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CASE REPORT

Trabecular juvenile ossifying fibroma in the maxilla: from diagnosis to rehabilitation

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Abstract:

Juvenile ossifying fibroma is a benign fibro-osseous lesion with an unusual presentation that predominantly affects individuals in the first decade of life. The aggressiveness added to the high rates of recurrence causes real diagnostic and therapeutic challenges for the dental surgeon and makes post-operative follow-up over the years indispensable. We present the case of a 21-year-old girl with a rapid onset and abrupt increase in volume in the left maxilla. After clinical, radiographic and histopathological exams, the diagnosis of trabecular juvenile ossifying fibroma was obtained. The lesion was surgically removed and the patient was rehabilitated with a removable partial prosthesis, due to the involvement of some teeth during surgical access. The present clinical case demonstrates that the adequate treatment must consist of complete surgical excision, early functional and aesthetic prosthetic rehabilitation and long-term preservation.

Keywords: Ossifying fibroma; Oral pathology; Margins of excision; Treatment outcome.

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INTRODUCTION

Juvenile ossifying fibroma (JOF) is an uncommon benign fibro-osseous lesion, classified by the World Health Organization - WHO (2017) as a synonym of conventional ossifying fibroma because it affects predominantly young individuals between the ages of 8 and 12 years old, and may also affect patients in the third life decade[1]. A study carried out with 80 patients affected by FOJ, in which 69.6% were women and 30.4% men and, in relation to ethnicity, 83.93% were black, 12.50% Caucasian and 3.57% Asian, demonstrated a greater predilection for FOJ cases in females and blacks[2].

JOF has two histological subtypes, trabecular (TJOF) and psammomatoid (PJOF), which present similar clinical behavior with an increase in painless volume and expansive growth, affecting mainly the gnathic bones and paranasal sinuses[3,4]. A retrospective study conducted with 15 JOF cases (10 of which were trabecular and 5 psammomatoid), showed that 60% of the cases presented as an clinical manifestation the asymptomatic volume increase in mandible accompanied by facial asymmetry and 40% of the cases, 4 in the maxilla and 2 in the mandible, reported pain and occasional clinical findings such as diplopia, nasal congestion and rapid progression[5].

With regard to the trabecular histological subtype, it affects younger patients (8-12 years) without a predilection for sex[1]. A systematic review study6, in which the distribution frequency of 403 JOF cases was analyzed according to the histological subtype and age, revealed that about 50% of the TJOF cases manifested between 6 and 10 years, 45% among 11 and 15 years and 5% between 21 and 25 years. The literature also points out that the main site of TJOF involvement is the maxilla, with a predilection for the anterior region. The TJOF common radiographic aspect is unilocular radiolucency bounded by a thin radiopaque line, in addition to the presence of irregular and dispersed calcifications[7,8,9].

Histologically, TJOF is described as a non-encapsulated lesion with hypercellular connective tissue composed of varying morphology fibroblasts (either ovoid or fusiform), with little collagen production and irregular proliferation of highly cellularized osteoid10. Immature bone trabeculae without an osteoblastic margin are also present[11]. In addition, areas of connective tissue with varying cellularity are separated from each other by thin chains of hemorrhagic foci with a cluster of multinucleated giant cells of the osteoclastic type and pseudocystic degeneration areas[10].

JOF treatment is surgical and the modalities consist of total or partial resection of the affected bone, enucleation, curettage or association of both[1,4,10]. Resective surgery is indicated for extensive tumors, locally aggressive and with a potential for recurrence, while enucleation or curettage are indicated for small tumors and usually diagnosed at an early stage[6,8,12]. In the study that addressed the therapeutic management of 10 juvenile ossifying fibroma cases (6 in the mandible and 4 in the maxilla) treated by total resection, there was no recurrence during the 3-year follow-up period[12].

Still, another study6 compared the surgical treatment modalities for 72 TJOF cases, comprised of: curettage; enucleation; enucleation, curettage and peripheral osteotomy; and resection. The results showed that enucleation and curettage had a considerably high recurrence rate (63.5% and 45.5%, respectively), regardless of the lesion anatomical location. Enucleation followed by curettage and peripheral osteotomy showed lower recurrence rates than enucleation (33.3%). However, when the resection was performed, only one TJOF case presented recurrence[6].

