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Case Report

Juvenile trabecular ossifying fibroma of the maxilla: Case report of a diagnostic dilemma

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ABSTRACT

Ossifying fibromas (OFs) are benign, well-demarcated lesions in the craniofacial region, particularly in the jaws, with clinical, radiographic, and histopathological similarities to other lesions, which make their diagnosis challenging. Herein, we report a case of a fibro-osseous lesion in the anterior maxilla of a 13-year-old boy, consisting of an intraosseous and an extra-osseous part, which created a diagnostic dilemma.

Key Words: Case report, fibroma, maxillary neoplasms, ossifying

INTRODUCTION

Ossifying fibromas (OFs) are benign, well-demarcated lesions in the craniofacial region, frequently in the jaws.[1] Molecular mechanism underlying the development of these lesions is still unknown, however, few studies identified chromosomal abnormalities.[2-5] They can be classified into two types namely cemento-ossifying fibroma and juvenile OF. Due to the numerous clinical and radiographic similarities to other lesions, and overlapping histopathological features, the classification of fibro-osseous lesions has been challenging. Moreover, rapid growth and osteolytic nature of these lesions can mimic malignant lesions, making the clinical decision-making more puzzling.[6-8] In this report, we describe the diagnosis and management of a rare case of a fibro-osseous lesion with confusing clinical, radiographic, and histopathological features.

CASE REPORT

A 13-year-old male patient presented to the Department of Oral and Maxillofacial Surgery of Shahid Beheshti Dental School in Tehran, with the chief complaint of a swelling on his maxillary gingiva in the left side, and ectopic eruption of maxillary left canine noticed 1 month ago according to his mother. There was no history of pain or bleeding from the site of swelling. The patient's past dental history

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was unremarkable. His past medical history was noncontributory, and there was no history of trauma.

Extra-oral examination showed facial asymmetry in the lip line and upper lip area due to the underlying swelling with no change in the overlying skin. No lymphadenopathies were detected in the head and neck region.

Intraoral examination revealed a solitary, well-circumscribed, nontender, dome-shaped growth of approximately 2 cm located in the anterior aspect of the left maxillary alveolar ridge. The surface of the lesion was smooth, and pink to red in color. The lesion was nonfluctuant, and had a firm consistency. No sign of blanching was seen with pressure [Figure 1]. Ectopic eruption of tooth #11 into the buccal vestibule was evident. Grade 2 mobility of tooth #10 was documented. No obvious local irritant was present at the time of examination, and the patient's oral hygiene was acceptable.

Panoramic examination revealed the presence of an ill-defined lesion, extending from tooth #10 to the first premolar region, presenting a fine ground-glass pattern [Figure 2]. Cone-beam computed tomographic (CBCT) images revealed an ill-defined lesion, extending from the palatal aspect of tooth #10 anteriorly, with significant buccal cortical plate expansion and destruction. The lesion had a slightly higher radiodensity than the surrounding soft tissue. Furthermore, thinning of the palatal cortical plate in the canine region was detected, but sinus and nasal floor were both intact. Superior and distal displacement of tooth #11 and a "floating in the air" appearance were also evident. External resorption and dilaceration of the mesial root of tooth #12 were also noted [Figure 3].

Based on the clinical and radiographic findings indicating a relatively invasive bony destruction with a high growth rate, differential diagnosis was central giant cell granuloma and sarcoma, respectively. Incisional biopsy under local anesthesia was planned for the patient. Needle aspiration was performed before the procedure, which was negative. The peripheral lesion was completely excised, and incisional biopsy was performed for the intraosseous part of the lesion. Histological evaluation of the peripheral specimen revealed highly cellular fibro-vascular connective tissue with active fibroblasts, woven bone, cementum-like structures, and osteoid formation. The tissue was partially

