CASE REPORT

doi: 10.5455/medarh.2016.70.470-472
Med Arch. 2016 Dec; 70(6): 470-472
Received: OCT 15, 2016 | Accepted: NOV 25, 2016

Juvenile Trabecular Ossifying Fibroma of the Maxilla: a Case Report

Samia Aboujaoude[1], **Georges Aoun**[2]

[1]Department of Pediatric Dentistry, Faculty of Dentistry, Lebanese University, Beirut, Lebanon
[2]Department of Oral Pathology and Diagnosis, Faculty of Dentistry, Lebanese University, Beirut, Lebanon

Corresponding author: Dr. Georges Aoun, Associate Professor. Department of Oral Pathology and Diagnosis, School of Dentistry, Lebanese University, Beirut, Lebanon. ORCID ID: http://orcid.org/0000-0001-5073-6882. E-mail: aoungeorges@yahoo.com

ABSTRACT

Introduction: Juvenile ossifying fibromas are uncommon benign tumors. Their aggressiveness added to their high tendency to recur, provoke real diagnostic and therapeutic challenges for the dental practitioner and make a postoperative follow-up over the years indispensable. **Case report:** In this report, we present a case of a seven-year-old girl presented with a swelling in the face at the upper right maxillary region. After clinical, radiological, and histopathological examinations the diagnosis of trabecular juvenile ossifying fibroma was made. The lesion was surgically excised and followed up for two years with no evidence of recurrence.

Keywords: Juvenile, ossifying fibroma, trabecular.

1. INTRODUCTION

Juvenile ossifying fibromas (JOFs) are rare benign fibro-osseous tumors (1). They usually appear under the age of fifteen (2), and concern the facial bones with possible intracranial and orbital extensions (3). Generally, JOFs are asymptomatic, aggressive and of osteolytic nature (4).

Due to their high recurrence rate, early diagnosis, suitable treatment and a long-term follow-up are essential (5).

Histologically, and like in all the ossifying fibromas, the normal bone is replaced by a fibrous cellular stroma containing mineralized bone trabeculae and cementum-like material (4, 6).

According to their prototype of mineralization, JOFs have been classified into two types: the psammomatous characterized by small uniform spherical ossicles resembling psammoma bodies located mainly around paranasal sinuses and orbits, and the trabecular distinguished by fibrous trabeculae and usually affecting the jaws (4).

This report describes a case of a maxillary trabecular JOF of a seven-year-old girl removed surgically and followed up for two years without evidence of recurrence.

2. CASE REPORT

A seven-year-old girl presented to our specialized pediatric dentistry office referred from her dentist for a persistent firm swelling localized at the upper right maxillary region.

Medical and physical examinations revealed a healthy girl with no extra-oral findings except the facial asymmetry. Intra-orally, a painless tumefaction of hard consistency obliterating the vestibule of the right maxilla and extending from the tooth # 53 to the distal of the tooth # 16 was detected (Figure 1).

The overlying mucosa was normal in color and texture. On palpation, no regional lymphadenopathy was noticed.

The panoramic radiograph, done previously by her dentist, showed a well-defined radiolucent lesion in the right maxilla and absence of the tooth # 54 which was extracted as an attempt of treatment for her condition (Figure 2).

In order to assess the bone and to evaluate the extension of the lesion, a

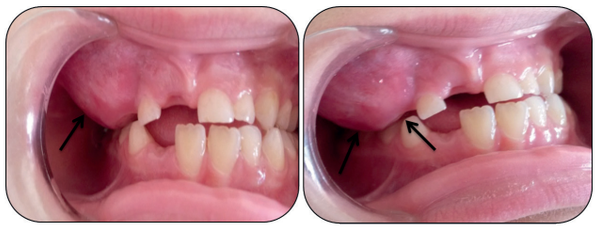

Figure 1. A well-defined swelling obliterating the vestibule and extending from the tooth # 53 to # 16.

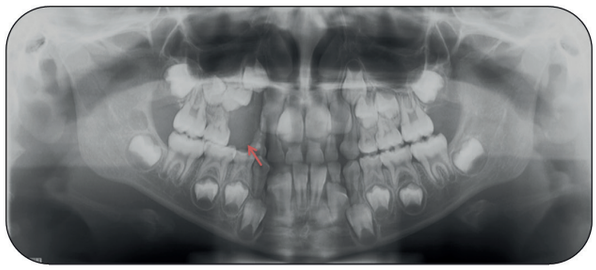

Figure 2. A panoramic radiograph showing a well-defined radiolucent lesion in the right maxilla and absence of tooth # 54.

