

MANDIBULAR AMELOBLASTIC FIBROMA – A CASE REPORT

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Abstract

Ameloblastic fibroma is a rare odontogenic tumour of the jaw, in which both the epithelial and ectomesenchymal elements are neoplastic and is commonly seen in the first two decades of life. The common clinical manifestation is a slow-growing swelling. Ameloblastic fibroma is most common in adolescents and young adults, and is generally found in the posterior mandible. Ameloblastic fibroma is diagnosed on routine radiographic evaluation and is clinically and radiographically similar ameloblastic fibroodontoma or odontoma making the accurate diagnosis mandatory. There is a controversy in literature whether the treatment should be conservative or aggressive. A conservative treatment strategy such as enucleation and curettage is usually sufficient. However the extensive lesion requires the radical treatment. The objective of the present study is to report a clinical case of a surgical treatment of ameloblastic fibroma involving the body of mandible by enucleation in a 19 year old female patient. The tumor responded to the surgical treatment and there was no recurrence one year postoperatively.

Key words: Ameloblastic Fibroma, Enucleation, Odontogenic Tumor.

Introduction

Ameloblastic fibroma (AF) is a relatively uncommon neoplasm of odontogenic origin comprising about 1.5-4.5% of all odontogenic tumors.¹ It is characterized by simultaneous neoplastic proliferation of mesenchymal and epithelial components, with no formation of dental hard tissues.² Ameloblastic fibromas are composed of proliferating odontogenic epithelium embedded in a cellular ectomesenchymal tissue resembling dental papilla.³

It was first described by Kruse (1891) and later classified as a separate entity by Thoma and Goldman (1946).^{4,5} It is reported in a wide age range (0.5–62 years), most of the cases are seen in the first two decades of life with 77.7% of cases being diagnosed before the age of 20. Males show slightly higher prevalence with Male:Female ratio of 1.4:1.⁵ It can occur either in the mandible or maxilla but over 80% of tumours occur in the posterior mandible.^{6,7}

Clinically the tumor grows slowly and painlessly, expanding the jaw. Radiographically, it appears as a unilocular/multilocular area of radiolucency with a smooth outline.⁸ Grossly, it appears as a solid, soft tissue mass with a smooth surface. It may or may not be encapsulated.⁹ Histologically, the tumor consists of strands and groups of epithelial cells in a connective tissue background and does not invade bone.⁸

Treatment of AF in general is a conservative approach, such as enucleation with curettage of the surrounding bone along with the removal of the affected tooth.^{4,10} Long-term followup is necessary.¹¹

In this article we report a case of ameloblastic fibroma in posterior region of mandible involving the premolar and molar tooth in the body of the mandible.

Case Report

A 19 year old female patient has come to the department of oral and maxillofacial surgery at our institution with a chief complaint of pain associated with swelling in her lower left jaw region since 3 years. Patient gives history of swelling

which was slowly increasing in its size since it noticed and also has difficulty in mastication from the left jaw since 6 months. Patient also gives a history of lesion changing its consistency from soft to hard over a period of time. The medical history was non contributory with no history of trauma to the teeth or jaw. Personal history revealed that patient was vegetarian in diet and there was no history of any deleterious habit like smoking, tobacco or betel nut chewing, alcohol etc.

On extra oral examination, the upper and middle 1/3rd of the face appears to be apparently normal. Facial asymmetry is seen wrt lower left side of the face with a swelling measuring about 2cm x 1.5 cm in diameter in the region of body of mandible. (Figure 1)



Figure 1: -Extraoral swelling seen in the lower left side of the face, body of mandible region.

On palpation it was firm in consistency, tender, non-compressible, non-fluctuant and afebrile to touch. Intraoral examination revealed that there is a gross swelling that extends from distal surface of 33 to mesial surface of 37 with the obliteration of buccal vestibule. Overlying surface was of same colour as that of surrounding mucosa. Teeth were carious wrt 35, 36 and displacement was seen wrt 34, 35, 36 with calculus and stains present. (Figure 2) On palpation it was firm in consistency, tender and no discharge was present. Aspiration was negative which indicates non cystic lesion.