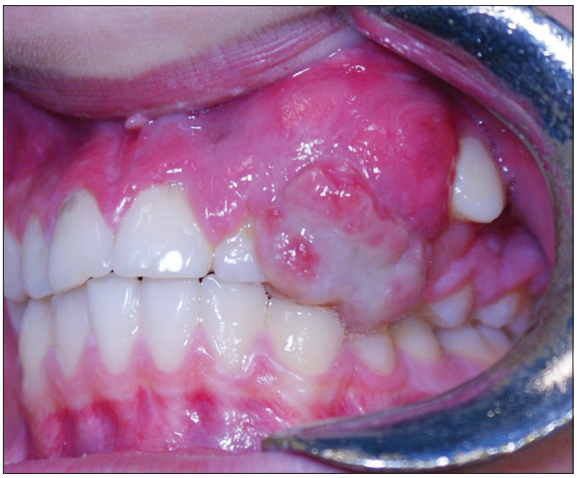

Figure 1: Intraoral view of the lesion in the left maxillary alveolar ridge.

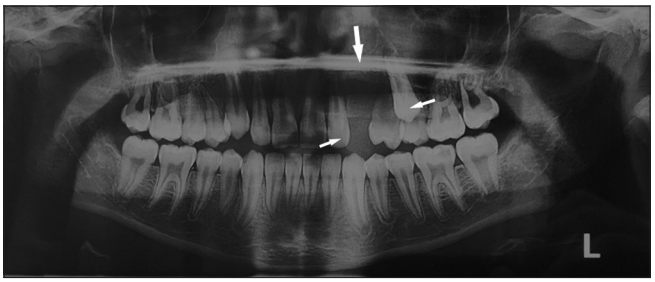

Figure 2: Panoramic radiograph revealing an ill-defined lesion extending from tooth #10 to the #12 region.

covered by ulcerated para-keratinized stratified squamous epithelium [Figure 4]. The findings were suggestive of peripheral OF (POF). The intraosseous part of the lesion showed a similar histopathological pattern.

The patient was admitted to a hospital 2 months after his first surgical procedure for enucleation and curettage under general anesthesia. The lesion was completely enucleated along with teeth #10 and #11, followed by a complete curettage of the site. Tooth #10 was completely mobile and had a hopeless prognosis at the time of surgery. The gross specimen was approximately 3 cm × 2.5 cm × 2 cm with a creamy color and firm to hard consistency [Figure 5]. The assessment of the excisional biopsy specimen showed a partially encapsulated fibro-osseous lesion composed of fragments of woven bone with osteoblastic rim, particles of cementum-like calcifications, and some osteoid formation in cellular fibrous connective tissue. The specimen was partially covered by parakeratinized stratified squamous epithelium [Figure 6]. It should be noted that histopathologic features of juvenile ossifying fibroma (JOF) and OF are similar. Based

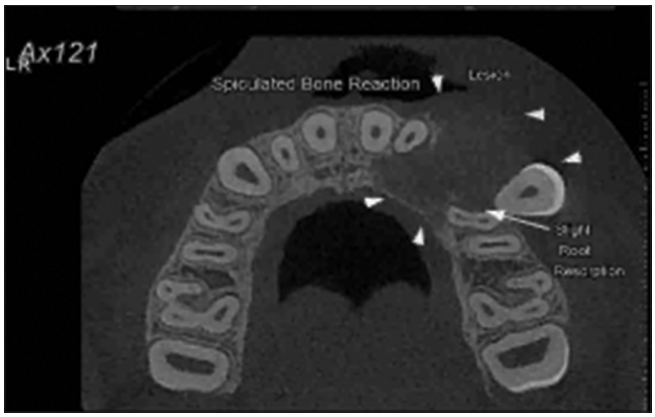

Figure 3: Cone-beam computed tomographic image of the maxillary alveolar ridge showing the lesion.

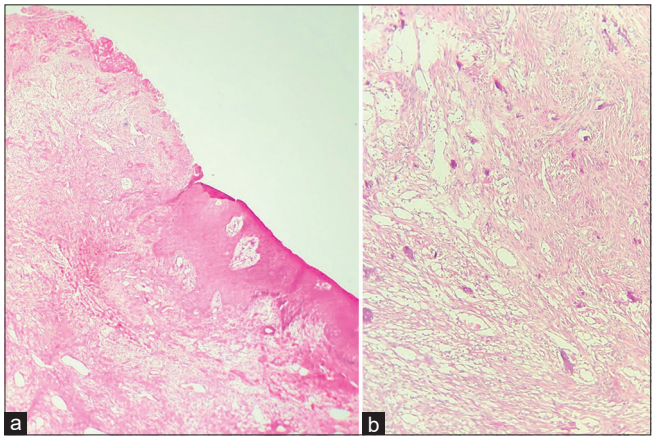

Figure 4: (a) Photomicrograph showing a gingival polypoid mass covered by ulcerated parakeratinized stratified squamous epithelium (H and E staining, ×40). (b) Photomicrograph showing numerous active fibroblasts with osteoid formation, cementum-like materials, and dystrophic calcifications (H and E staining, ×100).

