Juvenile ossifying fibroma of maxilla: Concurrent presentation of psammomatoid and trabecular variant

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Abstract

Juvenile Ossifying Fibroma (JOF) is a type of ossifying fibroma which occurs in younger individuals and manifests as trabecular and psammomatoid variants. The nature and behaviour of these variants vary, and they exhibit characteristic histopathological appearance. The solitary presentation of these subtypes is reported in numbers, but co-occurrence of both these entities is very few. Here, we present a case of JOF with the co-occurrence of both trabecular and psammomatoid variants in relation to an incompletely healed extraction socket.

Keywords: Juvenile ossifying fibroma, juvenile psammomatoid ossifying fibroma, juvenile trabecular ossifying fibroma, maxilla

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Submitted: 12-Oct-2023, **Accepted:** 19-Oct-2023, **Published:** 20-Dec-2023

INTRODUCTION

Juvenile ossifying fibroma (JOF), a rare subtype of ossifying fibroma, occurs in young individuals and is characteristic of its aggressiveness and high recurrence. Unlike conventional ossifying fibroma, which is slow-growing with less recurrence potential, JOF poses rapid growth and osteolytic behaviour.[1,2] It affects the craniofacial structures including ethmoid, frontal and paranasal sinuses, with maxilla being the predominant site of occurrence than mandible.[3] Variation in the histopathological features of JOF resulted in further sub-classification of the entity as Juvenile Trabecular Ossifying Fibroma (JTOF) and Juvenile Psammomatoid Ossifying Fibroma (JPOF).[4] Though the name being 'juvenile', usually occurring between the age group of 5 and 15 years, both JTOF and JPOF also manifest in the broader age range.[5] Solitary manifestation of these histological subtypes occurs commonly, while the concurrent presentation of both patterns is a rarity. Here, we report one such case with concurrent histopathological presentation of JTOF and JPOF.

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Website: https://journals.lww.com/JPAT/

DOI: 10.4103/jomfp.jomfp_443_23

Case presentation

A 44-year-old male reported to the outpatient department with severe pain and discharge in his upper left posterior tooth region for the past 2 months. Patient revealed a history of trauma 25 years back in the maxillary region, on the left side of his face. Extraction of 27 (FDI System) was performed one month back elsewhere for persistent discharge from that site. Extraoral findings revealed no swelling or facial asymmetry. Intra orally, 27 was missing, and the socket was not healed completely.

How to cite this article: Sivapathasundharam B, Prabhakar M, Sabarinath B. Juvenile ossifying fibroma of maxilla: Concurrent presentation of psammomatoid and trabecular variant. J Oral Maxillofac Pathol 2023;27:756-9.

A solitary swelling, measuring about 1 × 1 cm^2 was evident on the buccal aspect of 27 regions extending from the distal aspect of 26 to the mesial aspect of 28, without obliteration of the vestibule [Figure 1]. On palpation, the surface of the swelling was smooth, hard and non-tender. Intra-oral periapical radiograph revealed no evidence of maxillary sinus floor perforation in extracted 27 regions. A provisional diagnosis of oroantral fistula in relation to 27 was given.

Histopathological findings

Pre-operative incisional biopsy was performed and the histopathological feature revealed hypercellular fibrous connective tissue stroma with numerous foci of irregular bony trabeculae and calcified spherical structures [Figure 2]. The bony trabeculae showed varying degrees of maturity without the presence of osteoblastic rimming in few areas. Proliferation of fibroblastic-spindle cells, with areas of osteoid formation, was noticed surrounding the trabeculae, giving a paintbrush stroke appearance [Figures 3 and 4]. Numerous deeply stained basophilic concentric calcifications were also seen in the stroma, representing cementum-like psammomatous structures [Figure 5]. These structures were surrounded by parallely arranged slender strands of osteoid with their proximal (inner) ends being basophilic suggestive of initial stage of mineralization [Figure 5]. The lesion was then excised surgically and the post-operative specimen showed similar findings.

DISCUSSION

Ossifying fibroma affecting the jaw bones are categorized clinicopathologically as ossifying fibroma of odontogenic origin, usually called as cementifying fibroma, and the other two distinct juvenile variants such as psammomatoid and trabecular.[6] Several case series about JPOF and JTOF were reported earlier, mentioning their demographics and clinical presentation. A wide distribution of age is noticed in both variants, ranging from 16 to 33 years for psammamatoid and 8.5 to 12 years for trabecular.[2,6,7] This wide array of age distribution takes the nomenclature of this lesion towards contradiction. The name 'juvenile', with its varying demographics, raises a question and an opinion from the author, whether the same nomenclature can be retained for this entity of ossifying fibroma. Several authors had earlier titled this lesion as juvenile active and juvenile aggressive ossifying fibromas; however, the term 'juvenile' clings to the terminology until now. The adjective 'juvenile' is misleading, since the lesion occurs even in the adult and elderly age group.

