Case Report

Juvenile Trabecular Ossifying Fibroma: Unusual Radiographic Features

Santosh Kumar Singh[1], Ajay Kumar Pillai[2], Swapnil Moghe[3], Saksham Nahar[4], K. Kavitha[5]

[1]MDS, Sr. Lecturer, Department of Conservative dentistry, People's University, Bhopal, Madhya Pradesh, India, [2]MDS, Associate Professor, Department of Oral & Maxillofacial Surger, Peoples Dental Academy, Bhopal (M.P.), India, [3]PG Student, Department of Oral & Maxillofacial Surgery, People's Dental Academy, Bhopal, Madhya Pradesh, India, [4]PG Student, Department of Oral & Maxillofacial Surgery, People's Dental Academy, Bhopal, Madhya Pradesh, India, [5]Consultant Dental Surgeon Chowdhary Hospital, Bhopal, Madhya Pradesh, India

Corresponding Author: *Dr. Ajay Kumar Pillai, Department of Oral & Maxillofacial Surgery; People's University, Bhopal, Madhya Pradesh, India. E-mail- drajaypillaiomfs@gmail.com*

Abstract

Ossifying fibromas are uncommon benign lesion with aggressive local growth. These tumors are believed to originate from the periodontal ligament. Juvenile ossifying fibromas (JOF) were classified into juvenile pasammomatoid ossifying fibroma and juvenile trabecular ossifying fibroma (WHO 2005). These tumors though benign are clinically persistent thereby mandating aggressive ressection. We report a case of JOF of trabecular pattern in a 12 year old girl the treatment was radical en bloc resection.

Keywords: Ossifying Fibroma, Juvenile ossifying fibroma, Trabecular ossifying fibroma

INTRODUCTION

Benign fibro-osseous lesion of head and neck region are uncommon.[1] Because of their intense osteoblastic activity, these tumours are very aggressive and osteolytic nature.[2] However, it has been infrequently reported to be found in the frontal, ethmoidal, sphenoidal and temporal bones as well as the orbit and anterior cranial fossa. The Trabecular variant of juvenile ossifying fibroma was previously described by Reed and Hagy, in 1965.[3] We report a rare case of trabecular JOF of maxilla in a twelve-year-old female patient with its clinical, radiological features, our experience in its management and a brief discussion regarding the case.

CASE REPORT

A twelve-year-old systemically healthy girl, presented with 2 year history of a swelling on the left mid face (Figure 1a, 1b). Perceived 2 years before this consultation, the swelling was as a small peanut sized growth which gradually increased to the present size of 2.5 X 3 X 2 cms. Swelling was diffused and asymptomatic. There was no anaesthesia or paraesthesia of the upper lip, cheek, or jaw, and there was no history of trauma. Intra oral examination revealed the lesion to extend from the region of the left anterior maxilla to the ipsilateral first molar and superiorly from upper buccal vestibule extending inferiorly to the level of attached gingiva. The buccal cortical plate was expanded with no apparent mucosal changes. The swelling was hard in consistency with no signs of fluctuation. No orbital or nasal deformity was observed. However, there was mild facial asymmetry.

Palpation revealed the swelling to be firm with no associated tenderness or rise in local temperature. Intra orally, a solitary swelling, covered by normal mucosa, about 4x3 cms, oval in shape was seen extending from 21 to 42 (Figure 2,). It felt multi lobular, hard in consistency and there was slight obliteration of the buccal vestibule. The teeth in the affected area were normal with no signs of nonvitality. No lymphadenopathy was detected and mouth opening was normal.

Intraoral periapical and occlusal radiographic examination revealed a lesion displaying mixed radio density in the 22, 23 region causing root divergence(Figure 2a,2b). The orthopantomograph examination revealed an image showing displacement of teeth 22, and 23 with no evidence of root resorption of teeth was seen. The centre of the lesion appeared radiolucent with flecks of radiopaque foci, while the margins were more radiopaque. The lesion extended from the bone crest and fanning out laterally to approximately 3 cm above apical level of the adjacent teeth (Figure 3). Computed tomography of the facial bones was

advised which demonstrated a relatively well circumscribed lesion on the left maxilla with no involvement of the nasal septum, Maxillary antrum and left orbital floor. This heterogeneous tumour was of measuring 2.5×3×2 cm. Aspiration yielded negative results, ruling out any cystic lesion. As the lesion was asymptomatic and slow growing, and with no associated neurological symptoms, the diagnosis was concluded to be a benign neoplastic process. Ossifying fibroma, Pindborg tumour, the odontogenic adenomatoid tumour, cemento-ossifying fibroma The mixed odontogenic tumours, odontoma and ameloblastic fibro-odontoma also occur in younger patients and present as mixed density mass lesions in the tooth bearing portions of the jaws. The incisional biopsy was performed under local anaesthesia. Histopathological examination revealed that the tumour is composed of cell rich stroma with highly active proliferative fibroblast, abundant multi nucleated giant cells and many bony trabecullae which was lined by osteoblasts. These features were suggestive of Trabecular variant of juvenile Ossifying fibroma.

