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Ameloblastic fibroma: a case report

Ronald A, Bell, DDS, MEd
David L. Schaffner, DDS, MS
David R. Myers, DDS, MS

Abstract

The clinical and histopathologic features of a case of ameloblastic fibroma in a 9-year-old boy has been presented along with a review of the literature. The tumor responded to conservative surgical treatment and there was no recurrence one year postoperatively. Analysis of the literature suggests that the tumor may have a higher potential for recurrence than is generally appreciated. In addition, the possibility of maturational differentiation and/or malignant transformation should be recognized. The importance of early and accurate diagnosis, prompt treatment, and long-term followup is emphasized.

The ameloblastic fibroma is an uncommon, benign, mixed odontogenic neoplasm. It is the least differentiated of the odontogenic mixed tumors in that the neoplastic elements do not characteristically produce dentin or enamel matrix, the hallmark of the more differentiated tumors. Biologically, it is generally regarded as being less aggressive than the ameloblastoma, a feature which must be considered in the rational treatment and management of the patient with this tumor.

Report of Case

A 9-year-old Caucasian male was seen in February, 1980, in the pedodontic clinic at the Medical College of Georgia School of Dentistry for routine examination. The parent's only concern relating to the child's dentition was that there was "too much spacing and sticking out of front teeth."

Extraoral examination revealed a well-nourished, healthy child with no evidence of lymphadenopathy or facial asymmetry. Intraoral examination revealed that the permanent incisors, permanent first molars, primary cuspids and primary molars were present. The alignment of the posterior mixed dentition was normal with the exception of a partially erupted lower left first permanent molar which was displayed approximately 6-7 mm distal to the deciduous second molar (Figure 1). The opposing left maxillary first permanent molar had supraerupted about 2 mm. The soft tissues surrounding the displaced tooth were normal except for slight inflamation associated with the operculum. The patient indicated no history of symptoms associated with the lower left quadrant and there was no tenderness to palpation. No buccal or lingual cortical expansion was noted.

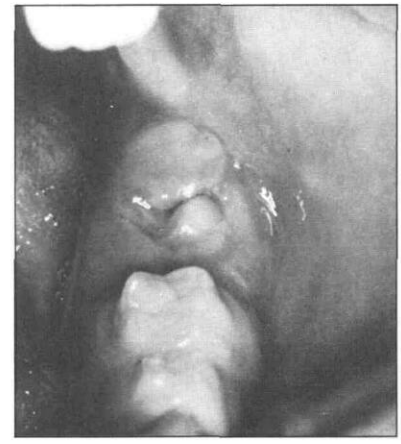

Figure 1. Initial clinical presentation of distally displaced mandibular left first permanent molar.

Radiographic examination revealed a multilocular, radiolucent lesion close to the crest of the alveolar ridge occupying the space between the first permanent molar and the second primary molar. The first permanent molar was displaced and tipped distally 6-7 mm (Figure 2). An occlusal radiograph (Figure 3) indicated no erosion but possible slight expansion of the lingual cortical plate.

An incisional biopsy was performed using intravenous meperidine hydrochloride and diazepam sedation in conjunction with local anesthesia. A mucoperiosteal flap was reflected facially from the mandibular left cuspid area to the distal of the first permanent molar, revealing the firm, white lesional tissue which had eroded through the cortical bone along with the alveolar crest. Several fragments of this tissue were curetted and submitted for histopathologic evaluation. The flap was repositioned and sutured interproximally with 3-0 plain gut sutures.

Histologic sections (Figure 4) showed a soft tissue specimen consisting of a benign neoplastic proliferation of fibrous connective tissue with numerous small islands and cords of epithelium dispersed throughout the specimen. The epithelial component generally consisted of a double layer of columnar cells. The immature connective tissue stroma was characterized by a loose network of delicate collagen fibers in association with spindle- and stellate-shaped fibroblasts. Some of the epithelial islands were rimmed by a zone of connective tissue hyalinization. The diagnosis was ameloblastic fibroma.