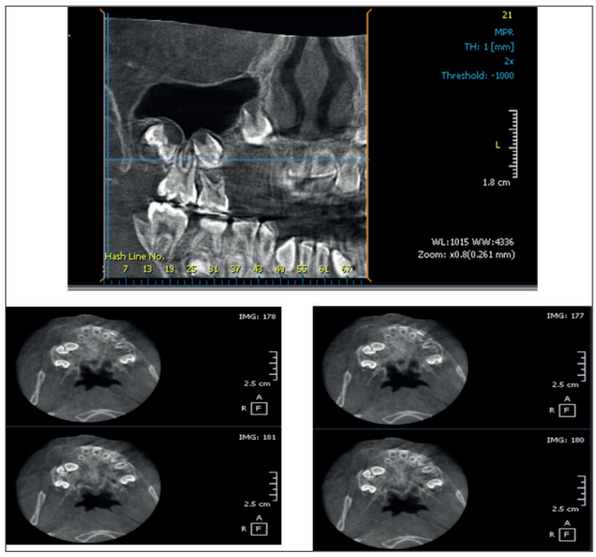

Figure 3. A CBCT radiograph showing a mixed image extending from # 53 to # 55 and from the crestal edge till the germ of the permanent canine; a diffuse border contains fine radio-opaque trabeculae with a buccal, palatal, and crestal expansion of the bone with disruption of the buccal bone.

cone-beam computed tomography (CBCT) radiography was requested.

The CBCT radiograph showed a well circumscribed mixed image, starting distal to the tooth #53 till the mesial of the # 55, and from the crestal edge till the germ of the permanent canine with a diffuse border containing fine radio-opaque trabeculae with a buccal, palatal, and crestal expansion of the bone; a disruption of the buccal bone was also noticed. The image of the right sinus was normal (Figure 3).

An incisional biopsy was performed and sent for histopathologic assessment.

Microscopic examination showed a proliferation of fusiform cells with regular nuclei arranged in bundles and storiform pattern associated with multinucleated giant cells. Within these irregular strands, mineralized pieces in the form of trabeculae were found. The stratified squamous epithelium surface showed moderate acanthosis and hyperkeratosis (Figure 4).

The histological diagnosis of trabeculae juvenile ossifying fibroma was made.

The tumor measuring approximately 1.8 x 2.5 cm was surgically excised under general anesthesia via intra-oral approach. During the procedure, the teeth # 53 and # 55 were removed (Figure 5).

Post-operative recovery was without complications, and the patient was discharged from the hospital 48 hours following the surgery.

Histopathological results of the excisional biopsy were consistent with that of the incisional one.

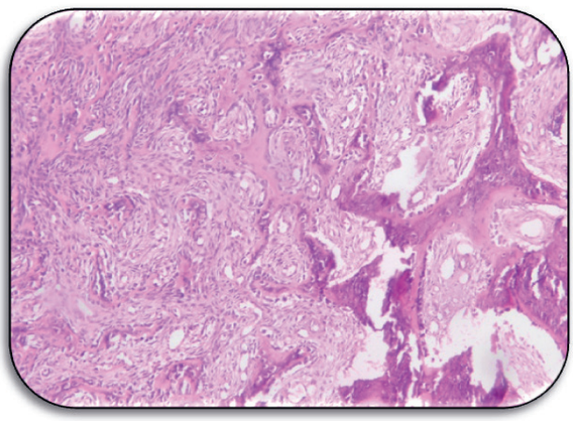

Figure 4. Histopathological section showing mineralized pieces in the form of trabeculae of woven bone.

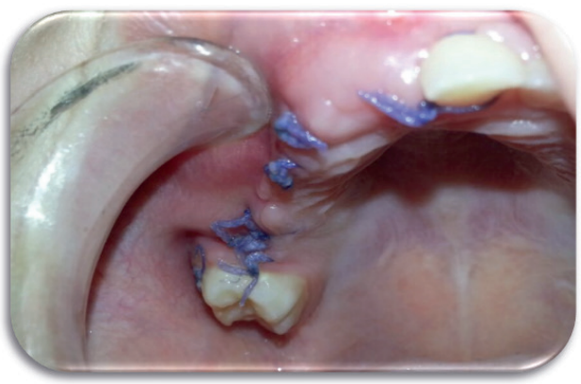

Figure 5. One week postoperative intraoral view (the stitches in place).

