastic fibroma (AF) was first reported by Kruse in 1891.<sup>[1]</sup> It is an uncommon benign mixed odontogenic tumor with a relative frequency between 1.5% and 4.5%.<sup>[2]</sup> The ameloblastic fibroma is generally considered to be a true mixed odontogenic tumor in which both the epithelial and ectomesenchymal components are neoplastic.<sup>[3]</sup>

There has been a long debate as to whether ameloblastic fibroma represents an anomalous hamartomatous growth, or is a true benign neoplasm. This is due, in part, to the difficulties to differentiate between the histology of the neoplastic and the hamartomatous lesions with the histologic features of ameloblastic fibroma.<sup>[3]</sup> Recently, it has been proposed that two variants of ameloblastic fibroma exist: A neoplastic type with no induction phenomenon and a hamartomatous type showing inductive capabilities. But still final proof for this hypothesis is missing.<sup>[2]</sup>

Here, we describe a case of ameloblastic fibroma in six years pediatric male patient, with details of some unusual and aggressive clinical, radiographic, and histologic features suggesting that it was a true neoplasm.

**CASE REPORT**

A 6-year-old male patient was presented in the Department of Oral and Maxillofacial Pathology, National Dental College, Derabassi with a painless left mandibular swelling [Figure 1].

The patient first noticed the swelling one month prior to the presentation. Initially the swelling was minimal but it had grown slowly with time to the present size. The swelling was painless throughout its course.

**Clinical features**

Extraoral examination revealed a large irregular swelling in relation to the body of the mandible on left side extending to involve the ramus on the same side. The approximate size of the swelling was 4 cm × 2 cm. The overlying skin was normal in color and smooth.

On palpation, the swelling was non tender with bony hard consistency, non compressible, non fluctuant and fixed to the underlying structures. Submandibular lymph nodes were non palpable and non tender.

Intraoral examination revealed an irregular swelling on the left side of the mandible obliterating the buccal vestibule extending from deciduous first molar to the ramus of the mandible on the same side. The swelling was irregular in shape. The overlying mucosa was normal. Left mandibular first permanent molar had not erupted in the oral cavity.

**Radiographic features**

Radiographically, orthopantomograph (OPG) showed a radiolucent lesion with scalloped margins on the left side of the mandible involving the body and extending upto the ramus of the mandible, associated with the crown of permanent first molar pushing it against the inferior border of mandible and antero superiorly causing resorption of distal root of 75 [Figure 2].

**Macroscopic examination**

The lesion was enucleated under general anesthesia and sent for histopathological examination. The tissue received was

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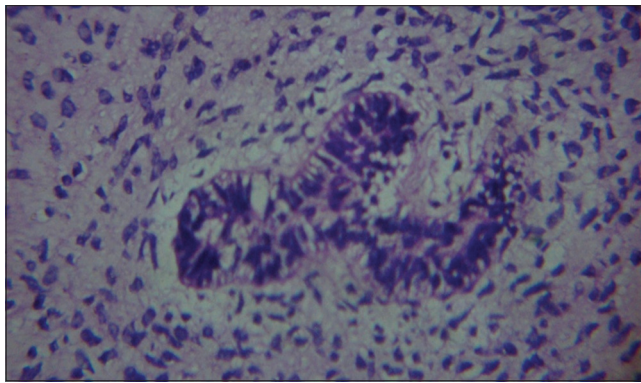
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**Figure 1:** Swelling in the left mandibular region involving the body of the mandible and extending upto the ramus on the same side



**Figure 3:** Macroscopic appearance of the tumor



**Figure 5:** Photomicrograph showing island of odontogenic epithelium in a primitive connective tissue background resembling dental papilla without the formation of dental hard tissues. (H and E, 40×)

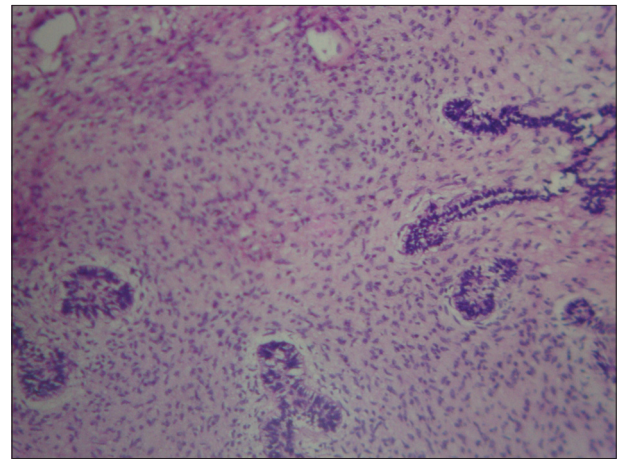
white in color but in pieces with the largest one measuring 6 cm × 2 cm [Figure 3]. The permanent first molar was also removed along with the tumor.

