

Trabecular juvenile ossifying fibroma in the mandible: clinical, radiographic and histopathologic features

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Abstract:

Juvenile ossifying fibroma is an uncommon, aggressive, asymptomatic fibro-osseous lesion. An 11-year-old white boy was seen at the stomatology outpatient clinic with a complaint of pain and a one-year history of tooth mobility and extensive swelling in the mandible. Extraoral clinical examination showed major facial asymmetry and swelling in the left mandibular body. Intraoral examination revealed expansion of the buccal and lingual cortical bones. Imaging identified a multilocular mixed lesion in the mandible with hyperdense areas. An incisional biopsy was performed and the combination of clinical, imaging and histopathologic data led to the final diagnosis of juvenile ossifying fibroma. Treatment consisted of segmental mandibulectomy and reconstruction with a microsurgical fibular flap, after 5 years of follow up, no local recurrence was observed. Strategies for the clinical management and treatment of pediatric patient should be designed to offer the best prognosis and quality of life for the patient.

Keywords: Jaw Abnormalities; Jaw Diseases; Oral Medicine.

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INTRODUCTION

Juvenile ossifying fibroma (JOF) is an uncommon fibro-osseous lesion. Although benign and generally asymptomatic, this lesion exhibits an aggressive behavior and strong tendency towards recurrence. JOF preferentially involves the craniofacial skeleton of young patients¹ and a slight predilection for males has been reported².

According to the World Health Organization (WHO), the two variants, psammomatoid (PsJOF) and trabecular JOF (TrJOF), differ in terms of clinical and histopathologic presentation³. Clinically, TrJOF is a lesion of the gnathic bones with a preference for the maxilla, while PsJOF mainly involves the paranasal sinuses¹. Histopathologic findings include a cellularized connective tissue. Psammomatoid JOF is characterized by the formation of small and uniform spherical ossicles that resemble psammoma bodies, while TrJOF differs by the formation of fibrillar osteoid and immature bone trabeculae².

Early treatment of this benign lesion by complete excision is indicated¹. However, controversy exists regarding its management², and different treatment modalities that range from conservative treatment by curettage to more invasive approaches such as en bloc or segmental resection have been reported¹.

This study reports the approach to a case of locally aggressive, rapidly growing TrJOF in an 11-year-old boy.

CASE REPORT

An 11-year-old white boy was seen at the stomatology outpatient clinic with a complaint of pain and a one-year history of tooth mobility and extensive swelling in the mandible. Extraoral clinical examination showed major facial asymmetry and a swelling in the left mandibular body. Intraoral examination revealed expansion of the buccal and lingual cortical bones in the region of the lower molars, which exhibited grade 3 mobility (Figure 1). Imaging (Figure 1) identified a multilocular mixed lesion in the mandible (region of the left molars) with hyperdense areas, which measured 7.2 cm in greatest diameter.

An incisional biopsy was obtained and histopathologic analysis (Figure 2) revealed non-encapsulated fragments of richly vascularized loose connective tissue amidst immature bone trabeculae that assumed different shapes, sometimes curvilinear,

and consisted of osteoid with varying degrees of mineralization. The combination of clinical, imaging and histopathologic data led to the final diagnosis of TrJOF.

Based on the age of the patient and the clinical, imaging and histopathologic findings, treatment consisted of segmental mandibulectomy and microsurgical mandibular reconstruction with a right fibular flap. The patient presented good recovery during the postoperative period. During follow-up, the patient reported slight sensory and motor loss in the right lower limb, including the loss of extension and flexion strength of the toes.

Postoperative extraoral examination showed improvement of the mandibular contour. Intraoral clinical examination revealed the accumulation of a dental biofilm and poor hygiene, but the site of microsurgical reconstruction had a normal appearance without any alterations in soft tissues and position of the fibular flap. No recurrence of the lesion was observed after more than 5 years of follow-up (Figure 3).

DISCUSSION

Juvenile ossifying fibroma is an uncommon benign, but potentially aggressive, fibrous-osseous lesion⁴, that accounts for 2% of all benign oral neoplasms in children⁵. Although usually asymptomatic, Phattarataratip et al.⁴ observed that 38.5% of their patients complained of swelling associated with pain. In the present case, the patient reported pain associated with extensive swelling.