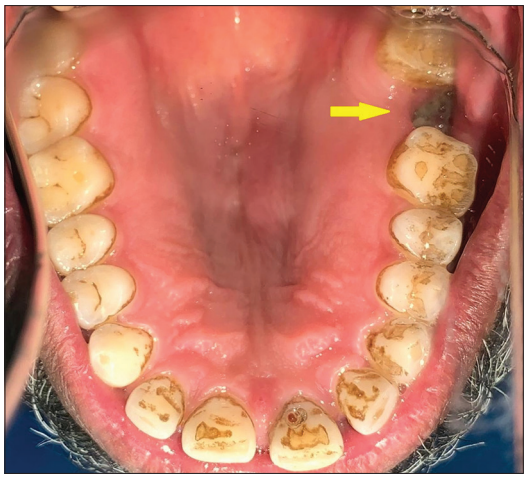

Figure 1: Intraoral photograph showing unhealed extraction socket with mild swelling in relation to 27 region

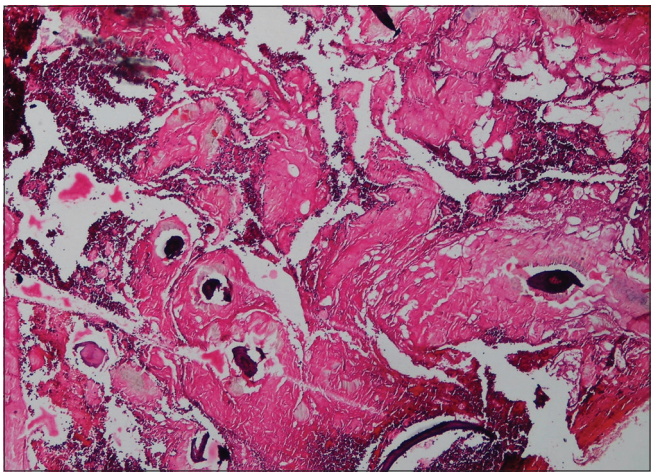

Figure 2: Hypercellular fibrous connective tissue stroma showing irregular bony trabeculae and calcified spherical structures

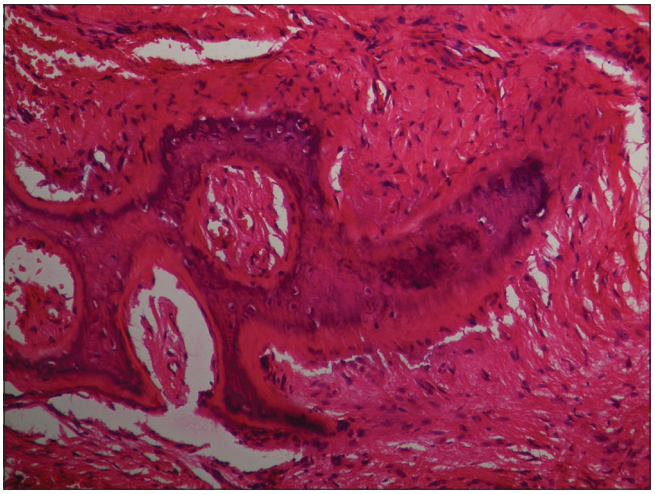

Figure 3: Fibroblastic-spindle cells surrounding the trabeculae, giving a paintbrush stroke appearance

Moreover, co-occurrence of psammamatoid and trabecular histopathological pattern is infrequent, and such a

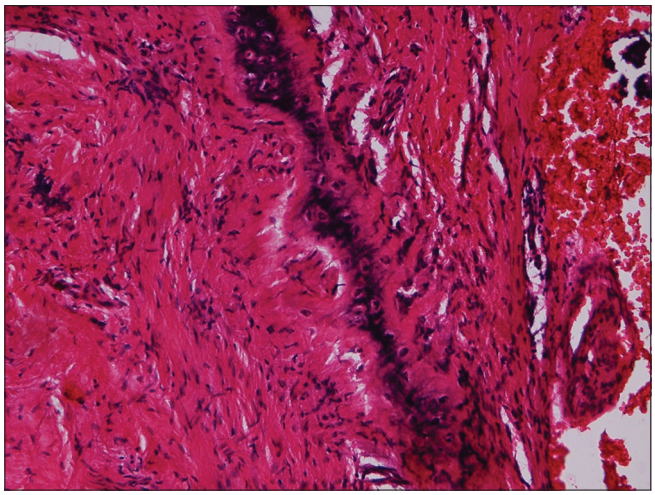

Figure 4: Fibroblastic-spindle cells surrounding the trabeculae, giving a paintbrush stroke appearance

