Case Report

Ameloblastic fibroma or fibrosarcoma: A dilemma of oral surgeon

Nitin Verma, Neha

Department of Oral and Maxillofacial Surgery, Government Dental College and Hospital, Amritsar, Punjab, India

Address for correspondence:
Dr. Neha,
Government Dental Hostel for Girls, Near Petrol Pump, Batala Road, Amritsar, Punjab, India.
E-mail: dr.neha89nm@gmail.com

ABSTRACT

Ameloblastic fibroma (AF) is an uncommon true mixed odontogenic tumor, with a relative frequency between 1.5% and 4.5% of all odontogenic tumors. It may behave either as a true neoplasm or as a hamartomatous proliferation of odontogenic epithelium of the enamel organ and odontogenic mesenchyme of the primitive dental pulp. Frequently diagnosed between the first and second decades of life with 75% of cases was diagnosed before the age of 20 and present with a well-defined unilocular or multilocular radiolucencies. A conservative approach, enucleation with curettage, and long-term follow-up are absolutely necessary for any recurrence or change to fibrosarcoma. We report a case of AF in a 10-year-old male patient who presented with a chief complaint of swelling in the right mandibular posterior region. Enucleation and curettage were done under general anesthesia, followed by immunohistochemical markers (Ki-67, Mib-1) to assess the sarcomatous changes and aggressiveness of the tumor.

Key words: Ameloblastic fibroma, ameloblastic fibrosarcoma, proliferative markers

Introduction

Ameloblastic fibroma (AF) is an uncommon true mixed odontogenic tumor, with a relative frequency between 1.5% and 4.5% of all odontogenic tumors. It may behave either as a true neoplasm or as a hamartomatous proliferation of odontogenic epithelium of the enamel organ and odontogenic mesenchyme of the primitive dental pulp. It was first described by Kruse in 1891 and later classified as a separate entity by Thoma and Goldman in 1946.

Case Report

A 10-year-old male patient reported to our department with the chief complaint of hard, painless swelling present on the right side of his cheek since 1 month. The swelling was reported to be insidious in onset and no history of trauma was elucidated. Medical history and family history were also noncontributory.

Extraoral examination revealed a diffuse swelling on the right lower third of face measuring 4 cm × 2 cm in size extending superoinferiorly from 1 cm below ala tragus line to lower border of mandible and anteroposteriorly from corner of mouth to angle of mandible. On palpation, the swelling was firm in consistency, nonfluctuant, noncompressible, and nontender in character. No ulceration and no drainage were seen.

Intraoral examination revealed the presence of hard swelling in the buccal vestibule, extending from

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distal surface of deciduous canine to mesial surface of permanent first molar with an obvious expansion of buccal and lingual cortex [Figure 1].

Orthopantomogram (OPG) revealed a multilocular, well-defined radiolucency having well-demarcated smooth borders, extending from permanent first molar posteriorly to canine anteriorly. Permanent impacted premolars were present in a line below the roots of deciduous first molar [Figure 2].

Based on the clinical and roentgenographic findings, a presumptive, preoperative diagnosis of dentigerous cyst was made. An intraoral incisional biopsy was done and a section of approximately 1.5 cm of tissue was sent for the histopathological examination [Figure 3].

Histopathologically, the lesion showed bilaminar strands which appeared to divide the cellular stroma into lobules. The epithelium had small, bulbous thickenings or clusters of follicular islands, in which basal cells were tall, columnar and showed reversal of polarity. An inner zone like stellate reticulum was seen in these islands. The mesenchymal component consisted of evenly distributed stellate cells in a loose myxoid to predominantly eosinophilic matrix, resembling the primitive dental papilla with very few cells showing atypical mitoses. No hard tissue structures such as enamel or dentin were detected. The overall features confirmed the diagnosis of AF [Figure 4]. However, due to the presence of a few atypical mitotic cells, markers of cellular proliferation, namely, Ki-67 and Mib-1, were used to determine the aggressive growth potential. The results of Ki-67 and Mib-1 labeling index came out to be approximately 20%–25% (mild).

Hence, complete curettage was done intraorally with extraction of deciduous first molar under general anesthesia (GA). Intraoperatively, lesion was extending anteriorly up to premolars and posteriorly up to buccal plate of lower first molar. The site was packed with iodoform gauze and allowed to heal by secondary intention. After 3 weeks, an acrylic, removable plate was made to cover the defect.