Figure 2: - Intraoral examination reveals a gross swelling with the obliteration of buccal vestibule.

With the above clinical findings, provisional diagnosis of unicystic ameloblastoma was given with differential diagnosis of ameloblastic fibroma.

Radiographic examinations were performed OPG revealed a oval, well defined, unilocular radiolucent lesion, with sclerotic radioopaque margin located in the left mandibular region between the roots of distal surface of canine and mesial surface of 2nd molar measuring approximately 2.5 cm x 2 cm in diameter. The divergence of root was also observed wrt 34, 35, 36 (Figure 3).

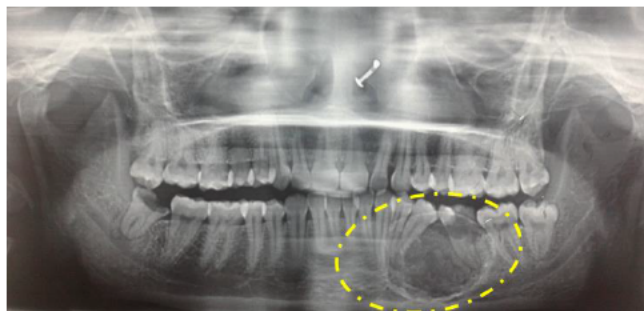


Figure 3: - OPG revealing the shape, size and extent of the lesion.

Occlusal view reveals expansion of the buccal and lingual cortical plates (Figure 4). Radiographic diagnosis ameloblastic fibroma was given .



Figure 4: - Occlusal view showing the expansion of the buccal and lingual cortical plates.

Routine base line investigations were performed, which were non-contributory. After taking the informed consent of the patient complete surgical enucleation of the lesion

along with the extraction of the involved teeth #34, #35 and #36 was done under general anaesthesia (Figure 5) and excised specimen grossly, appeared as firm, lobular soft tissue mass with a smooth surface (Figure 6) was sent to histopathological examination of the specimen reveals odontogenic epithelium in a sharply defined strands and islands of cell which are tall columnar cells resembling ameloblast, a less conspicuous area of which resemble the stellate cells, in the mesenchymal tissue, suggestive of ameloblastic fibroma (Figure 7)



Figure 5: - Surgical enucleation of the lesion

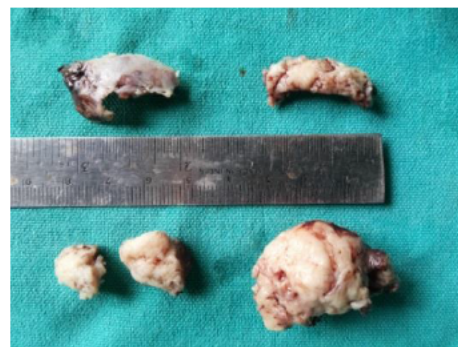


Figure 6: - Gross specimen of the lesion obtained following surgical enucleation

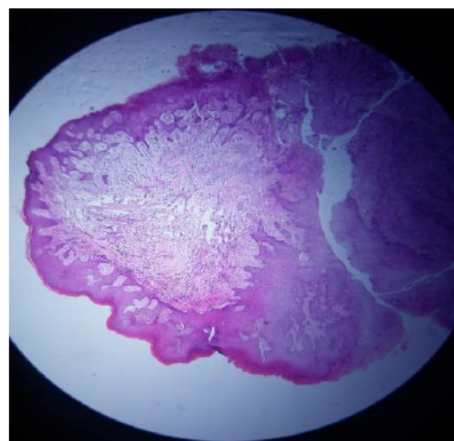


Figure 7: - Histopathological examination of the specimen Based on the clinical, radiographic and histological findings final diagnosis of ameloblastic fibroma was given.

The patient was followed up regularly for a period of 1 year and the healing was found to be satisfactory with no tendency for recurrence.