The patient was admitted to Eugene Talmadge Memorial Hospital for surgical removal of the lesion

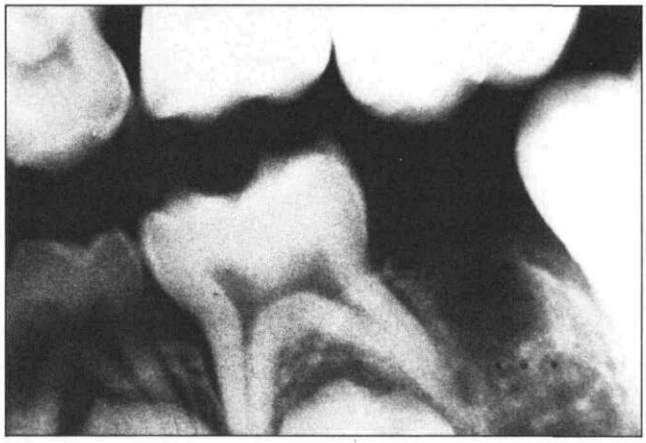

Figure 2. Bitewing radiograph showing multilocular radiolucent lesion, distally displaced mandibular first permanent molar, and supraeruption of the maxillary left first permanent molar.

under general anesthesia. The patient's family and medical history, and review of systems were unremarkable. Preoperative laboratory work, including CBC, SMA-6, urinalysis, and chest x-ray films were within normal limits.

A full thickness mucoperiosteal flap extending from the retromolar pad to the left primary canine was reflected allowing removal of the buccal plate in the area of the tumor. On exposure it became evident that the lesion extended inferiorly and lingually in proximity to the roots of the second primary molar. The second primary molar was removed and the tumor, the bulk of which was situated just beneath the lingual cortex, was cleanly enucleated. The bone was smoothed with a bone file and the area vigorously irrigated. The gingiva was repositioned with multiple 3-0 plain gut sutures. The recovery and postoperative period were uneventful and the patient was discharged the day following surgery. The surgical specimen was submitted for histopathologic examination and the diagnosis of ameloblastic fibroma was confirmed.

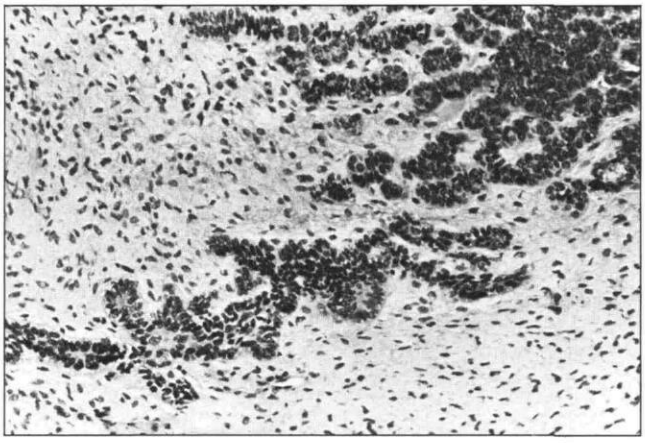

Figure 4. Photomicropgraph of the biopsy specimen showing numerous cords of odontogenic epithelium dispersed throughout an immature connective tissue stroma (Hematoxylin and Eosin, original magnification 100 x).

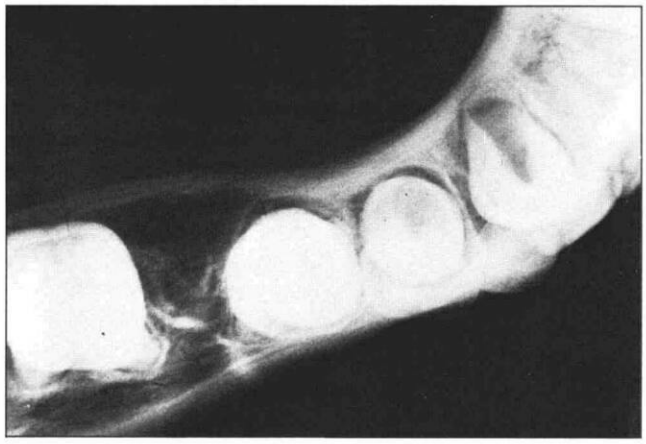

Figure 3. Occlusal radiograph on the left mandible showing lingual orientation of the lesion with no apparent cortical erosion.

Follow-up examinations have been made at three, six, and twelve months. At one year, the second bicuspid had emerged into the oral cavity with no evidence of developmental defect and the first permanent molar had migrated mesially approximately 4-5 mm. Radiographically, significant regeneration of bone and no evidence of residual or recurrent tumor were noted (Figure 5). The patient will be maintained on a regular six-month recall schedule.