The WHO classifies JOF into two variants³, with PsJOF being more prevalent than TrJOF². Phattarataratip et al.⁴ described 13 cases of JOF, 9 of the psammomatoid type and 4 of the trabecular type. Other authors also reported a higher prevalence of the psammomatoid type^{1,6}. The psammomatoid type affects patients over a broad age range (mean of 16 to 33 years) compared to the trabecular type (8.5 to 12 years). This is compatible with the present case which was diagnosed with TrJOF at 11 years of age.

Trabecular JOF predominantly involves the maxillary gnathic bones¹ and can cause root resorption and tooth displacement in the affected area. In the mandible, a higher prevalence is found in areas such as the ramus and mandibular angle², similar to the present case in which, in addition to these sites, the lesion extended to the region of the mandibular body. No tooth displacement or root resorption was observed, but the teeth exhibited intense mobility.

The trabecular variant of JOF is formed by richly cellular, fibrous tissue, exhibiting trabeculae of cellular

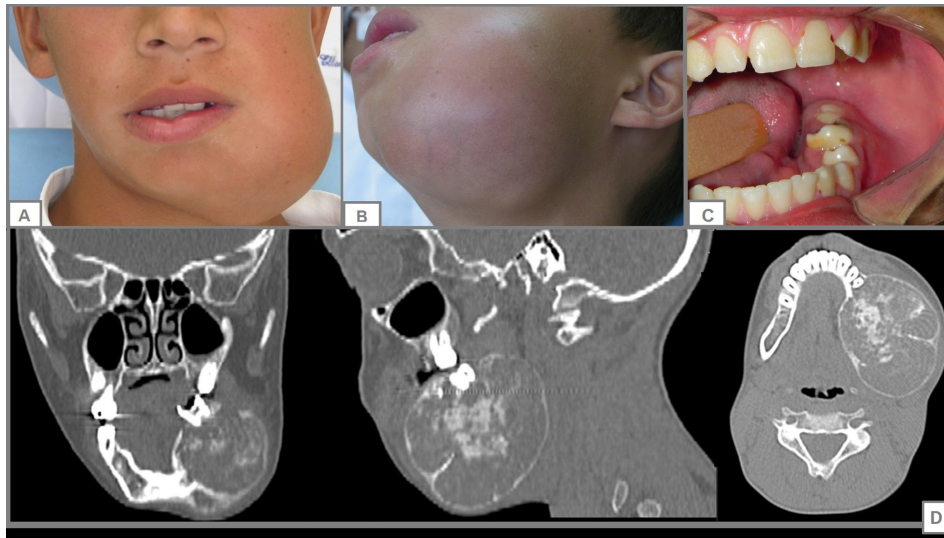


Figure 1. Extraoral clinical view showing a swelling in the region of the left mandibular body and tomographic sections of the lesion.

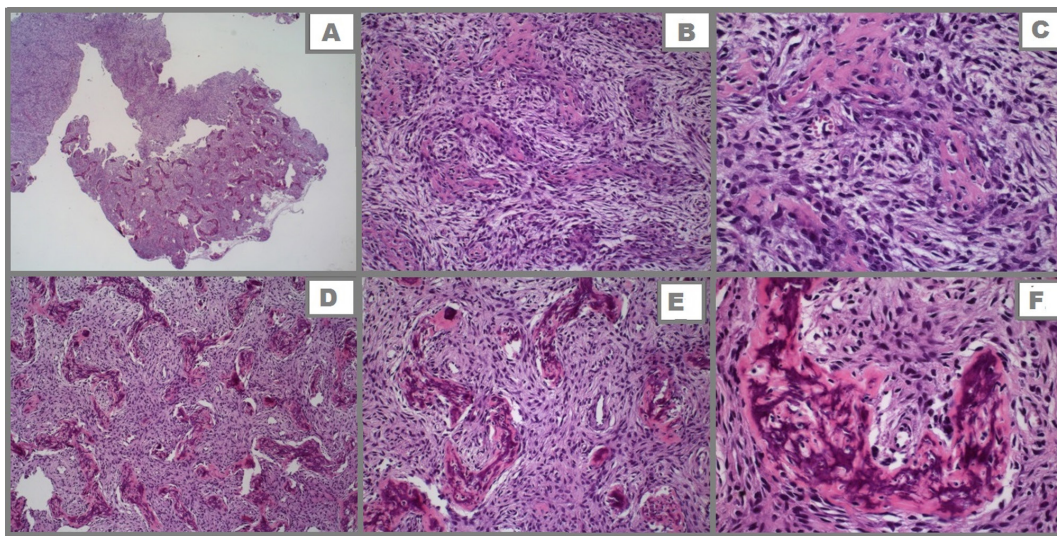


Figure 2. Histopathologic findings (HE staining). A: Panoramic view of the lesion, 25x. B, C, D, E and F: Immature bone trabeculae exhibiting different degrees of mineralization amidst richly cellularized connective tissue (D: 100x, B and E: 200x, C and F: 400x).

osteoid and delicate trabeculae of immature bone. These trabeculae sometimes anastomose, forming a network³. These findings were observed in the present case. Additionally, but less typical, multinucleated giant cells, pseudocystic stroma, degenerations and hemorrhage may be present³.