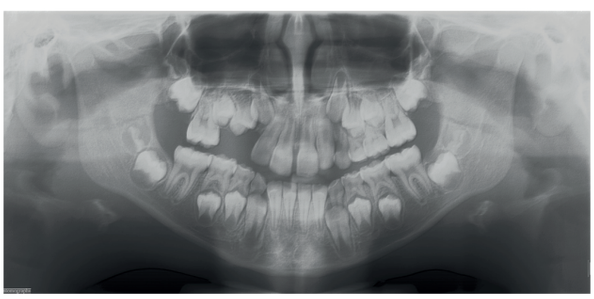

Figure 6. A panoramic radiograph showing new positions of the germs of the teeth # 13, #14, and #15.

The patient was followed for two years, once every six months. She remained recurrence-free clinically and radiologically (Figure 6).

3. DISCUSSION

Juvenile ossifying fibromas are rare aggressive fibro-osseous tumor arising in the craniofacial bones (2).

Usually, JOFs occur under the age of fifteen without any significant gender predilection (5, 7).

Clinically, JOFs are mostly asymptomatic with slow or rapid development leading to facial asymmetry (8).

As for the radiological aspect, JOFs can be seen as uni- or multilocular radiolucencies/mixed radiolucencies (9); rare root resorption can be observed (4).

Cone-beam computed tomography assessment may show well-defined sclerotic borders with a variable amount of calcifications (10).

Histologically, JOFs are lesions characterized by cell-rich fibrous tissue with giant cells and bands of cellular osteoid trabeculae (11).

Juvenile ossifying fibromas are of two types. One is characterized by spherical ossicles, the psammomatoid, and the second less commonly reported, the trabecular, distinguished by trabeculae osteoid and woven bone as seen in our case (4).

Many bone pathologies usually found in the region may constitute a differential diagnosis challenge for JOF. Fibrous dysplasia remains the most prominent condition (1, 2, 12); it can be ruled out as it typically shows normal marginal bone with less cellular stroma and an important amount of lamellar bone instead of woven one (13).

Cemento-ossifying fibroma, a histological variant of JOF, can also be considered; however, giant cells which are evidently found in JOFs are not present in cement-ossifying fibromas (14). Cementoblastoma, osteoblastoma, osteosarcoma, and others entities must be separated as well when diagnosing JOFs (1, 12); this type of separation must be made with the aid of clinical, radiological and histological examinations.

The treatment of JOFs remains controversial. Complete resection, conservative local excision, or enucleation with curettage, are among the treatment techniques (15-17). Abuzinad and Alyamani (2) suggested that conservative management with less aggressive approach must be considered the treatment of choice of JOFs. The same was proposed by Slootweg and Müller (18) who recommended conservative surgery; for them, there are no differences in the results between the JOFs that had limited surgical excision and those removed with major surgery.

On the other hand, Waldron (19) stated that the local excision/curettage of the lesion is the best treatment of JOFs. However, many other authors reported a high recurrence rate after conservative or mini-invasive treatment (in 30–56% of cases) (15, 20, 21), and thus, a complete surgical resection remains the preferred line of treatment (1, 5, 12, 22).

It is to be noted that whatever the surgical technique was, a long-term postoperative surveillance is essential (1, 2, 7, 12, 23). In our case, we have presented a trabecular JOF localized in the right maxilla of a seven-year-old girl. After histological and radiological confirmations, a complete surgical resection was performed with no recurrence till date.

4. CONCLUSION

Juvenile trabecular ossifying fibroma is a rare tumor with a rather high risk of recurrence. A careful evaluation of the clinical, radiological, and histological components of this lesion is needed to surmount the diagnostic and therapeutic challenges connected with it. Furthermore, its early diagnosis and adequate treatment consisting in a complete surgical excision followed by long-term follow-up of the patient is indispensable.