Discussion

World Health Organization (WHO 1992) classifies Ameloblastic Fibroma as a true mixed neoplasm of odontogenic origin with both epithelial and ectomesenchymal components, without hard tissue formation.¹² The ameloblastic fibroma is a rare odontogenic tumor comprising of approximately 2% of all odontogenic tumors^{13,14} found primarily in children and adolescence, the youngest patient reported is a 7 week old infant.¹⁵ It has been diagnosed at a mean age of 15 years¹⁵ with males being more affected than females with slight male predilection.^{16,17} The tumor occurs exclusively in the first and second decades of life^{14,16}.

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 complaint¹⁸. An impacted tooth may be associated with the tumor in approximately three quarters of the cases.^{13,17}

The similar finding was seen in our case where a patient with a complaint of pain and swelling in the left mandibular premolar molar region in her 2nd decade of life.

The exact pathogenesis is not clear. The tall columnar ameloblast-like cells in the epithelial component are too primitive to induce the cells of the ectomesenchyme, and only little is known about their interactions. It is also unknown why induction of odontoblastic differentiation is lacking in AF.⁵ Immunohistochemical analysis shows positive staining of odontogenic epithelium for cytokeratin, mesenchymal tissue around the dental lamina-like epithelium for tenascin, focal areas of immature dental papilla-like cells, and basement membrane of the epithelium for vimentin. These findings suggest that AF develops at an early stage of tooth formation.⁴

Ameloblastic fibroma radiographically presents as a unilocular or multilocular radiolucency with a smooth, well-defined periphery. Associated features may include unerupted or displaced teeth, divergence of the roots of adjacent teeth, or expansion of the cortical plates.¹⁸ The presented case revealed the similar radiographic features such as well defined, unilocular radiolucency, with sclerotic radioopaque margin. The divergence of root and the expansion of the buccal and lingual cortical plates were observed.

Grossly, ameloblastic fibroma appears as firm, lobular soft tissue mass with a smooth surface. If a tooth is associated with the lesion it may accompany the specimen. A capsule is generally not appreciated.¹⁴

Histologically, the ameloblastic fibroma is characterized by the proliferation of odontogenic epithelium supported by a primitive mesenchymal connective tissue stroma.^{19,20} 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.¹⁸ The above finding was in line with our case where histological features of the lesion represented odontogenic epithelium in a sharply defined strands and islands of cell which are tall columnar cells resembling ameloblast, a less conspicuous area of which resemble the stellate cells, in the mesenchymal tissue.

Treatment of ameloblastic fibroma by conservative enucleation or by aggressive modality is still a topic of debate. A recurrence rate of 43.5% and 18% was reported by Trodahl and Zallen, respectively. Gundlach was of the opinion that simple enucleation would not be sufficient for AF.⁵ Most of them agree for a conservative surgical approach initially, and a more aggressive excision for recurrent lesions, larger tumors, or those involving maxilla.^{5,9} The extraction of the involved teeth should be done along with the excision of the lesion. A long-term followup is recommended.¹¹ Total excision of extraosseous AF with no recurrence is reported.²¹ In the present case the surgical enucleation of the lesion was done along with the extraction of the involved tooth.

Regardless of the treatment done, the patient of ameloblastic fibroma should be followed up for a long period to enable early detection of the possible recurrence or development of ameloblastic fibrosarcoma, which is a malignant counterpart of ameloblastic fibroma. In the case described here the patient has been followed up for 1 year with no signs of recurrence or sarcomatous transformation, thereby demonstrating success of the treatment.

Conclusion

Ameloblastic fibroma is a rare odontogenic tumour. It consists of odontogenic ectomesenchyme resembling the dental papilla and epithelium resembling dental lamina and enamel organ without dental hard tissues. It mainly affects the posterior mandible. It has been the subject of much debate over years with respect to its treatment modalities. The treatment plan varies from conservative surgical excision that includes enucleation and curettage to the radical treatment. The type of treatment chosen depends on several factors like patient age, lesion location and size and whether the lesion is primary or recurrent. In this case the obvious advantages of our treatment technique were the Complete removal of the pathologic lesion, reduction of the potential for recurrence as the case was regularly followed for 1 year and no signs of recurrence was seen and Preservation of the continuity of the mandible, thus maintaining jaw function and shape.

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