It is worth noting that post-surgical sequelae, mainly in the maxilla region, result in extensive bone defects that predispose the appearance of facial and functional deformities and compromise the individual's life quality, such as oroantral communication and phonetic, masticatory and swallowing difficulties[13]. In order to minimize the aesthetic, psychological and social impacts resulting from mutilating surgeries, the literature[14,15] proposes alternatives for rehabilitation ranging from bone grafts to prosthetic devices. A study compared the influence of prosthetic rehabilitation and reconstructive surgery in the patients' life quality with a maxillary bone defect, finding a statistically significant improvement in the patients' life quality undergoing these rehabilitation modalities[16].

Thus, the objective of the present study is to report a trabecular juvenile ossifying fibroma clinical case in the maxilla, addressing clinicopathological findings, treatment and prosthetic rehabilitation.

CASE REPORT

The female patient, 21 years old, melanoderma, sought a private office for maxillofacial surgery, reporting a rapid and sudden increase in volume in the maxilla 2 months ago (Figure 1A). On intraoral clinical examination, she had an increase in volume in the bottom of the maxillary vestibule and in the anterior region of

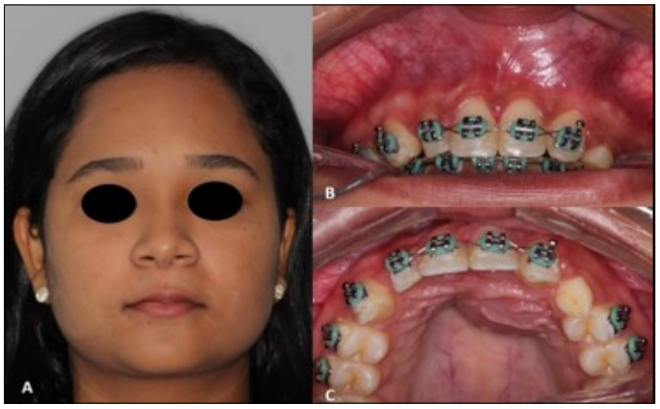

Figure 1. Extraoral and intraoral clinical aspects. A: swelling in the left periorbicular/ paranasal region. B: swelling increase in the bottom of the maxillary vestibule. C: swelling in the anterior region of the hard palate.

the hard palate, both on the left side and measuring 5 cm in its largest diameter (Figure 1B-1C). The tomographic examination revealed the presence of a hypodense image, with defined limits and a considerable proportion, located in the left maxilla in the region between teeth 21 to 24, and small, slightly hyperdense images inside, suggestive of calcification. In addition, there was an expansion of the bony corticals (vestibular and palatal) and expansion to the upper of the lower wall of the left nasal cavity (Figure 2A-2B-2C-2D).

Considering the clinical and tomographic findings, the established diagnostic hypotheses included ameloblastoma, odontogenic keratocyst, fibrous dysplasia and juvenile ossifying fibroma. In view of this, an incisional biopsy was performed through intraoral access with subsequent referral of the removed specimen to the pathological anatomy laboratory (Figure 3A).

Microscopic examination of these specimens, through 5 micrometers thick slices and stained using the hematoxylin and eosin technique, reveal a connective tissue presence consisting of elongated bundles of collagen fibers, of variable density, interposed by sometimes ovoid-shaped fibroblasts, sometimes fusiform, arranged in storiform arrangement. The parenchymal component was associated with deposition areas of bone trabeculae at different degrees of maturation. The focal clusters presence of multinucleated giant cells was also evidenced (Figure 3B).

Considering the removal of the soft and hard tissue components from the lesion, the histopathological diagnosis established was of trabecular juvenile ossifying fibroma. After diagnosis, the patient underwent complete excision of the lesion through enucleation, curettage and peripheral osteotomy. Due to the extension and location of the tumor, we opted for intraoral surgical access, extraction of teeth 21, 22, 63 and 24 (Figure 4D). The surgical wound was sutured with resorbable threads and analgesic and anti-inflammatory medications as well as mouthwashes with 0.12% chlorhexidine were prescribed. There was no trans-surgical complication and the specimen was sent again to the pathological anatomy laboratory, confirming the initial histopathological report.