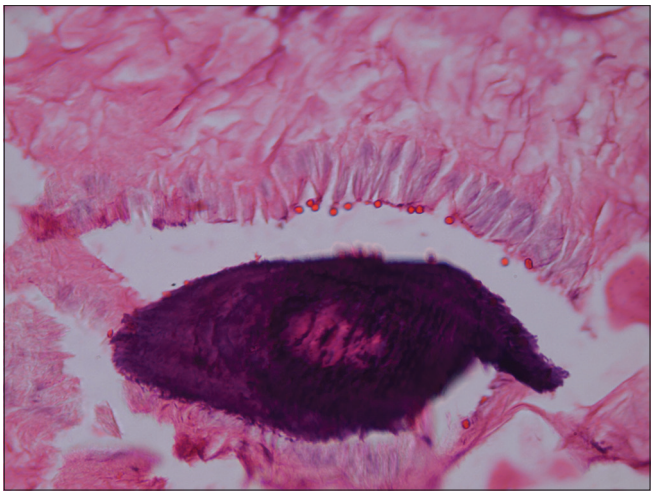

Figure 5: Psammomatous structures surrounded by parallelly arranged slender strands of osteoid

presentation is reported in this case. A couple of cases with the co-existence of both variants were reported earlier, one in the skull base and the other in the maxilla.[2,8] The reason for the concurrence of both these entities may be attributed to the cell-rich stroma containing fibroblastic-spindle cells, surrounding osteoblasts and immature cellular osteoid. Since the behaviour of the lesion varies between psammomatoid and trabecular, with trabecular being more aggressive, the behaviour of this dual-type lesion is foreseen to be even more erratic. Recurrence rate, malignant transformation and secondary changes are to be continuously monitored and followed up in this type of conglomerated variant.

Generally, JOF has varying distribution of site of occurrence, with JPOF predominantly manifesting in the sinonasal and orbital bones, whereas JTOF affects the jaws. Psammomatoid variant shows deeply stained basophilic spherical or ossicles in an intense or loose fibroblastic stroma, called 'psammoma bodies'. These basophilic acellular mineralized structures are encompassed by thorn-like or thread-like calcified eosinophilic strands, giving a 'brush border appearance'. The trabecular variant is composed of multiple areas of inter-connecting bony trabeculae with varying degrees of maturation, forming a lattice-type network, with few areas of the trabeculae exhibiting osteoblastic rimming. The osteoid surrounding the trabeculae blends with the surrounding fibrocellular stroma in few areas, forming a paintbrush strokes appearance.[2,3,9,10] The present case exhibits slender strands of osteoid surrounding the basophilic psammomatoid structures, a distinctive presentation rarely reported.

Differentiating ossifying fibroma from other fibro-osseous lesions becomes intricate, when it occurs in the tooth-bearing region. Rich cellular fibroblastic stroma with osteoid formation, anastomosing trabeculae of immature woven bone, and basophilic structures with brush borders distinguish the present lesion from other benign fibro-osseous lesions like Cemento osseous dysplasia and fibrous dysplasia. The presence of concentric basophilic structures resembles cementicles at times, which may create a diagnostic perplexity between JOF and Cemento-ossifying fibroma. However, these two entities can be differentiated by their histologic and radiographic appearance.[2,5] Surgical excision is the preferred way of managing this kind of lesion, as it shows an aggressive nature. The present case was also managed surgically by excising the lesion completely with the surrounding clear margin. The patient is under follow-up and is monitored for any recurrence.

Summary

Several cases have been reported earlier in the literature explaining both the patterns of JOF; but the question of retaining the terminology as 'juvenile', when the lesion actually occurs in a wide age group has not yet been addressed. Moreover, reports with co-occurrence of both the patterns of JOF are very minimal, and the behaviour of this concurrent type needs a mere understanding of its origin to manage the lesion appropriately. In addition, the occurrence of JOF from an improperly healed extraction socket is unusual. Usually, this kind of unhealed socket may mislead clinicians towards alveolar osteitis, ending up with localized and conservative management. Any unhealed extraction socket should never be missed or ignored, as it may portray a different histopathological picture later, as in this case. Correlation of both clinical and pathological entities will help clinicians to arrive at a definite diagnosis in such a scenario.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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