Indian Journal of Medical Case Reports ISSN: 2319–3832
Online, International Journal, Available at http://www.cibtech.org/jcr.htm
2021 Vol.10, pp. 51-55/Pourna and Murali
Case Report (Open Access)

ODONTOGENIC FIBROMYXOMA IN AN EARLY ADOLESCENT – A RARE CASE REPORT AND REVIEW OF LITERATURE

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ABSTRACT
Odontogenic fibromyxoma is a rare benign odontogenic tumor of mesenchymal origin. It is common among young adults with a slight female predilection. It is unencapsulated, locally aggressive tumor with high recurrence rate. It occurs commonly in mandible than maxilla. Its histological and radiological features make it difficult to differentiate from other odontogenic tumors. This case report presents a case of odontogenic fibromyxoma of mandible in a 12-year-old female patient.

Keywords: *Odontogenic Fibromyxoma, Mesenchymal Tumor, Benign, Mandible*

INTRODUCTION
Odontogenic fibromyxoma (also called odontogenic myxofibroma) is a relatively rare benign tumor of mesenchymal origin that may contain odontogenic epithelium. It accounts for only 3–6% of all odontogenic tumors and cysts of the jaws. The common site of occurrence is in the order of posterior mandible, anterior mandible, and maxilla (Trehan *et al.,* 2020). The tumor most frequently arises in the second to fourth decades of life with female predilection. The tumor usually presents as a slow-growing painless swelling; however, patients may be symptomatic with pain and paresthesias in advanced cases. Loosening of the teeth is relatively common and it may be associated with impacted or unerupted tooth (Kaur *et al.,* 2020). Radiographically, majority of the cases present as expansile mass demonstrating unilocular or multilocular radiolucency with or without scalloped borders (Chauhan and Guruprasad, 2012). This case report presents a rare case of odontogenic fibromyxoma of mandible with impacted tooth in a 12-year-old female patient.

CASE
A 12-year-old female patient presented to our department with the chief complaints of swelling in the lower front tooth region for past 5 months. The swelling was gradual in onset and slowly progressed to its present size. There is no associated pain and pus discharge. Patient had no significant medical history. Extraoral examination revealed no facial asymmetry and mouth opening appeared normal. Intraoral examination revealed a well-defined swelling of size 2.5 X 1.5cm in the left mandibular canaine region extending from the distal aspect of 73 to the mesial aspect of 31 obliterating the labial vestibule. On palpation, the swelling was hard in consistency and non-tender. There was mobility in the associated teeth 31, 32, 73 and 74.
Based on the history and clinical findings, the provisional diagnosis was made as benign odontogenic tumor. The differential diagnosis was suggested as dentigerous cyst and adenomatoid odontogenic tumor.

The radiographic examination comprised of Intraoral periapical radiograph and Digital orthopantomogram. The IOPA radiograph and Orthopantomogram revealed ill-defined mixed radiopaque-radiolucent lesion extending from distal to 32 to mesial to 34 with underlying impacted 33. Radiographically, the differential diagnosis was dentigerous cyst and adenomatoid odontogenic tumor.