Due to the lack of teeth and to the bone defect resulting from the surgical procedure, an immediate

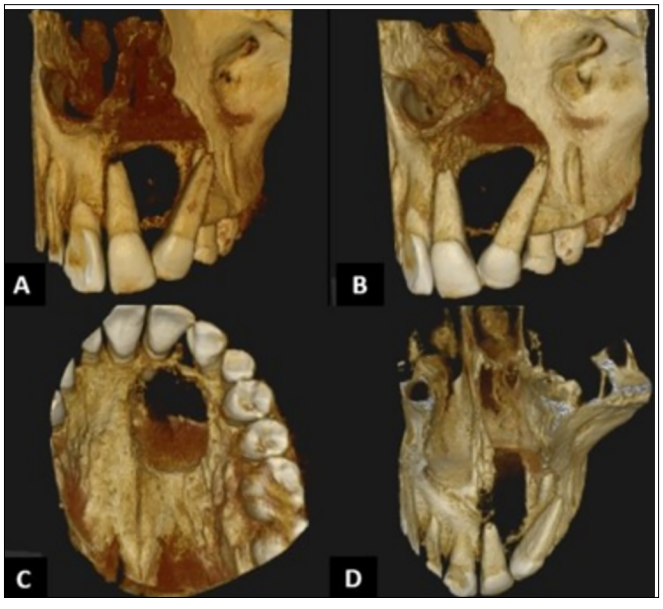

Figure 2. 3D reconstruction of cone beam tomography showing the limits of the lesion and its relationship with noble structures.

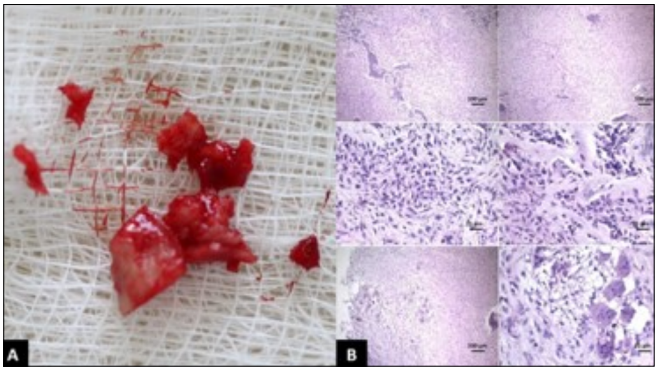

Figure 3. A: lesion fragments removed for microscopic examination. B: HE--stained histological sections revealing connective tissue fragments made up of elongated bundles of collagen fibers, of variable density, interposed by fibroblasts of either ovoid or fusiform shape, arranged in storiform arrangement. The parenchymal component is associated with deposition areas of bone trabeculae at maturation different degrees. Focal clusters of multinucleated giant cells are also evident. Such findings closed the diagnosis of trabecular juvenile ossifying fibroma.

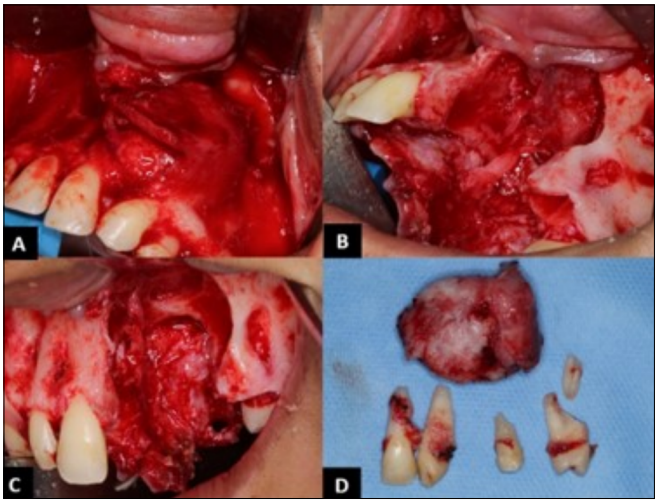

Figure 4. Surgical approach to the lesion. A: intraoral surgical access and lesion aspect after a divided flap keeping the periosteum over the lesion. B and C: surgical store after lesion removal and involved teeth. D: surgical specimen and involved teeth that were sent for confirmation of the initial diagnosis.

removable prosthesis was provisionally made in order to guarantee aesthetics and masticatory function to the patient (Figure 5A-5B-5C-5D), minimizing the psychological impact resulting from the sequelae post-surgical. After 6 months of post-surgery, a new cone beam tomography was performed (Figure 6A-6B-6C). In the absence of clinical and tomographic recurrence signs, in addition to the absence of oroantral communication signs, the patient was definitively rehabilitated with a 4-element removable partial tooth supported prosthesis. The treatment instituted returned the aesthetics and function to the pacient satisfactorily. The case has 6 months of proservation and the patient will be followed-up for another 18 months, with three follow-up visits on a six-monthly basis (Figure 7A-7B-7C-7D).