Phattarataratip et al.⁴ highlighted the importance of surgical removal to treat this type of benign lesion. However, different treatment modalities have been proposed depending on the size and invasion of adjacent tissue. Patil et al.⁷ and Banu & Palikat² reported that small neoplasms can be treated successfully by enucleation and curettage, while resection should be considered in

cases of recurrence of the lesion or invasion of adjacent tissues and cavities. The authors also suggested that, in the absence of paresthesia and involvement of the lower mandibular border, a more conservative treatment can be planned.

Rao et al.⁶ emphasized the aggressiveness of JOF, with recurrence rates ranging from 30% to 58%, and suggested complete surgical excision, en bloc or hemisection, as the best treatment option to prevent recurrence. Bohn et al.⁸ observed high recurrence of JOF after partial resection due to the infiltrative nature of the tumor front. However, the authors emphasize that enucleation of the lesion is sometimes not possible



Figure 3. Extraoral aspect obtained during follow-up 10 months after surgery.

because of the area affected, compromising the prognosis of the patient.

The mandible defines the profile and appearance of the lower third of the face and is essential for adequate occlusion, mastication, swallowing and speech⁹. Consequently, reconstructive treatment should be studied in an attempt to restore esthetics and function¹⁰. In view of the high prevalence of JOF in young patients, we highlight the importance of reconstruction since these patients could develop psychological disorders related to bullying because of their appearance. Studies show that children and adolescents with malignant tumors are more introverted than their peers¹¹⁻¹³. In addition, the appearance-affecting sequelae of treatment such as weight gain, scars, amputation and disfigurements can expose these children to bullying¹³.

Within this context, Wan et al.¹⁴, evaluating the quality of life of patients undergoing mandibular reconstruction, described mastication and appearance as the domains frequently chosen by the patients as the most important. In addition, these patients reported improvement in their quality of life after mandibular reconstruction. Fang et al.¹⁵ indicated possible improvement in mastication, deglutition and sense of taste in this group of patients. In agreement, most patients of the study of Zavalishina et al.¹⁶ reported satisfaction with their overall quality of life one year after mandibular resection followed by reconstruction with a fibular flap.

However, mandibular reconstruction continues to be a challenge due to two main factors: the anatomic diversity at this site and the complex movements that the mandible performs¹⁰. Thus, the standard treatment for reconstruction of mandibular defects continues to be a free vascularized bone flap¹⁷ and microvascular surgery is widely used as an auxiliary treatment of head and neck defects¹⁶.

Spinelli et al.¹⁷ consider the fibular flap to be the first choice due to easy access to the donor site, permitting a surgical approach of two teams at the same time, great length and adequate bone width and height for placement of the dental implant. Disa & Cordeiro⁹ suggested the fibula to be a versatile donor site for mandibular reconstruction, which was indicated in more than 90% of the cases of their study. According to Rashid et al.¹⁸, the vascularized fibular flap is an excellent option for mandibular reconstruction in pediatric benign neoplasms, which require large bone resection.

However, donor site morbidity is uncertain despite the high overall success rate of free flap surgery of 95% to 97%¹⁶. The present patient exhibited loss of extension and flexion strength of the toes at the donor site. Ferreira et al.¹⁰ highlighted the possibility that about 24% of the patients submitted to this type of reconstruction may have some difficulty walking, with a reduction in the range of motion of the feet and in extension and flexion strength, as well as sensitivity in the donor area. On the other hand, Fang et al.¹⁵ observed no donor site morbidity in the 28 patients of their study.

With respect to the reconstructed area, it was observed success in the present case in the follow-up period. Results similar to those found by Rashid et al.¹⁸.

CONCLUSION

The treatment of uncommon, benign, aggressive lesions, such as TrJOF, should promote adequate functional and esthetic conditions for the pediatric patient, in addition to minimizing negative effects on facial development.

Strategies for the clinical management and treatment of this condition should be designed to offer the best prognosis and quality of life for the patient.

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