Under general anaesthesia, the tumour was exposed and enucleation of was performed (Figure 4) with a clear demarcation was evident between tumour and healthy bone. The postoperative course was uneventful and there were no signs of recurrence after a periodic follow-up of 6 months (Figure 5).

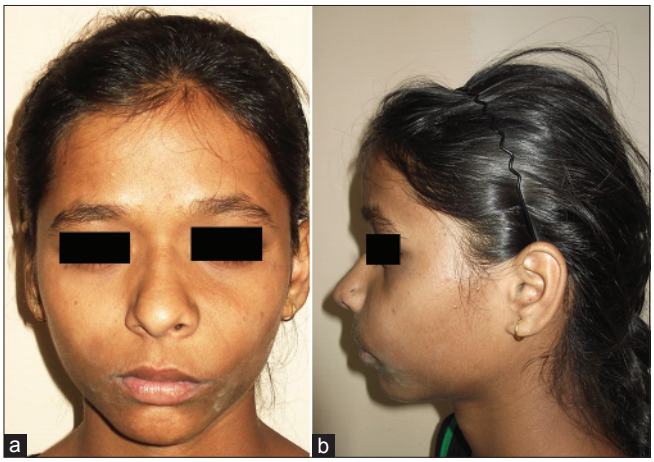

Figure 1: (a) Front view of swelling of left side of face. (b) Lateral view of swelling of left side of face

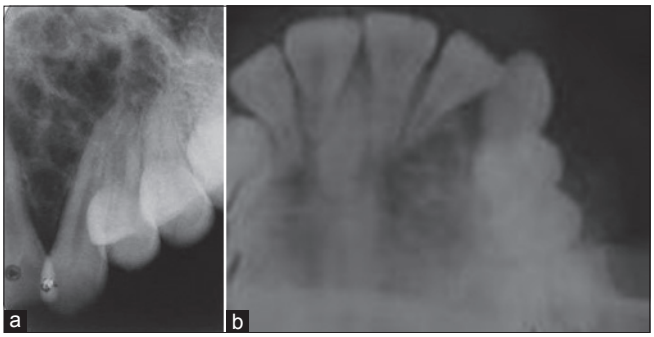

Figure 2: (a) Intraoral periapical radiograph showing mixed radiodensity in 22, 23 region with root divergence. (b) Occlusal radiograph showing root divergence & mixed radiodensity in 22,23 region

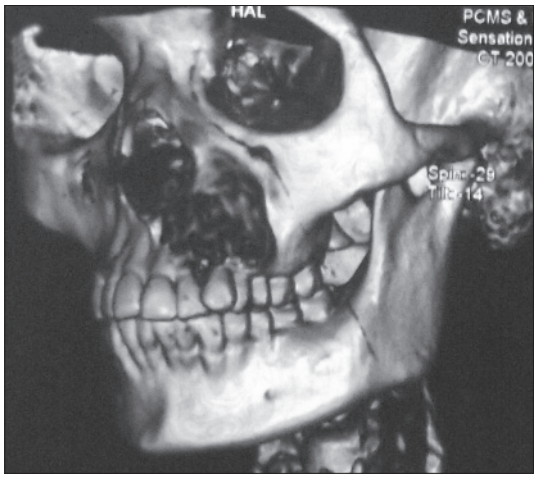

Figure 3: 3D CT Showing relatively well circumscribed lesion on the left maxilla

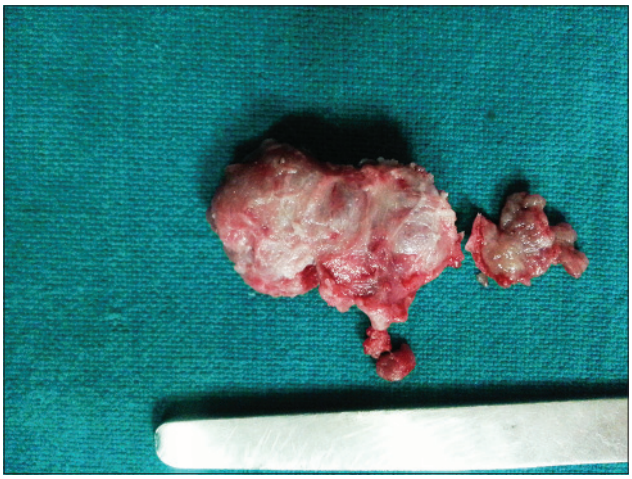

Figure 4: Excised tumor specimen

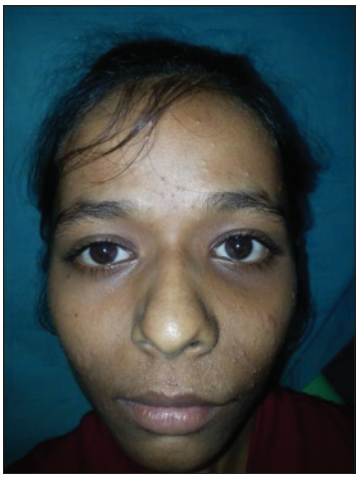

Figure 5: Postoperative picture after follow up

DISCUSSION

Ossifying fibroma is an uncommon benign osteogenic neoplasm, accounting for 2% of oral tumours in children.[4] Juvenile ossifying fibroma behaves aggressively and has a high recurrence rate when not treated adequately, the correct treatment being en bloc resection with free surgical margins. The juvenile form could be distinguished from ossifying fibroma by the following features: earlier onset (at childhood or adolescence), locally aggressive growth and osteoid trabeculae on histological examination.[1]

Regezzi and Sciubba[5] stated that multiple Ossifying fibroma are sporadic but there is familial inclination in few cases. They also stated that chromosomal translocations were seen in few instances.

