Review Article

Psammomatoid and Trabecular Juvenile Ossifying Fibromas of the Jaws: Two Aggressive Tumors

ABSTRACT

Juvenile ossifying fibromas (JOFs) of the jaws are uncommon benign fibro-osseous tumors affecting children under 15 years of age. Based on histological criteria, JOFs have been classified into psammomatoid and trabecular. Their aggressiveness, added to their high tendency to recur, provokes real diagnostic and therapeutic challenges for the dental practitioner and makes postoperative follow-up over the years indispensable. The aim of this article was to review the clinical, histological, and radiological features of these lesions as well as their treatment modalities.

Keywords: Juvenile, ossifying fibroma, trabecular, psammomatoid.

1. INTRODUCTION

Juvenile ossifying fibromas (JOFs), also known as juvenile active ossifying fibromas and juvenile aggressive ossifying fibromas, are rare benign fibro-osseous tumors [1,2]. They present as neoplastic lesions affecting the facial bones, with possible intracranial and orbital extensions [3-5]. Typically, JOFs occur under the age of 15, with no gender predilection [3,4].

Histologically, and like in all the ossifying fibromas, the normal bone is replaced by a fibrous cellular stroma including mineralized bone trabeculae and cementum-like material [6,7].

Due to their high recurrence rate, early diagnosis, appropriate treatment, and long-term follow-up are indispensable [3].

The main objective of this article was to review the clinical, histological, and radiological features of JOFs as well as their treatment modalities.

2. CLINICAL, RADIOLOGICAL, AND HISTOLOGICAL FEATURES OF JOF

JOFs are variants of ossifying fibroma affecting young patients. According to a study conducted by Slootweg et al. [8], the average age at the time of the lesion diagnosis was 11.8 years. The majority of the cases occur in the sinonasal area and the jaws more frequently in the maxilla than in the mandible [9,10].

Due to their aggressive nature, JOFs grow asymptomatically to very large sizes, leading to severe facial asymmetry and giving suspicion of malignancy [10] (Figure 1).

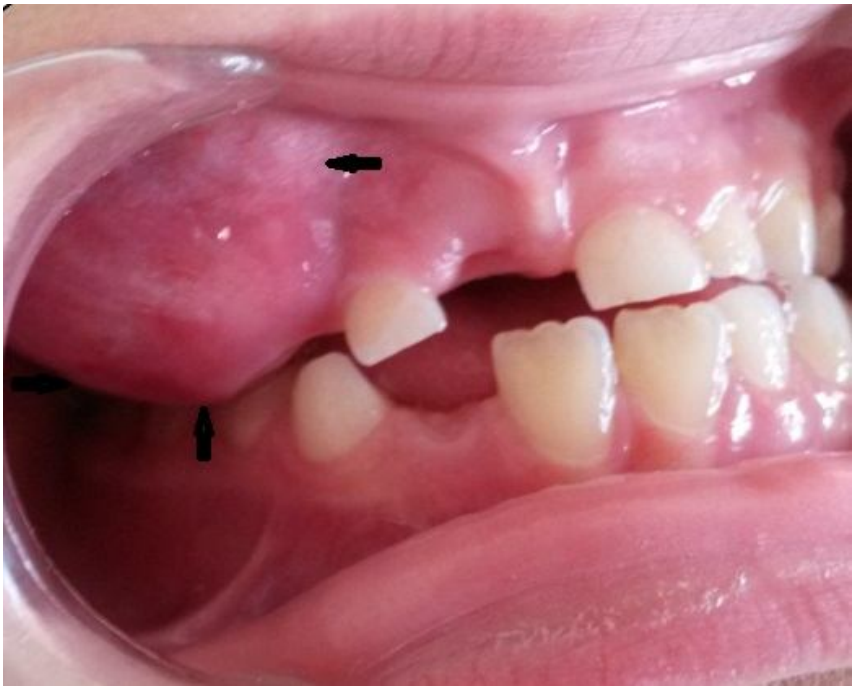

Fig. 1. Intraoral photograph showing a well-defined, large tumefaction of hard consistency extending to the vestibule of the right maxilla. The color and texture of the overlying mucosa are normal.

In many cases, JOFs reach the maxillary sinus, the nasal cavity, and push the globe superiorly, causing symptoms related to mass effects, including sinus dysfunction and visual changes [7]. Extension to the cranial base is also reported [10,11]. JOFs can provoke cortical thinning and

perforation as well as tooth displacement and root resorption [3,6]. Paresthesia is not commonly seen [3].

Radiologically, JOFs can be seen as well demarcated uni-or multilocular lesions; the amount of calcified tissue produced leads to a variable degree of radiolucency and radiopacity [6,12]. Cone-beam computed tomography (3D radiography) assessment may show well-defined sclerotic borders with an inconsistent number of calcifications [13] (Figure 2).

