Juvenile Ossifying Fibroma of Maxilla

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ABSTRACT

Ossifying fibroma (OF) is a rare, benign, non-odontogenic tumor of the jaw which comes under the group of fibro-osseous lesions. Ossifying fibromas of the mandible are more common than in the maxillary region. Juvenile ossifying fibroma has been distinguished from conventional ossifying fibroma on the basis of patient's age, site predilection, and clinical behavior. The lesion should be differentiated from other fibro-osseous lesions as its management varies from surgical enucleation to complete resection. Present report describes the case of juvenile ossifying fibroma of anterior maxillary region in a 7 year old male patient, with a detailed description of clinical, radiographic, histopathologic features, and its surgical management.

KEY WORDS

Fibro-osseous lesions, Juvenile, Maxilla, Ossifying fibroma

INTRODUCTION

Ossifying fibroma (OF) is a benign osseous neoplasm, non-odontogenic in origin, consisting of highly cellular, fibrous tissue containing varying amounts of calcified tissue which may resemble bone, cementum or both.[1] The lesion is believed to be derived from the cells of the periodontal ligament and is usually seen in tooth bearing areas.[2] It is usually associated with craniofacial bones, mandible being the most common site and has a female predilection with a wide age range, with highest number of cases occurring during the third and fourth decades of life.[3,4] Cases with the involvement of the orbit, paranasal sinuses or maxilla have been reported.[5] Once it is completely excised, OF does not usually reoccur.[6] Juvenile ossifying fibroma is an aggressive form of ossifying fibroma which occurs in younger individuals and has been designated as Juvenile central ossifying fibroma and separated into juvenile psammomatoid ossifying fibroma and juvenile trabecular ossifying fibroma.[7]

CASE REPORT

This is a case report of a 7 years old male patient who presented to the Department of Oral and Maxillofacial Surgery, Kathmandu University School of Medical Sciences, Dhulikhel Hospital with the complaint of swelling in the anterior maxillary region for 3 months. History revealed that the swelling was small in size and had gradually increased to attain the present size. The swelling was insidious in onset without any history of trauma or dental pain and was not associated with pus discharge or paresthesia. General examination revealed a moderately built boy with no other systemic problems. His past medical history, family and dental history was non-contributory. Since the patient was asymptomatic and did not show any facial deformity earlier, he did not visit any hospital.

Extra oral examination revealed a slight visible swelling over the left anterior maxillary region with obliteration of the nasolabial fold and raised nasal floor on same side (fig. 1). On palpation, lymph nodes were non tender and not

enlarged. Temperature over the lesion was same as that of the adjacent area. Intraoral examination revealed oval shaped swelling measuring approximately 3X3 cms i.r.t. 21, 22, 23 and obliterating the vestibule (fig. 2). The overlying oral mucosa was intact and normal in appearance. The swelling was non-tender, bony hard in consistency with well-defined margins. The teeth in that region showed no signs of mobility and non-tender to pressure and percussion with no evidence of displacement.

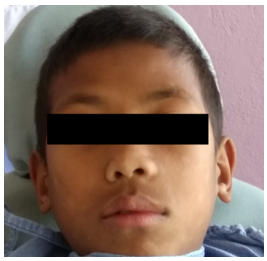

Figure 1. Extra oral photograph showing slight visible swelling over the left anterior maxillary region

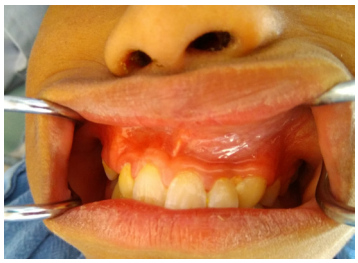

Figure 2. Clinical appearance of patient at intra oral examination

Multi Detector Computed Tomography (MDCT) revealed a well-defined, unilocular radiolucency showing expansible lytic lesion measuring 2.8x2.4x2.2 cm with enhancing component within left paramedian maxillary alveolar process along with displacement of nasal floor (fig. 3).

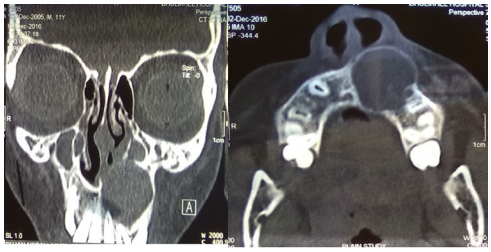

Figure 3. Cut section through tumor mass

Based on the clinical, and MDCT features, a radiographic diagnosis of Adenomatoid Odontogenic tumour (AOT) was made and differential diagnosis considered were calcifying epithelial odontogenic cyst (COC), and Ossifying fibroma (OF).