Initially the tumour is asymptomatic, but as it progress in size and invade surrounding bone it causes functional alterations and cosmetic deformities.[6] Displacement of the teeth may be seen but the teeth remain vital and the overlying mucosa is characteristically intact, although most lesions are discovered during routine dental examinations. Centrifugal growth of the tumour usually causes bowing of the inferior border of the mandible but cortical perforation is rare.

In Fibro-osseous lesions, the morphology of the normal bone is replaced by fibroblasts, collagen fibres, and immature osteoid cells. This group comprises of fibrous dysplasia, benign fibro-osseous neoplasms (central ossifying fibroma), and a heterogeneous group of reactive lesions (osseous dysplasias).[5] Juvenile ossifying fibromas are more radiopaque than conventional lesions and they have a 'ground glass' appearance or may form dense lobulated masses.[7] Radiologically, juvenile ossifying fibroma presents as a clearly circumscribed, concentrically expanding, solitary mass with bone density. It is the circumscribed nature of ossifying fibroma which distinguishes it from fibrous dysplasia.

Histologically, these lesions are always benign, composed of highly vascular and fibroblast-rich connective tissue, which produces a calcified substance that often cannot be clearly attributed to either cement or bone. Clumps of osteoblasts are also present. The incidence of juvenile ossifying fibroma is unknown. A literature review revealed 17 cases reported between 2003 and 2010 with a sex-ratio of five females for one male in adults, while a male predominance is observed in the juvenile form.

Complications of the lesions involving the paranasal sinuses includes occular disturbances, intracranial extension, cysts, and recurrences.[8]

As far as the management of these lesions, it is generally agreed that juvenile ossifying fibroma is a locally aggressive lesion and without adequately treated, it has a high recurrence rate. Hence there is no place for simple curettage of the lesion. The treatment should aim for the complete resection with free surgical margins and grafting.

TAKE HOME MESSAGES

1. Juvenile ossifying fibromas occur predominately in young people under the age of 15 years & may grow rapidly.
2. A key feature to differentiate ossifying fibroma from fibrous dysplasia is the pattern of mineralization. The pattern of mineralization in ossifying fibroma varies from place to place, whereas in fibrous dysplasia it is uniform throughout the lesion.
3. The advocated treatment for juvenile trabecular pattern of ossifying fibroma is complete surgical resection and partial/incomplete resection leads to recurrence.

REFERENCES

1. Dominguete P, Meyer T, Alves F, Bittencourt W. Juvenile ossifying fibroma of the jaw. *Brit J Oral Maxillofac Surg*. 2008; 46:480-481.
2. R Breheret, C Jeufroy, E Cassagnau, O Malard. Juvenile ossifying fibroma of the maxilla. *Eur Ann Otorhinolaryngol Head Neck Dis.* 2011; 128(6):317-320.
3. Emerson LP, Alexander M, Job A. Juvenile trabecular and psamomatoid variant of ossifying of the maxilla with secondary aneurysmal bone cyst. *The Internet Journal of Otorhinolaryngology* 2012; 14: 1.
4. Regezzi JA, Sciubba JJ. Oral pathology. 3rd ed. Philadelphia: W.B. Saunders; 1999:357-360.
5. Martín-Granizo R, Sánchez-Cuellar A, Falahat F. Cemento ossifying fibroma of the upper gingivae. *Otolaryngol Head Neck Surg*. 2000;122:775.
6. Gondivkar SM, Gadbail AR, Chole R, Parikh RV. Ossifying fibroma of the jaws: Report of 2 cases and literature review. *Oral Oncology* 2011; 47(9):804–809.
7. Speight P, Carlos R. Maxillofacial fibro-osseous lesions. *Current Diagnostic Pathol.* 2014; 12 (1):1-10.
8. Vaidya A, Chow J, Goldberg K, Stankiewicz J. Juvenile aggressive ossifying fibroma presenting as an ethmoid sinus mucocele. *Otolaryngol Head Neck Surg*. 1998; 119 (6):665-668.

How to cite this article: Santosh Kumar Singh, Ajay Kumar Pillai, Swapnil Moghe, Saksham Nahar, K. Kavitha. "Juvenile Trabecular Ossifying Fibroma: Unusual radiographic features". International Journal of Scientific Study. 2014;1(6):60-62.

Source of Support: Nil, **Conflict of Interest:** None declared.