Discussion

The ameloblastic fibroma is an odontogenic tumor found primarily in children and teenagers (Table 1) with no apparent sex or race predilection. The lesion may occur in either jaw, although 80% of the reported cases have been in the mandible, usually in the premolar-molar area. The tumor enlarges by gradual expansion and often exhibits an asymptomatic clinical course. Pain or swelling may be the patient's initial complaint.[1-5]

Ameloblastic fibroma radiographically presents as a unilocular or multilocular radiolucency with a

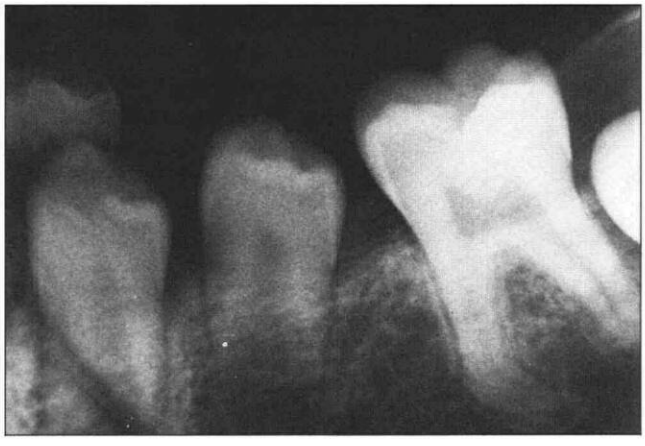

Figure 5. Twelve-month postoperative radiograph of the left mandibular area showing no sign of tumor recurrence. Significant regeneration of bone, normal bicuspid eruption, and mesial movement of the first permanent molar are noted.

smooth, well-defined periphery.[1,2,5] Associated features may include unerupted or displaced teeth, divergence of the roots of adjacent teeth, or expansion of the cortical plates.[1-6]

Histologically, the ameloblastic fibroma is characterized by the proliferation of odontogenic epithelium supported by a primitive mesenchymal connective tissue stroma.[1,2] The epithelium presents as nests, buds, and cords of cuboidal or columnar cells which may develop a central portion resembling stellate reticulum. The cell-rich mesenchymal component closely resembles the dental papilla of the developing tooth germ. The ameloblastic fibroma contains no calcified tissue elements.

Generally credited as demonstrating benign behavior, the recommended treatment for ameloblastic fibroma consists of curettage or enucleation.[1-6] A few recurrences have been documented by Gorlin et al.,[3,4] Heringer,[6] Tanaka[7] and Lysell and Sund.[8] Trodahl,[5] in a survey of 24 cases from the Armed Forces Institute of Pathology, indicated 10 patients required further surgical procedures after the initial treatment, an apparent recurrence rate of 43.5%. In most of these recurrent cases the initial surgical procedure was "believed to have completely removed the tumor, and yet it recurred."[5] Interestingly, 4 of the 10 cases recurred more than two years after initial treatment; the majority of other cases, in which evidence of recurrence was reported, were not followed for that length of time.

The possible pathogenesis of ameloblastic fibroma has been correlated with the events of normal odontogenesis.[5,9,10] This maturational theory of mixed odontogenic tumor origin proposes that the ameloblastic fibroma and ameloblastic fibro-odontoma are progressive phases in the development of odontomas. In support of this viewpoint, Trodahl[5] and Carr et al.[11] have pointed out that some recurrent lesions initially diagnosed as ameloblastic fibroma showed maturation toward ameloblastic fibro-odontoma or odontoma. On the other hand, the fact that no histologic evidence of additional maturation has been observed in many other examples of recurrent ameloblastic fibroma provides support to the theory that the ameloblastic fibroma is an independent entity.[7,8,12,13]

Further support for the maturational theory is derived from the observation that all these tumors show a similar distribution in the jaws and occur in the same general patient age population.[14,15] A more detailed analysis of patient data by Slootwig,[16] however, showed the mean age of occurrence for the ameloblastic fibroma to be 14.6 years in contrast to 8.1 years for the ameloblastic fibro-odontoma.