Incisional biopsy of the lesion was performed under local anesthesia and microscopic examination revealed loose cellular stroma, composed of plump fibroblasts and numerous bony trabeculae showing osteoblastic rimming with few acellular calcifications resembling cementum (fig. 4). Occasionally multinucleated giant cells were also seen. A diagnosis of Juvenile ossifying fibroma was considered.

After obtaining consent from both the patient (patient's guardians), the treatment was planned under general anaesthesia. An intraoral vestibular incision was made from 11 to 25 in most prominent part of the lesion. The mucoperiosteal flap was raised to expose the lesion which was found to be well-encapsulated and measured

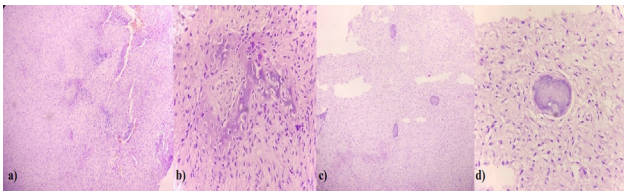

Figure 4. a) Low power view shows loose cellular stroma, composed of plump fibroblasts and numerous bony trabeculae (H and E, x10)

b) High power shows bony trabeculae showing osteoblastic rimming and osteocytes within lacunae (H and E, x40)

c) Low power view reveals acellular calcifications resembling cementum (H and E, x10)

d) High power shows focal acellular calcification (H and E, x40)

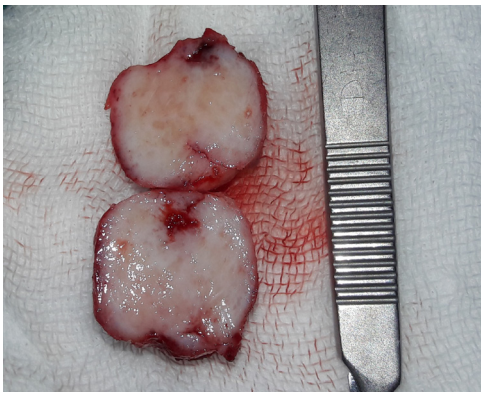

Figure 5. Gross specimen showing a well-circumscribed tumor that shelled out in one piece

aprooximately 3X3 cm. The lesion was removed in toto with no perforations and preserving the nasal floor (fig. 5). Surgical cavity was debrided completely and packed with antibiotic impregnated ribbon gauze. The wound was closed in single layer using 3-0 vicryl sutures and the ribbon gauze was removed on the second post-operative day. Post-operative recovery was uneventful and the patient was discharged on 2nd postoperative day. Periodic follow-up was planned and one-month follow-up did not reveal any symptoms. The patient is under regular follow-up and 3 months post-operatively, has not shown any signs of recurrence.

The histopathology report of the excised specimen confirmed the diagnosis of Juvenile central ossifying fibroma of trabecular variant.

DISCUSSION

Ossifying fibroma, juvenile psammomatoid ossifying fibroma (JPOF), juvenile trabecular ossifying fibroma (JTOF), fibrous dysplasia (FD) and cemento-osseous dysplasia (COD) all belong to the same group of the fibro-osseous lesions. In 1872, Menzel first described central ossifiying fibroma (COF) as a variant of ossifying fibroma or a benign fibro-osseous neoplasm.[8] Ossifying fibroma is considered as a benign neoplasm arising from undifferentiated cells of the periodontal ligament tissues. It can produce bone, cementum, spheroidal calcifications and fibrous tissues,

due to its origin from multipotent mesenchymal cells. In 1971, WHO classified four types of cementum containing lesions: fibrous dysplasia, cementifying fibroma, ossifying fibroma and cemento-ossifying fibroma.[9] According to the second WHO classification, benign fibro-osseous lesions in the oral and maxillofacial region were divided into two categories, osteogenic neoplasm and non-neoplastic bone lesions.[10] Waldron in 1985 sub classified COFs as medullary or periodontal ligamentous in origin.[11] However, the term ''cementifying ossifying fibroma'' was replaced with ossifying fibroma in the new WHO classification in 2005.[12] Based on their pathogenesis of COF, WHO classification of odontogenic tumors in 2005, included juvenile ossifying fibroma occuring in younger individuals as Juvenile COF and segregated into juvenile psammomatoid ossifying fibroma and juvenile trabecular ossifying fibroma.[7]

Although COFs are usually seen in the third and fourth decades of life, our patient was a 7 years old child.[4] Most of the studies have shown prevalence in females, but in our case, the patient was a male.[4] Mandible being the most common site, a few cases in the maxilla have been reported as seen in our case.[13,14] The patients usually present with a painless swelling which may become large enough to cause visible facial deformation, whereas the present case had only a moderate swelling.[15]

