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

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Authors' contributions

This work was carried out in collaboration between both authors. Both authors read and approved the final manuscript.

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Review Article

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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].

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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] (Fig. 1).

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] (Fig. 2).

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

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].

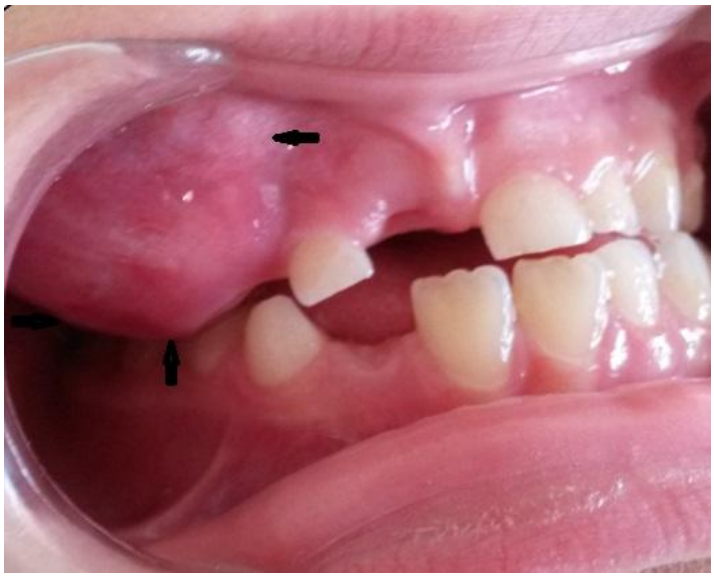

Fig. 1. Intraoral photograph showing a well-defined, large tumefaction of hard consistency extending to the vestibule of the right maxilla. The color and texture of the overlying mucosa are normal

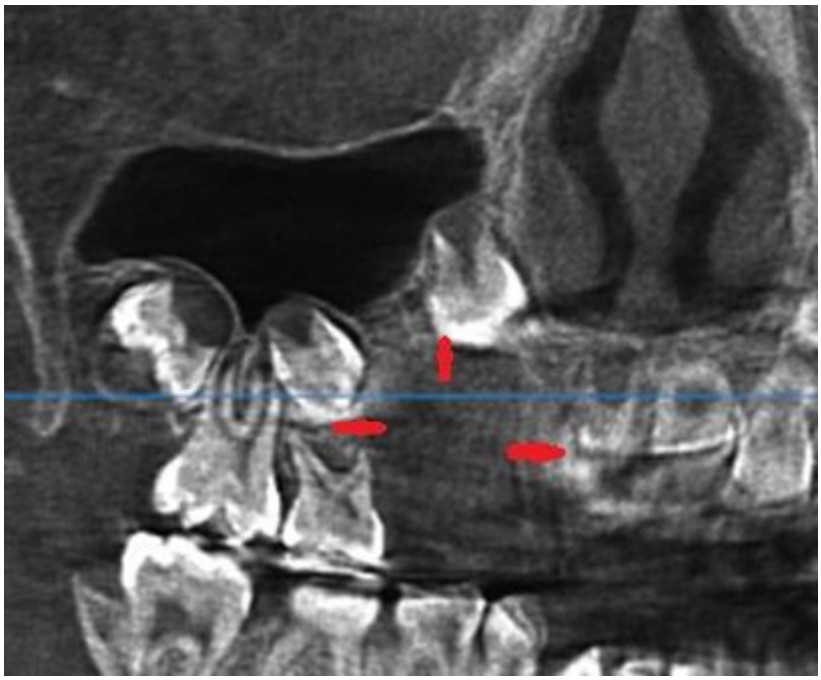

Fig. 2. A CBCT image showing a large, well-defined mixed lesion extending from tooth #53 (*anteriorly)* to tooth #55 (posteriorly) and till the germ of the permanent canine (superiorly)

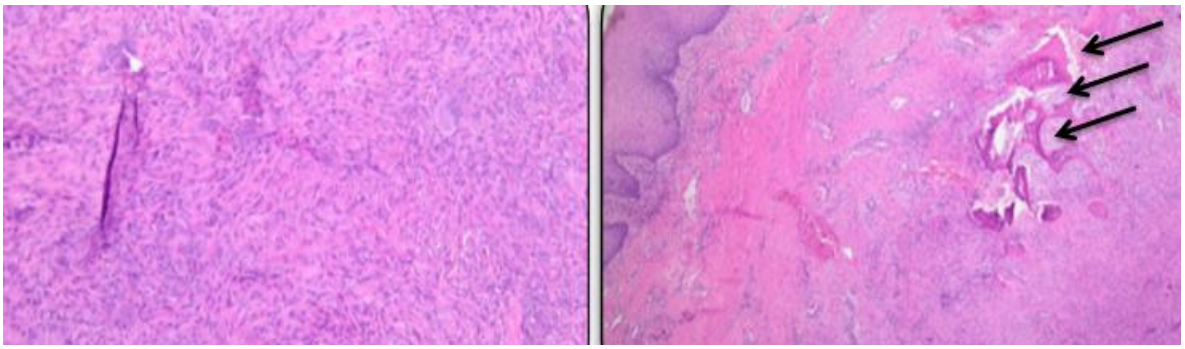

Fig. 3. Histological cuts showing mixed spindle and giant cells and trabeculae of immature bone

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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CONSENT

It is not applicable.

ETHICAL APPROVAL

It is not applicable.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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