on histologic features such as abundance of osteoid and woven bone formation and the zonal pattern of cementum-like ossicles and correlation of clinical and radiographic findings, the final diagnosis was JOF.

The patient has been on the regular follow-up ever since. Radiographic examination was performed after 6 months by CBCT [Figure 7]. There was no clinical or radiographic evidence of recurrence, and complete healing of the surrounding tissues was observed [Figure 8]. No sign of recurrence was evident in 14-month recall.

DISCUSSION

JOF is a rare benign lesion with high tendency to recur. It is usually characterized by early age of onset (under 15 years) and aggressive behavior. It

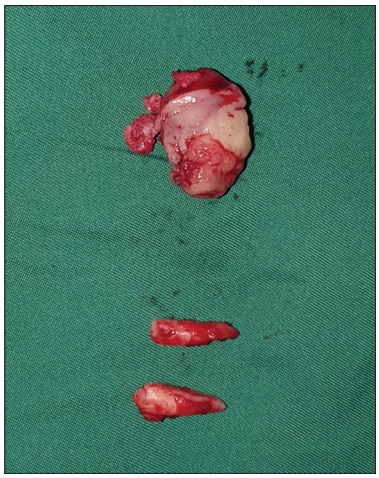

Figure 5: Gross specimen after enucleation including teeth #10 and #11.

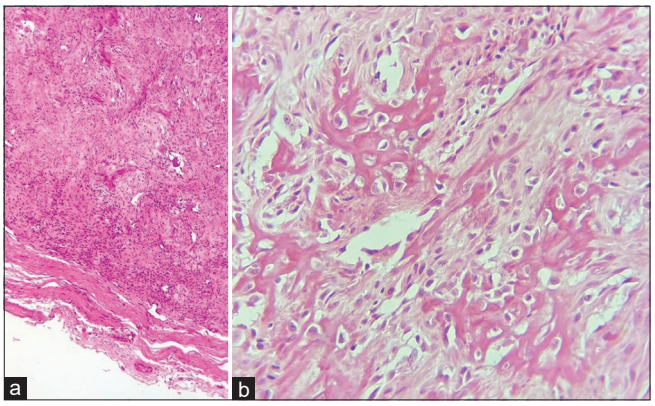

Figure 6: (a) Photomicrograph showing the capsule surrounding the tumor (H and E staining, ×100). (b) Photomicrograph showing irregular osteoid formation and cellular woven bone (H and E staining, ×400).

involves more commonly maxilla, paranasal sinuses, orbit, and frontal and ethmoid bones, therefore nasal obstruction, proptosis, and exophtalmous may be seen in patients.[9] Recurrence rates of 20%–58% have been reported.[10] According to the WHO classification, there are two histological subtypes of this lesion: Juvenile trabecular ossifying fibroma (JTOF) and juvenile psammomatoid ossifying fibroma (JPOF).[1] JPOF is more commonly reported than the trabecular type.[2] JTOF is more common in the maxilla and is usually seen in children and adolescents. The latter form occurs in patients with a wider age range (16–33 years), and more commonly arises from the paranasal sinuses.[1,6] JTOF is often asymptomatic and generally, tooth displacement is the primary clinical symptom.[7,11,12] Radiographically, JTOF tends to show concentric expansion in all directions, and this expansion may result in tooth displacement. Cortical

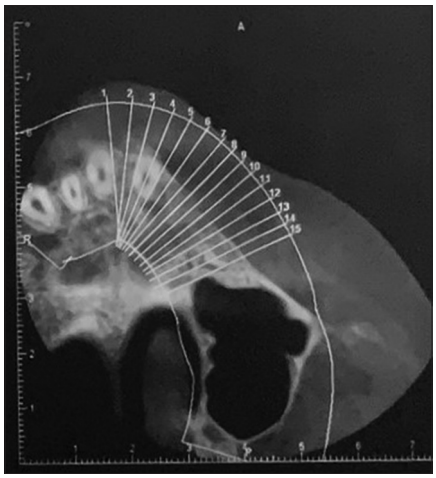

Figure 7: Cone-beam computed tomographic image of the maxillary alveolar ridge 6 months after surgery.