REFERENCES

1. Osunde O, Iyogun C, Adebola R. Juvenile aggressive ossifying fibroma of the maxilla: A case report and review of the literature. Ann Med Health Sci Res. 2013; 3(2): 288-90.
2. Abuzinada S, Alyamani A. Management of juvenile ossifying fibroma in the maxilla and mandible. J Maxillofac Oral Surg. 2010; 9(1): 91-5.
3. Khademi B, Niknejad J, Mahmoudi J. An aggressive psammomatoid ossifying fibroma of the sinonasal tract: report of a case. Ear Nose Throat J. 2007; 86(7): 400-1
4. El-Mofty S. Psammomatoid and trabecular juvenile ossifying fibroma of the craniofacial skeleton two distinct clinicopathologic entities. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2002; 93(3): 296-304.
5. Sun G, Chen X, Tang E, Li Z, Li J. Juvenile ossifying fibroma of the maxilla. Int J Oral Maxillofac Surg. 2007; 36(1): 82-5.
6. Brannon RB, Fowler CB. Benign fibro-osseous lesions: a review of current concepts. Adv Anat Pathol. 2001; 8(3): 126-43.
7. Keles B, Duran M, Uyar Y, Azimov A, Demirkan A, Esen HH. Juvenile ossifying fibroma of the mandible: a case report. J Oral Maxillofac Res. 2010; 1(2):e5. doi:10.5037/jomr.2010.1205.
8. Noffke CE. Juvenile ossifying fibroma of the mandible. An 8 year radiological follow-up. Dentomaxillofac Radiol. 1998; 27(6): 363-6.
9. Papadaki ME, Troulis MJ, Kaban LB. Advances in diagnosis and management of fibro-osseous lesions. Oral Maxillofac Surg Clin North Am. 2005; 17(4): 415-34.
10. Brügger OE, Reichart PA, Werder P, Altermatt HJ, Bornstein MM. Asymptomatic ossifying fibroma of the mandible: a case presentation. Quintessence Int. 2012; 43: 381-5.
11. Kramer IR, Pindborg JJ. Histological classification of odontogenic tumours histologic typing of odontogenic tumours. 2nd edition. Berlin: Springer-Verlag: 28. 1992.
12. Rai S, Kaur M, Goel S, Prabhat M. Trabeculae type of juvenile aggressive ossifying fibroma of the maxilla: Report of two cases. Contemp Clin Dent. 2012; 3(Suppl1): S45-S50.
13. Khoury NJ, Naffaa LN, Shabb NS, Haddad MC. Juvenile ossifying fibroma: CT and MR findings. Eur Radiol. 2002; 12 (Suppl 3): S109-13.
14. Williams HK, Mangham C, Speight PM. Juvenile ossifying fibroma. An analysis of eight cases and a comparison with other fibro-osseous lesions. J Oral Pathol Med. 2000; 29(1): 13-8.
15. Espinosa SA, Villanueva J, Hampel H, Reyes D. Spontaneous regeneration after juvenile ossifying fibroma resection: a case report. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2006; 102(5): e32-5.
16. Dominguete PR, Meyer TN, Alves FA, Bittencourt WS. Juvenile ossifying fibroma of the jaw. Br J Oral Maxillofac Surg. 2008; 46(6): 480-1.
17. Saiz-Pardo-Pinos AJ, Olmedo-Gaya MV, Prados-Sánchez E, Vallecillo-Capilla M. Juvenile ossifying fibroma: a case study. Med Oral Patol Oral Cir Bucal. 2004; 9(5): 456-8.
18. Slootweg PJ, Müller H. Juvenile ossifying fibroma. Report of four cases. J Craniomaxillofac Surg. 1990; 18(3): 125-9.
19. Waldron CA. Fibro-osseous lesions of the jaws. J Oral Maxillofac Surg. 1993; 51(8): 828-35.
20. Shekhar MG, Bokhari K. Juvenile aggressive ossifying fibroma of the maxilla. J Indian Soc Pedod Prev Dent. 2009; 27(3): 170-4.
21. MacDonald-Jankowski DS. Fibro-osseous lesions of the face and jaws. Clin Radiol. 2004; 59(1):11-25
22. Thankappan S, Nair S, Thomas V, Sharafudeen KP. Psammomatoid and trabecular variants of juvenile ossifying fibroma-two case reports. Indian J Radiol Imaging. 2009; 19:116-9.
23. Breheret R, Jeufroy C, Cassagnau E, Malard O. Juvenile ossifying fibroma of the maxilla. Eur Ann Otorhinolaryngol Head Neck Dis. 2011; 128(6): 317-20