In view of this data, the maturational theory seems unlikely in that the more differentiated tumor should not occur at a younger age than the tumor from which it is hypothetically derived. Slootwig's data also showed significant differences in the distribution of these two tumors. His data does supoort the concept that the ameloblastic fibro-odontoma may repre-

Table 1. Ameloblastic fibroma: data summary of documented cases through 1980.

Author	Number of cases	Sex M	Sex F	Age Range	Average Age	Site Max.	Site Mand.	Recurrences
Gorlin et al.[4]	36*	20	15	1½-39y	14y	8	26	2
Trodahl[5]	24**	16	8	1½-4ly	15y	3	21	10
Heringer[6]	19***	9	10	1m-42y	15y	3	16	1
Additional Reports****								
Tanaka et al.[7]	1	1		7y	7y		1	1
Lysell & Sund[8]	2	1	1	7-13y	10y	2		1
Nilsen & Magnusson[10]	2	1	1	9-14y	11½y	1	1	0
Reichart & Zobl[18]	1	1		16y	16y		1	1
Rodney & Carrington[23]	1	1		6y	6y	1		0
Lewin-Epstein et al.[24]	1		1	8y	8y	1		0
Singh & Agarwal[25]	1	1		35y	35y		1	0
Hager et al.[26]	1		1	7y	7y		1	0
Edwards & Goubran[27]	1	1		18y	18y		1	0
Present Case	1	1		9y	9y		1	0
Totals	91	53 59%	37 41%	1m-42y	14y	19 21%	70 79%	16 18%

*Gorlin et al.[4] reported 10 of their own cases and 26 compiled from published reports.

**Trodahl[5] reported on a survey of cases from the Armed Forces Institute of Pathology.

***Heringer[6] documented 19 cases not included in the tabulations of Gorlin et al.[4] and Trodahl.[5]

****Additional reports include cases not previously tabulated in comprehensive reviews.

sent an immature form of an odontoma, but the ameloblastic fibroma probably arises as a separate odontogenic tumor. Additionally, the ameloblastic fibroma is appropriately designated as a neoplasm as it has the potential for unlimited growth, recurrence, and malignant transformation. This is in distinction to the odontoma which lacks these properties and may be more appropriately considered a hamartoma.

Of concern is the possibility of malignant transformation of the ameloblastic fibroma into ameloblastic fibrosarcoma.[11,17,18] The ameloblastic fibrosarcoma is a rare malignant tumor of clusters and strands of benign epithelial components within a cell-rich mesenchymal component exhibiting the cytologic features of a fibrosarcoma.[1,17,18,19] Leider et al.[20] and Goldstein et al.[21] documented recurrent ameloblastic fibromas that had transformed histologically into ameloblastic fibrosarcoma. These tumors were characterized by progressive overgrowth and increased cellularity of the malignant mesenchymal component, leaving only a few remnants of odontogenic epithelium.

In a review of fibrosarcomas of bone, Dahlin and Ivins[22] reported that 2 of 13 fibrosarcomas of the mandible occurred at sites of previously diagnosed ameloblastic fibroma. In their review of the literature, Howell and Burkes[11] identified 22 cases of ameloblastic fibrosarcoma and 89 cases of ameloblastic fibroma and fibro-odontoma. They suggest that although this high ratio of malignant lesions may in part be artefactual due to the greater likelihood that a sarcoma will be reported, the malignant potential of ameloblastic fibroma may be somewhat higher than is generally appreciated. They concluded, however, that the use of radical surgical procedures in the treatment of ameloblastic fibroma is still not justified, although careful microscopic review of the histopathology and a prolonged period of followup is mandatory.

Summary

A case of ameloblastic fibroma of the posterior mandible in a 9-year-old boy has been presented. The clinical and histological aspects of the case are typical of ameloblastic fibroma, as is the lack of recurrence during the initial 12-month postoperative period following conservative surgical treatment. Given the questions and concerns in the literature with regard to recurrence rate, possibility of maturational differtiation, and transformation into fibrosarcoma, the importance of early and accurate diagnosis, prompt treatment, and long-term followup must be emphasized.

Dr. Bell is associate professor and director of clinical pedodontics, Department of Pedodontics; Dr. Schaffner is assistant professor, Department of Oral Pathology; and Dr. Myers is professor and chairman, Department of Pedodontics, School of Dentistry, Medical College of Georgia, Augusta, GA 30912. Requests for reprints should be sent to Dr. Bell.

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