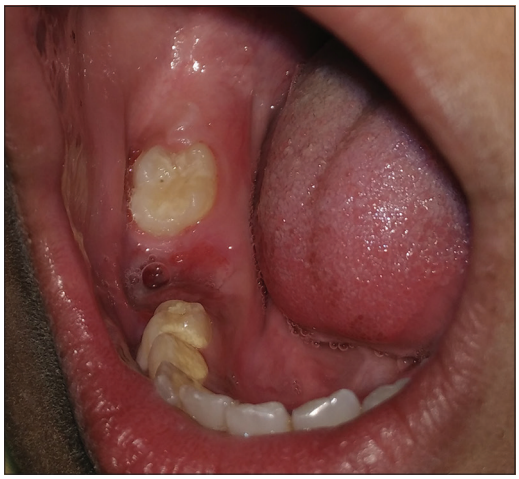

Figure 1: Buccal and lingual cortex expansion

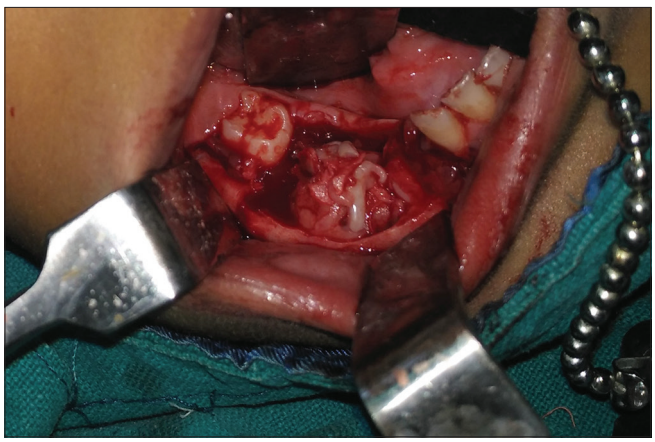

Figure 3: Firm lobular soft tissue mass

Discussion

The ameloblastic fibroma is an uncommon, benign, mixed odontogenic neoplasm.[1] These tumors are frequently diagnosed between the first and second decades of life with slightly higher prediction for males (male:female = 1.4:1).[2]

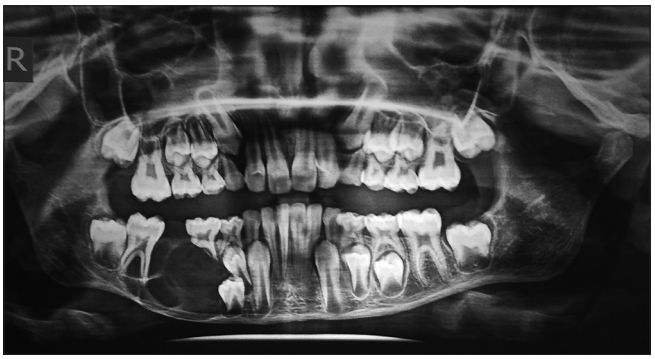

Figure 2: Orthopantomogram showing multilocular radiolucency with impacted premolars

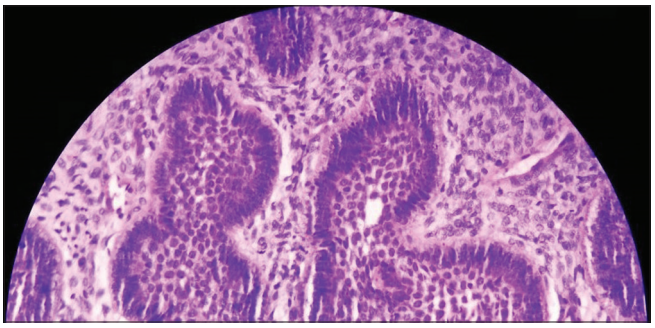

Figure 4: Island of odontogenic epithelium in a primitive connective tissue background resembling dental papilla without the formation of dental hard tissues (H and E, ×40)

Radiographically, they appear unilocular or multilocular radiolucencies associated with impacted tooth, often misdiagnosed as dentigerous cyst. In our case, it appeared as multilocular radiolucent lesion having smooth well-demarcated borders associated with impacted premolars. Intraorally, cortical expansion of the affected bone was observed as increase in the mesiodistal distance between mandibular first molar and adjacent mandibular deciduous second molar was evident. All of these findings were in concurrence with the literature suggesting it to be often diagnosed as dentigerous cyst.[3,4]

However, other entities such as odontogenic keratocyst, ameloblastoma, AF, or ameloblastic fibrosarcoma were also kept in differential diagnosis.[3]

Histologically, the epithelial component occupied the mesenchymal stroma in various patterns such as thin long strands, cords, nests, or islands. Unlike the strands in ameloblastoma, the strands in AF exhibit double or triple layer of cuboidal cells,[5] which was seen in histopathological details of our case. Hence, the diagnosis of AF was confirmed. Since a few atypical mitotic figures were reported in the biopsy, we got the proliferative markers (Ki-67 and Mib-1) done. They were found to be in mild-labeling indices suggesting of less aggressive tumor. Hence, we considered a conservative approach for the treatment.

Treatment options for AF swings between conservative excision and aggressive resection. Philipsen *et al*.[1] proposed that the innocuous behavior of the lesion does not justify aggressive initial treatment but rather meticulous surgical enucleation with close clinical follow-up.[6] While uncommon, the possibility of malignant transformation of AF into ameloblastic fibrosarcoma is well documented.[7] Furthermore, the recurrence rate after reviewing the literature with 85 cases of AF by Trodahl *et al*. and Zallen *et al*. was found to be 43.5% and 18.3%, respectively.[8,9] Hence, an aggressive surgical treatment is often suggested by some authors. No matter whatever the reason of recurrence is, a long-term follow-up is definitely necessary.[8,10] In our case keeping in view the biopsy report suggesting of AF, an intraoral excision and thorough curettage of the surrounding bone followed by the removal of first and second deciduous molars were done under GA.

However, mandibular first and second premolars were not found to be involved with pathology, so intraoperative decision to retain both mandibular premolars was taken. It is important to note that the further eruption of these teeth would be done by surgical orthodontics. In addition, we found pocketing of buccal cortex in relation to mandibular first molar without involving lingual cortex. Since complete removal was done from this site too, we did not remove permanent mandibular first molar. Iodoform roll gauze dressing was done followed by weekly regular follow-ups. After 3rd week, a removable plate to cover the defect was given.

Management of AF has been ranging from conservative to aggressive resection protocols. Since it is difficult to label the recurrence potential and transformation of AF into sarcoma, the proliferation markers such as Ki-67 and Mib-1 would be a good option in evaluating its growth potential and further helping in planning treatment options for the management of tumor.

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Conflicts of interest
There are no conflicts of interest.

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