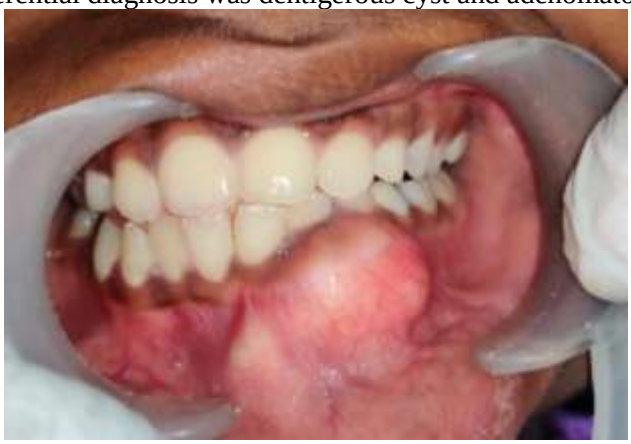

Figure 1: Preoperative image

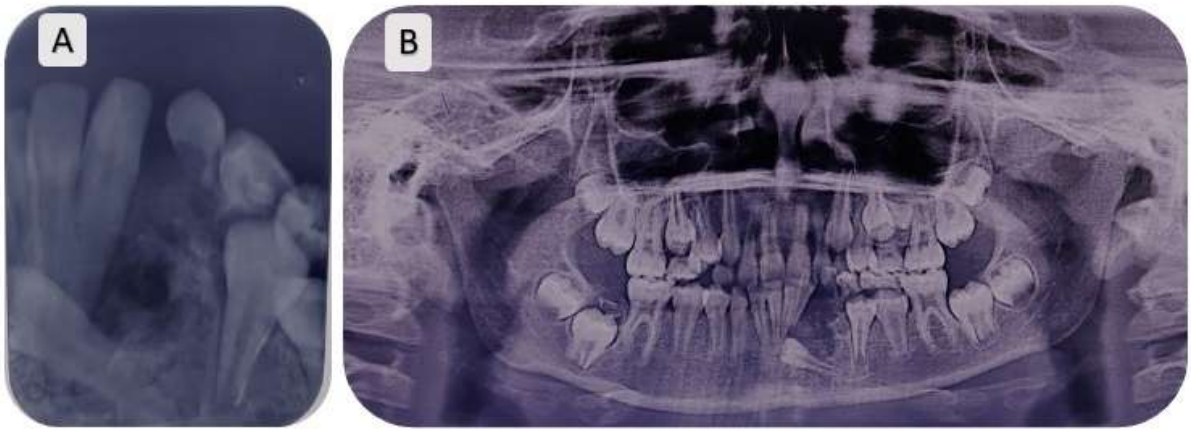

Figure 2: Radiographs showing mixed radiopaque-radiolucent lesion with impacted tooth
A) Intraoral Periapical radiograph B) Orthopantomogram

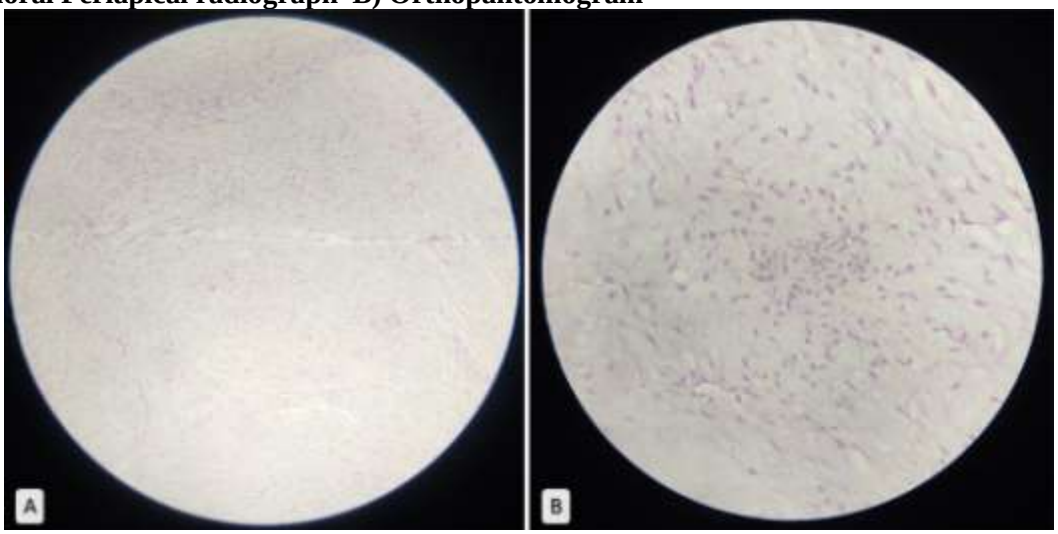

Figure 3: Histopathologic photomicrograph showing fibrous and myxoid stoma.
A) Under 10X magnification B) Under 40X magnification

The histopathological examination revealed fibrous and myxoid matrix in lobular configuration with suspended spindle to stellate shaped cells. The cellular elements were arranged in short fasicules without prominent atypia. The features were suggestive of Odontogenic fibromyxoma. The presented case was concluded as Odontogenic fibromyxoma.
Based on the clinical findings, radiographic features and histopathology, the tumor was planned for enucleation. The follow-up was continued for the period of 3 months post-surgery and there were no complaints from the patient.