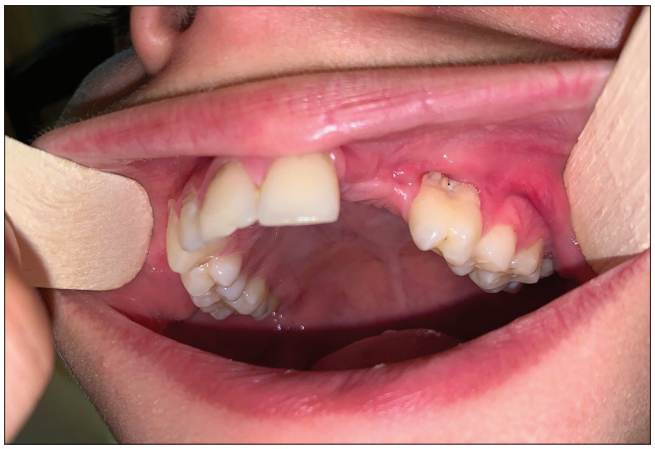

Figure 8: Intraoral view of the left maxillary alveolar ridge 6 months after surgery.

plate destruction is not a common finding in JTOF, despite significant expansion.[13]

POFs are rather common and account for 3.1% of all oral tumors.[14] Clinically, POF can be seen as a pedunculated or sessile nodular mass, which is usually located in the interdental papilla.[15] It predominantly affects the adolescents and young adults, with a peak prevalence between 10 and 19 years.[7] Its color is similar to that of the mucosa unless the lesion is ulcerated. POF is slightly more common in the maxilla and has a potential to cause tooth displacement.[16]

In this study, we reported a 13-year-old male with a fibro-osseous lesion in his anterior maxilla. The incisional biopsy of the lesion initially reported POF. However, after enucleation of the lesion, the specimen was histopathologically analyzed, and the findings were suggestive of JTOF. According to the current literature, JTOF is not associated with cortical bone destruction even in extremely locally advanced cases,[13,16,17] which is in contrast to the clinical and radiographic features of our case.

According to the aforementioned histopathological findings, we suggest that the concurrent eruption of permanent canine tooth led to the destruction of cortical plate and subsequent herniation of the central JTOF. This might have resulted in the formation of a peripheral lesion that was mistaken for a POF in the first-stage surgery. The location of the peripheral lesion, which was exactly beneath the malerupted canine tooth can support this hypothesis. The aforementioned coincidence might have led to early detection of the lesion present in the jaw. As reported in numerous cases, JTOF can enormously grow in bone with no pain or any prominent extra-oral deformity. The management of such cases may require a sacrifice of dental and anatomical structures.[16,18]

It is important to note that although there is no potential for malignant transformation of JTOF,[11,12,19] risk of recurrence is not negligible. The existing literature reports a recurrence rate of 30%–67% following incomplete excision.[11,19,20] However, according to a review article by Chrcanovic and Gomez,[5] the recurrence rate can be minimized by enucleation complemented by ostectomy or curettage. In our case, due to the small size of the lesion and its accessibility, enucleation, and curettage were performed. After 6 months, clinical examinations showed no recurrence, and CBCT images revealed reasonable remodeling of the affected bone. No sign of recurrence was observed in 14-month clinical follow-up.

CONCLUSION

In this study, we reported a 13-year-old male with a fibro-osseous lesion in his anterior maxilla. After enucleation of the lesion, histopathological findings of the specimen were suggestive of JTOF. The patient was successfully treated with no signs of recurrence after 14 months.

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Conflicts of interest
The authors of this manuscript declare that they have no conflicts of interest, real or perceived, financial or nonfinancial in this article.

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