### Microscopic features

The H and E stained section showed both epithelial and mesenchymal elements [Figure 4]. The mass was composed of embryonic mesenchyme which is traversed by elongated cords of cuboidal and columnar odontogenic epithelial cells that resemble the dental lamina. Some peripheral cells were tall columnar with reverse polarity. Organelle free distal zone was present in these cells. Some



**Figure 2:** OPG showing radiolucent lesion with scalloped margins involving the crown of left permanent first molar causing root resorption of 75



**Figure 4:** Photomicrograph showing both odontogenic epithelium and ectomesenchymal components. (H and E, 10×)

follicles containing stellate reticulum in the centre are also identified [Figure 5].

Mesenchymal component resembled dental papilla. These contain angular cells. There was presence of few delicate collagen fibres.

The overall features confirmed the diagnosis of ameloblastic fibroma.

### DISCUSSION

Ameloblastic fibroma is a benign, true mixed odontogenic tumor in which both the epithelium and the ectomesenchymal components are neoplastic without hard tissue formation.<sup>[3]</sup> It is generally intraosseous, but can also occur rarely in peripheral locations.<sup>[4]</sup> Ameloblastic fibroma is mostly encountered in young patients especially during the first two decades of life.<sup>[5]</sup> Males are affected by ameloblastic fibroma more often than females, with a male: female ratio of 1.4: 1.<sup>[2]</sup> Mandible is the predominant site of occurrence and the posterior mandible is affected more often than the maxilla by a factor of 3.1. Impacted, unerupted teeth are associated with ameloblastic fibroma in three quarter of the cases. Ameloblastic fibroma may develop in areas of congenitally missing teeth also.<sup>[3,6]</sup>

Clinically, ameloblastic fibroma is a painless slow growing expansile lesion of the jaw and in about 20% of the cases discovered accidentally on radiographs. Radiographically the tumor is characterized as a well defined uni or multilocular radiolucency often with a sclerotic border.<sup>[3,6,7]</sup>

Histopathologically, the epithelial tumor component is arranged in strands, cords and islands of proliferating odontogenic epithelium. The strands often reveal a double or triple layer of cuboidal cells thus resembling the dental lamina of early tooth development. The islands often show a peripheral row of high cuboidal or columnar ameloblasts like cells. The centre of individual tumor islands may enclose a number of cells resembling stellate reticulum. The ectomesenchymal cells are rounded or angular and there is a little collagen which is represented by a few delicate collagen fibrils. Occasionally some parts of ectomesenchymal component may reveal a loose myxomatous structure with a weakly positive metachromatic substance. There may be cell free zone bordering the epithelial islands and strands and in rare instances juxta epithelial hyalinization.<sup>[3]</sup>

The nature of ameloblastic fibroma is controversial. The 1992 WHO classification does not include a definition of ameloblastic fibroma as an entity.<sup>[8]</sup> The authors pooled ameloblastic fibroma with “related lesions,” which also covers the ameloblastic fibrodentinoma and ameloblastic fibroodontoma. The suggested definition for this group of lesion was “neoplasm composed of proliferating odontogenic epithelium embedded in a cellular ectomesenchymal tissue that resembles the dental papilla and with varying degree of inductive change and dental hard tissue formation”.

Cahn and Blum<sup>[9]</sup> proposed the continuum concept based on the assumption that an ameloblastic fibroma will, overtime, mature and finally result into the formation of odontoma. Some points contradicting this theory were raised:<sup>[3]</sup>

1. Recurrent cases of AF have never shown further steps of differentiation into dental hard tissue forming odontogenic tumor of more advanced histodifferentiation.
2. AFs are known to occur at ages beyond completion of odontogenesis, that is, after the age of 20 years.

Hence it was proposed<sup>[3]</sup> that AFs occurring after the age of 20 years are true benign neoplasm. All cases of AF developing during the entire period of odontogenesis however may represent non neoplastic hamartomatous lesion.

It has recently been suggested that ameloblastic fibroma and fibrodentinoma occur in two variants, one being a neoplastic lesion, which - if left *in situ* - does not seem to differentiate further. The second variant is a non neoplastic, hamartomatous lesion, which seems capable of developing into an ameloblastic fibro-odontoma and differentiating further into complex odontoma. The latter line of development has been termed as developing complex odontoma line.<sup>[10]</sup>

Interestingly, our present case had some unusual clinical, radiographic and histologic features that distinguished it from normal types of AF, and supporting its neoplastic nature.

Firstly, AFs are slow growing and relatively small in size. However, some large tumors have been reported<sup>[2]</sup> and the present case showed unusually rapid growth to more than 5 cm within one month as reported by the patient. Secondly, radiographic examination usually reveals a well defined radiolucency associated with impacted tooth,<sup>[3]</sup> but in the present case the radiolucency was large with scalloped margins, associated with root resorption of 75, which is quite uncommon. Thirdly, the voluminous nature of the lesion has pushed the crown of 36 to the inferior border of the mandible, thus preventing its eruption into the oral cavity. Fourthly, histopathologically no zone of induction could be appreciated around the epithelial follicles, suggesting it to be neoplastic subtype,<sup>[3]</sup> as being one of the criteria to differentiate the two subtypes.

Although considering the age of the patient, the AF developing during the period of odontogenesis should be considered as non neoplastic hamartomatous lesion. But based on the currently available evidence and findings in the present case, we consider AF in this case to be true neoplasm rather than a hamartoma.

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