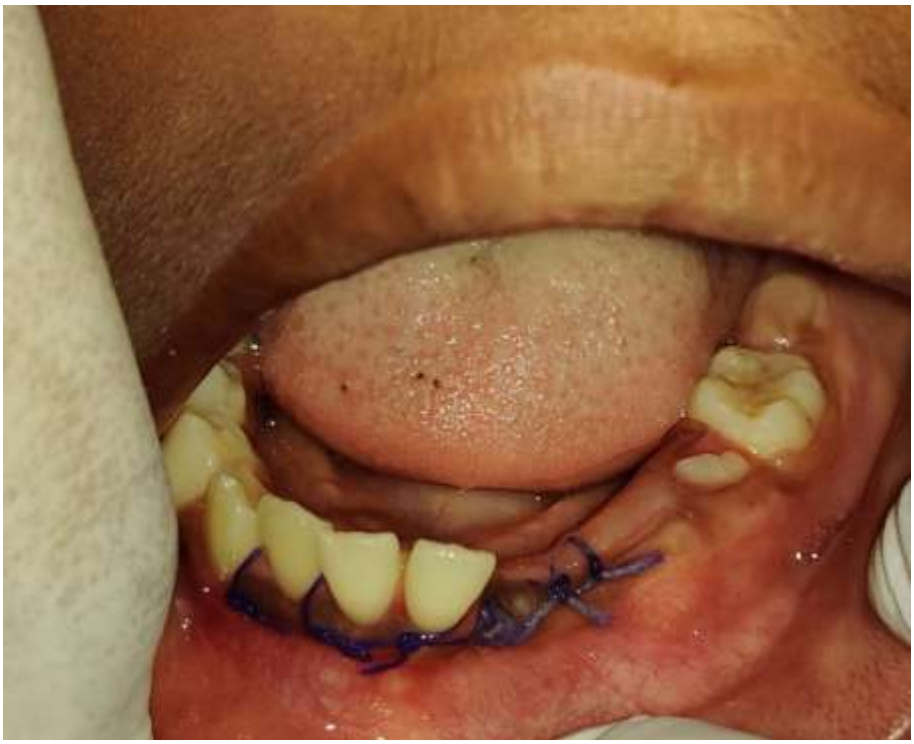

Figure 4: Post-operative image

DISCUSSION

Rudolph Virchow used the term "myxoma" first in 1863 to describe abdominal and soft-tissue lesions. In 1947 Thoma and Goldman described it as "a rare benign tumor of the tooth-bearing areas of the jaw bone" (Kaur *et al.,* 2020). While the World Health Organization uses the terms "myxoma" and "fibromyxoma" interchangeably, some authors intimate that fibromyxomas tend to have more prominent collagen fibers than myxomas (Alhousami *et al.,* 2018). These are two forms of fibromyxomas identified in the head-and-neck region obtained from the facial skeleton and the facial soft tissues, respectively (Kaur *et al.,* 2020).
Odontogenic fibromyxoma is a relatively rare intraosseous, benign odontogenic tumour which is locally aggressive. In the pathogenesis of Odontogenic Fibromyxoma, dental follicle, dental papilla, and periodontal ligament tissue have been implicated as possible germ centers (Kaur *et al.,* 2020). They exhibit a female to male ratio of 1.5:1 and are two times as common in the mandible as the maxilla. They can arise wherever in the jaws but have a strong predilection for the molar and premolar regions. Odontogenic myxomas are relatively less common in children under 10 years of age (Alhousami *et al.,* 2018). They are exclusively found in the tooth-bearing areas of the jaw and are usually located centrally in the mandible. Soft-tissue localization of the tumor is quite rare and is categorized as peripheral myxoma. Peripheral myxoma is slow growing, less aggressive with low recurrence rate, as compared to the central myxoma (Jain and Reddy, 2013). The intraosseous peripheral occurrences of the tumor in the

long bones have also been reported. These peripheral lesions exhibit a higher tendency of recurrence and malignant transformation (Kaur *et al.,* 2020). The discussed case was of central variety located in the canine-premolar region of the mandible and the features were consistent with the literature.

The radiographic presentation of fibromyxoma is highly variable. It has been reported in the literature as unilocular radiolucency, multilocular radiolucency, and mixed radiolucency and radio-opacity with multilocular pattern with sclerotic border as most common (Rakesh *et al.,* 2012). The multilocular radiolucencies demostrate different presentations including "soap bubble", "tennis racquet", "honey comb", "wispy", or "spiderweb" patterns. Though radiolucency is the most common pattern of this tumor, radiopaque or mixed radiolucent and radiopaque pattern has also been reported to some extent in the literature (Alhousami *et al.,* 2018).

The presented case radiographically shows mixed radiolucency and radiopacity with an impacted tooth and additionally displacement of adjacent tooth has been seen.

Histopathologically, odontogenic fibromyxoma has varying amounts of myxoid and fibrous connective tissue. Accurate pathologic diagnosis is essential to ensure proper management. It is commonly marked by randomly arranged stellate, oval or spindle-shaped cells in a myxoid stroma along with myofibroblasts and dense collagen fibres which were seen in the present case too (Kaur *et al.,* 2020).

The treatment of choice for this tumour is surgery since it is neither radio sensitive nor chemo sensitive. The surgical option of the fibromyxoma varies from a conservative local excision or curettage to a more radical en bloc resection (Trehan *et al.,* 2020). The tumour is associated with a high recurrence rate ranging from 10% to 43% with a mean of 25%, and this is attributed to its myxomatous nature, lack of capsule and infiltrative growth pattern. It has been reported that recurrence was slightly higher in maxilla when comparing to mandible. Although the tumor has high recurrence rate, conservative management could be considered first to avoid both morbidity and resection associated effects on the quality of life(Rakesh *et al.,* 2012). In our case also, conservative approach of local excision of the tumor was done with a consideration of patient's age and to preserve the function and esthetics.

CONCLUSION

Odontogenic fibromyxoma is a rare pathology in the pediatric ages. Though the tumor is not life threatening, its aggressiveness severely affects the quality of life. So the complete removal of the tumor along with the involved tooth is recommended. Even after a satisfactory surgical management, long-term follow up is suggested in view of the high tendency of the tumour to recur. A minimum of five years of surveillance is essential to confirm that the lesion has completely healed, and periodic clinical and radiographic follow up should be carried out indefinitely irrespective of the treatment modality applied to the tumor.

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