As the lesion is slow growing, there is no breach in the bony cortical plates and overlying mucosa or skin. The lesion usually occurs in the tooth bearing area with a centrifugal pattern of growth rather than a linear one which is an important diagnostic feature and is seen as an expansion that occurs equally in all the directions and presents as a round tumor mass clinically.[5]

Histopathologically, COF shows a fibrocellular connective tissue that is well vascularized with presence of immature bony fibrous dysplasia. Our case revealed loose cellular stroma composed of plump fibroblasts and numerous bony trabeculae showing osteoblastic rimming with few acellular calcifications resembling cementum. Occasionally multinucleated giant cells were also seen.

The initial treatment of choice for small COFs is surgical curettage or enucleation, whereas for the large lesion surgical resection is indicated where normal structures including teeth, neurovascular elements and bone should be preserved whenever possible.[13,16] Eversole et al. reported a recurrence rate of 28 % following curettage.[3] Hence, a long term follow-up of the patients is recommended. Our case was treated with surgical enucleation and curettage and the post-operative follow-up revealed normal healing.

We report a case of juvenile ossifying fibroma in a 7 years old child patient who came with a moderate sized swelling on anterior maxilla. Ossifying fibroma of the maxilla is not a common benign tumor and is clinically asymptomatic. A proper correlation of the clinical, radiological as well as histological features is necessary to establish a definitive diagnosis, as well as to classify the FOL lesions. Since chances of recurrence of ossifying fibroma are reported in the literature, long term follow-up of the patients is recommended.

REFERENCES

1. Liu Y, You M, Wang H, Yang Z, Miao J, Shimizutani K, Koseki T. Ossifying fibromas of the jaw bone: a study of 20 cases. *Dento maxillofacial Radiology.* 2010; 39: 57-63
2. Sarwar HG, Jindal MK, Ahmad SS. Cemento-ossifying fibroma-a rare case. *J Indian Soc Pedod Prev Dent.* 2008; 26: 128-131.
3. MacDonald-Jankowski DS. Cemento-ossifying fibromas in the jaws of the Hong Kong Chinese. *Dento maxillofac Radiol.* 1998;27:298–304.
4. Eversole LR, Leider AS, Nelson K. Ossifying fibroma: a clinicopathologic study of sixty-four cases. *Oral Surg Oral Med Oral Pathol.* 1985;60: 505–11.
5. Kashyap RR, Nair GR, Gogineni SB. Asymptomatic presentation of aggressive ossifying fibroma: A case report. *Case Rep Dent.* 2011; 2011:523751.
6. MacDonald-Jankowski DS. Ossifying fibroma: a systematic review. *Dento maxillofac Radiol.* 2009;38:495-513.
7. Slootweg PJ, Panders AK, Koopmans R, Nikkels PG. Juvenile ossifying fibroma. An analysis of 33 cases with an emphasis on the histological aspects. *J Oral Pathol Med.* 1994; 23: 385-88.
8. Cakir B, Karadayi N. Ossifying fibroma in the sasopharynx: A case report. *Clin Imaging.* 1991;15(4): 290-2.
9. Pindborg JJ, Kramer IRH. Histological typing of odontogenic tumors, jaw cysts and allied lesions. In: International histological classification of tumors. Geneva: WHO. 1971; 31–4.
10. Kramer IRH, Pindborg JJ, Shear M. Neoplasm and other lesions related to bone. Histologic typing of odontogenic tumors, World Health Organization. *Berlin, Springer-Verlag.* 1992:28–31.
11. Waldron CA. Fibro-osseous lesions of the jaws. *J Oral Maxillofac Surg.* 1985;43:249–62.
12. Reichart PA, Philipsen HP, Sciubba JJ. The new classification of head and neck tumours (WHO) – any changes? *Oral Oncol.* 2006; 42: 757-8.
13. Cemento-ossifying fibroma of the mandible: Presentation of a case and review of the literature. *J Clin Exp Dent.* 2011;3(1): e66-69.
14. Tamiolakis D, Thomaidis V, Tsamis I. Cemento-ossifying fibroma of the maxilla: a case report. *Acta Stomatal Croat.* 2005;39:319-21.
15. Summerlin DJ, Tomich CE. Focal cemento-osseous dysplasia: a clinicopathologic study of 221 cases. *Oral Surg Oral Med Oral Pathol.* 1994; 78: 611–20.
16. Koury ME, Regezi JA, Perrott DH, Kaban LB. Atypical fibro-osseous lesions: diagnostic challenges and treatment concepts. *Int J Oral Maxillofac Surg.* 1995; 24:162-69.