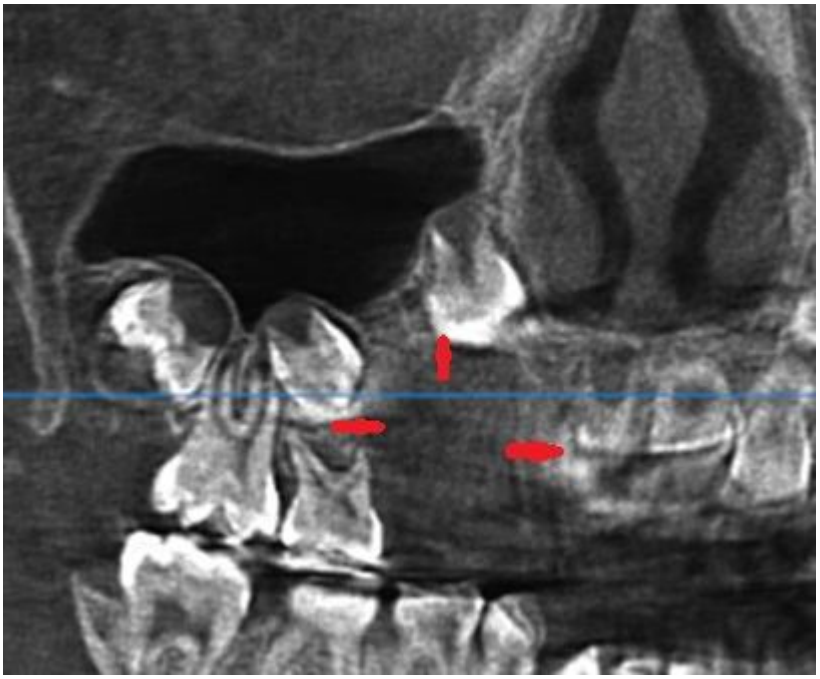

Fig. 2. A CBCT image showing a large, well-defined mixed lesion extending from tooth #53 (*anteriorly)* to tooth #55 (posteriorly) and till the germ of the permanent canine (superiorly).

Histologically, JOFs show heterogeneous morphology. Cell-rich fibrous tissue with giant cells and bands of cellular osteoid trabeculae is often present [10,14] (Figure 3).

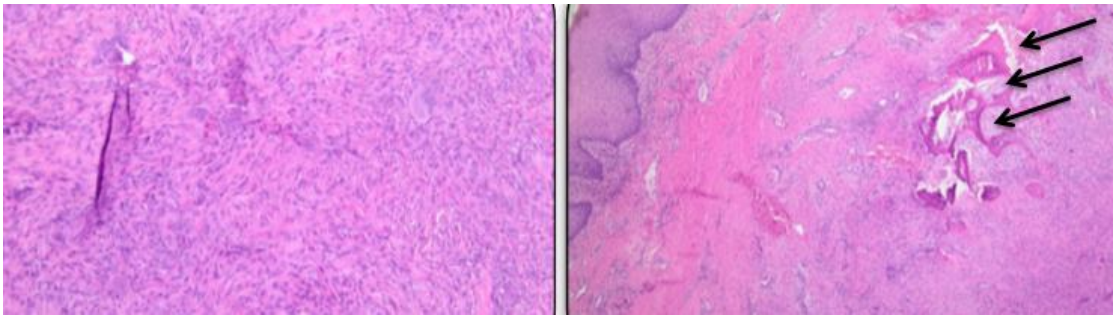

Fig. 3. Histological cuts showing mixed spindle and giant cells and trabeculae of immature bone

Based on histological criteria, JOFs have been classified into two types: the psammomatoid, distinguished by small uniform spherical ossicles that resemble psammoma bodies, sometimes grouped together to form large areas of mineralization; and the trabecular, distinguished by trabecular osteoid and woven bone with collections of osteoclastic giant cells [6,7,14].

Psammomatoid JOFs are reported more frequently than trabecular JOFs. They commonly involve the orbit and the sinuses, with the ethmoid sinus being the most prevalent. The affected patients' ages range from 3 months to 72 years (mean age, 16-33 years). As for trabecular JOFs, they mainly affect the jaws in patients aged between 2 and 12 years (mean age, 8-12 years) [6,7].

3. DIFFERENTIAL DIAGNOSIS

Many bone lesions, usually found in facial bones and jaws, may constitute a differential diagnosis challenge for JOFs. Fibrous dysplasia remains the most important one [1,2,15]; 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 [16].

Cemento-ossifying fibroma, a histological variant of JOF, can as well be considered. Nevertheless, giant cells, which are obviously found in JOFs, are not present in cement-ossifying fibromas [17].

Additionally, when diagnosing JOFs, cementoblastoma, osteoblastoma, osteosarcoma, and other lesions must be ruled out [1,15]; this can be done through careful clinical, radiological, and histological assessments.

4. TREATMENT OF JOF

The treatment modalities of JOF remain controversial. Aggressive surgical resection and conservative surgery are among the suggested treatment techniques [18-20].