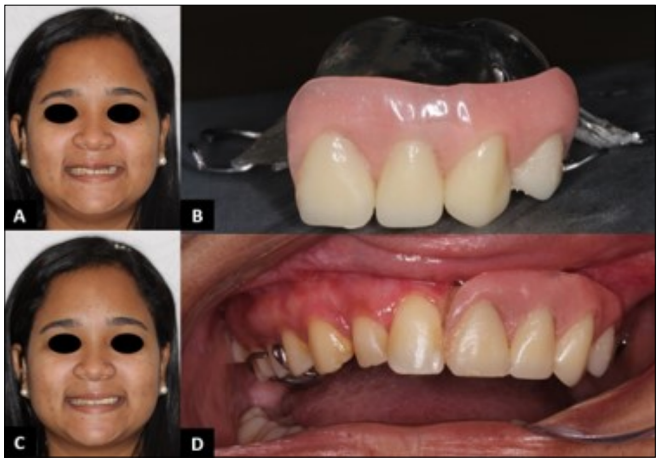

Figure 5. Provisional rehabilitation with immediate removable prosthesis. A: clinical aspect without the use of the prosthesis. B: installed prosthesis and demonstration of aesthetic gain. C: adaptation of the prosthetic device in the mouth. D: prosthesis installed.

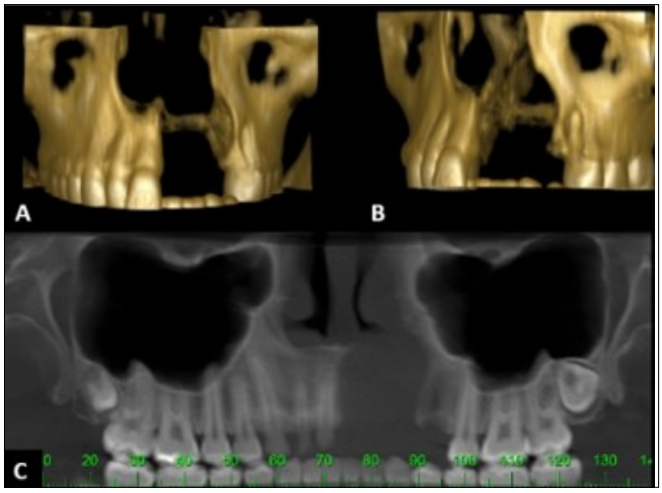

Figure 6. A and B: 3D reconstruction showing no recurrence signs. C: panoramic reconstruction showing no recurrence signs.

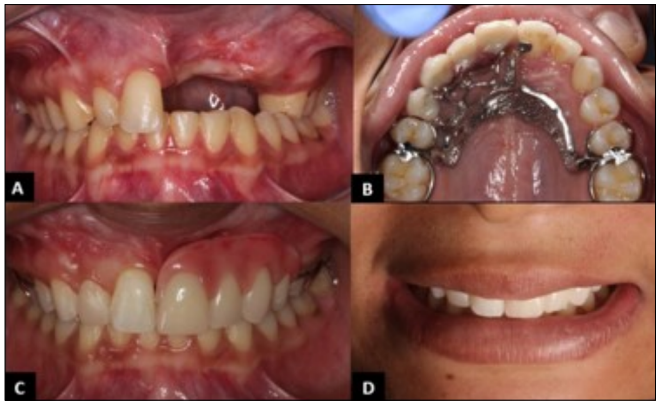

Figure 7. 6 months post-surgery. A: intact mucosa without recurrence signs and/or oroantral communication. B: occlusal view of the definitive prosthesis. C: frontal view of the definitive prosthesis. D: smile aspect after definitive prosthetic rehabilitation.