According to Abuzinad and Alyamani [2], the less aggressive approach should be considered first for JOF treatment in children. Likewise, Slootweg and Müller [21] recommended conservative surgery since they found no differences in the results between the limited surgical excision and the major approach.

On the other hand, many authors reported a high recurrence rate after conservative or mini-invasive treatment (in 30-56% of cases) [18,22,23], and therefore, a complete surgical resection remains the favorite choice of treatment [1,3,15,24].

It is to be noted that whatever the surgical technique was, a long-term postoperative follow-up is mandatory [1,2,4,15,25].

5. CONCLUSION

JOFs of the jaws are uncommon tumors with a high risk of recurrence. Thorough evaluations of the clinical, radiological, and histological features of these lesions are needed to surmount the diagnostic and therapeutic challenges. Furthermore, long-term follow-up of the patient after complete surgical excision is indispensable.

DISCLAIMER

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COMPETING INTERESTS

Authors have declared that no competing interests exist.

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):288-290.
2. Abuzinada S, Alyamani A. Management of juvenile ossifying fibroma in the maxilla and mandible. J Maxillofac Oral Surg. 2010; 9(1):91-95.
3. Sun G, Chen X, Tang E, Li Z, Li J. Juvenile ossifying fibroma of the maxilla. Int J Oral Maxillofac Surg. 2007; 36(1):82-85.
4. 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.
5. Khademi B, Niknejad N, Mahmoudi J. An aggressive psammomatoid ossifying fibroma of the sinonasal tract: report of a case. Ear Nose Throat J. 2007; 86(7):400-401.
6. 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.
7. Nelson BL, Phillips BJ. Benign fibro-osseous lesions of the head and neck. Head Neck Pathol. 2019; 13(3):466-475.
8. Slootweg PJ, Panders AK, Koopmans R, Nikkels PG. Juvenile ossifying fibroma. An analysis of 33 cases with emphasis on histopathological aspects. J Oral Pathol Med. 1994; 23(9):385-388.
9. Noffke CE. Juvenile ossifying fibroma of the mandible. An 8 year radiological follow-up. Dentomaxillofac Radiol. 1998; 27(6):363-366.
10. Khanna J, Ramaswami R. Juvenile ossifying fibroma in the mandible. Ann Maxillofac Surg. 2018; 8(1):147-150.
11. Barrena López C, Bollar Zabala A, Úrculo Bareño E. Cranial juvenile psammomatoid ossifying fibroma: case report. J Neurosurg Pediatr. 2016; 17(3):318-323.
12. Papadaki ME, Troulis MJ, Kaban LB. Advances in diagnosis and management of fibro-osseous lesions. Oral Maxillofac Surg Clin North Am. 2005; 17(4):415-434.

13. Brügger OE, Reichart PA, Werder P, Altermatt HJ, Bornstein MM. Asymptomatic ossifying fibroma of the mandible: a case presentation. Quintessence Int. 2012; 43(5):381-385.

14. Aslan F, Yazici H, Altun E. Psammomatoid variant of juvenile ossifying fibroma. Indian J Pathol Microbiol. 2018; 61(3):443-445.

15. 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(Suppl 1):S45-S50.

16. Khoury NJ, Naffaa LN, Shabb NS, Haddad MC. Juvenile ossifying fibroma: CT and MR findings. Eur Radiol. 2002; 12 Suppl 3:S109-S113.

17. 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):13-18.

18. 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-e35.

19. Titinchi F. Juvenile ossifying fibroma of the maxillofacial region: analysis of clinico-pathological features and management. Med Oral Patol Oral Cir Bucal. 2021; 26(5):e590-e597.

20. Paranthaman A, Shenoy V, Kumar S, Marimuthu L, Velusubbiah S, Vijayaraj S. Trabecular variant juvenile ossifying fibroma of the maxilla. Cureus. 2017; 9(9):e1684.

21. Slootweg PJ, Müller H. Juvenile ossifying fibroma. Report of four cases. J Craniomaxillofac Surg. 1990; 18(3):125-129.

22. Liu Y, Shan XF, Guo XS, Xie S, Cai ZG. Clinicopathological characteristics and prognosis of ossifying fibroma in the jaws of children: a retrospective study. J Cancer. 2017; 8(17):3592-3597.

23. Han J, Hu L, Zhang C, Yang X, Tian Z, Wang Y, et al. Juvenile ossifying fibroma of the jaw: a retrospective study of 15 cases. Int J Oral Maxillofac Surg. 2016; 45(3):368-376.

24. Chrcanovic BR, Gomez RS. Juvenile ossifying fibroma of the jaws and paranasal sinuses: a systematic review of the cases reported in the literature. Int J Oral Maxillofac Surg. 2020; 49(1):28-37.

25. Singh AK, Kumar N, Singh S, Pandey A, Verma V. Juvenile ossifying fibroma of the mandible: A case report and review. J Dent Allied Sci. 2018; 7:34-37.