DISCUSSION

Juvenile ossifying fibroma is a rare benign fibro-osseous lesion, classified as an aggressive variant of conventional ossifying fibroma because it affects young individuals. JOF generally occurs predominantly in the first life decade without any significant predilection for sex and ethnicity[1,11,17]. In the case report, the patient is female, 21 years old and melanoderma. This finding corroborates with studies[18,19] that consider the fact that the lesion can manifest itself in individuals in the third life decade, in addition to having a slight predilection for black ethnicity[6].

Among the clinical findings, it is extremely important that the professional recognizes the clinical signs of the disease early, such as asymptomatic swelling, rapid and expansive growth and facial asymmetry[8], in addition to the rare symptoms of diplopia, proptosis and

nasal congestion[15,20,21]. In our case, the early diagnostic determination was essential for the rapid surgical approach to be instituted, otherwise, the patient could have an evolution with more severe involvement.

Among the JOF imaging findings, in most cases it is not possible to identify peculiar characteristics that distinguish such lesion from odontogenic cysts and tumors. However, the differential diagnosis between these lesions is essential to establish the best clinical and therapeutic approach[3,9,22]. JOF is usually manifested by unilocular, multilocular or mixed radiolucency associated with bone cortical expansion and occasional findings of root resorption and tooth displacement[9,22,23]. In the findings of the reported case, tooth displacement and bone expansion were markedly pronounced, which suggested the diagnostic hypotheses of ameloblastoma and odontogenic keratocyst. Therefore, the differential diagnosis with these lesions is part of the diagnostic exercise[24].

In addition, other fibro-osseous lesions associated with gnathic bones can also be a challenge in the differential diagnosis with JOF. In turn, fibrous dysplasia remains the most prominent condition[25,26]. In our case, we rule out fibrous dysplasia, as it usually exhibits marginal bone with less cell stroma and a considerable amount of lamellar bone[27].

Histologically, JOF has two variants: trabecular and psammomatoid[1,4,10]. Both are characterized by proliferation of fibrous connective tissue with varying morphology fibroblasts. While small spherical ossicles spread the collagen matrix are identified in the FOJPs, immature bone trabeculae are present in the TJOF, in addition to multinucleated giant cells similar to osteocytes and foci of pseudocystic degeneration[3,4]. According to the histopathological report of the case described, we arrived at the diagnosis of juvenile trabecular ossifying fibroma by identifying the fibroblasts presence of either ovoid, or fusiform format, arranged in a storiform arrangement, and deposition areas of trabeculae bone in different degrees of maturation and of multinucleated giant cells[10].

The JOF treatment remains controversial. Some authors[25,28,29] suggested that conservative treatments such as enucleation and bone curettage, with a less aggressive approach, should be considered as the first choice for JOF. However, many other authors[6,12,15,20] reported a high rate of recurrence after conservative or minimally invasive treatment in 30-56% of cases and, therefore, advocate total surgical resection as the preferred treatment approach. It should be noted that, whatever the surgical technique, long-term post-surgical follow-up is essential[1,10]. In our case, we presented a TJOF located in the left maxilla of a twenty-one-year-old woman. After tomographic and histological confirmations, conservative treatment was chosen based on enucleation, curettage and peripheral osteotomy, with no recurrence in the 6-month proservation period.

After the surgical procedure, the patient's well-being and psychological condition contributed to the assessment of therapeutic success, due to the great impacts resulting from surgical trauma. Thus, the ideal reconstruction of post-surgical bone defects is widely discussed in the literature. Most authors[14,15,30] advocate surgical reconstruction using bone grafts, especially in cases of extensive bone defects, and subsequently, prosthetic rehabilitation with fixed, removable or obturating devices. In relation to the described case, we rehabilitated the patient with a four element removable partial tooth supported prosthesis that demonstrated excellent results, especially due to the fact that it filled the region of the bone defect and replaced the extracted teeth in the surgical procedure, avoiding a new reconstructive surgical approach and ensuring complete patient satisfaction.

CONCLUSION

Trabecular juvenile ossifying fibroma is a rare fibro-osseous lesion with a very high recurrence risk. A careful evaluation of the clinical, radiographic and histopathological components of this lesion is necessary to elucidate the diagnosis and overcome therapeutic challenges related to it. In addition, the appropriate treatment should consist of complete surgical excision followed by long-term postoperative follow-up, using rehabilitation alternatives for post-surgical sequelae in order to guarantee the functional balance of the stomatognathic system.

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