

Psammomatoid Juvenile Ossifying Fibroma of the Mandible – A Histochemical insight!

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Citation

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Abstract

Juvenile ossifying fibroma (JOF) are fibro osseous lesions that mostly occur in children. Although a benign entity, JOF is known to be locally aggressive and has a high tendency to reoccur. Two distinctive microscopic patterns have been described; a trabecular variant and a Psammomatoid variant. This latter variant is predominantly a craniofacial lesion and occurs rarely in the jaws. We report a rare case of Psammomatoid Juvenile Ossifying that occurred in the mandible of a young boy along with review of the relevant literature. In addition, the pathology of the lesion was also analyzed with picosirus red stain and polarizing microscopy

INTRODUCTION

Juvenile Ossifying fibroma is a fibro-osseous entity that arises within the craniofacial bones of young individuals below 15 years of age [1]. Juvenile Ossifying fibroma is a broad term to describe two distinct histopathological variants: Trabacular Juvenile Ossifying fibroma which is distinguished by the presence of trabeculae of fibrillar osteoid and woven bone and Psammomatoid Juvenile Ossifying fibroma (PsJOF) that is characterised by the presence of small uniform spherical ossicles that resemble Psammoma bodies [2].

PsJOF was first reported by Benjamins in 1938, who called the lesion “osteoid fibroma with atypical ossification of the frontal sinus” [3]. Later, Gogl in 1949 reported two cases of what he called “Psammomatoid ossifying fibroma of the nose and paranasal sinus” [4]. In 1952, Johnson termed the same lesion “Juvenile active ossifying fibroma” and described it as a cellular mass which generates innumerable small uniform sized osteoid bodies [5]. Makek in his review of fibroosseous lesions categorized these lesions as Psammous desmo-osteoblastomas [6]. Although PsJOF is an aggressive lesion with a predilection for children in the age range of 5-15 years [7] there are reports where the patient age ranged from 3 months to 72 years [568]. A slight male predominance has been observed in several case reports and clinico-pathological studies of PsJOF [2689].

Samik El Mofty [2] observed a significant demographic

difference between the two variants of Juvenile Ossifying fibroma, while the trabecular variant is predominantly a gnathic lesion, with a predilection for the maxilla, PsJOF occurs overwhelmingly in the sinonasal and orbital bones. Johnson et al [5] and Makek [6] in their case series reports also found that 70% of the PsJoF's occurred in the Paranasal sinus, 20% in the maxilla and only about 10% in the mandible. Here, we report an additional case of PsJOF that occurred in the mandible an uncommon site for this PsJOF. The histopathology of the lesion was also analysed with the aid of picosirus red stain and polarizing microscopy to correlate the pathology of the lesion with its clinical behaviour.

CASE REPORT

A 13 year old boy reported to dental clinic of Manipal college of Dental sciences, Manipal in June 2006 with a chief complaint of a swelling on the right side of his face.. The swelling had developed 2 months back, and the size and firmness of the swelling had remained the same since then.

An extra Oral examination revealed a 4x 4 cm swelling on the right body of the mandible extending from the angle of the mouth to the inferior body of the mandible (Fig1). The swelling was bony hard in consistency. There was no discharge pus, parasthesia of the lip or restricted mouth opening.

Figure 1

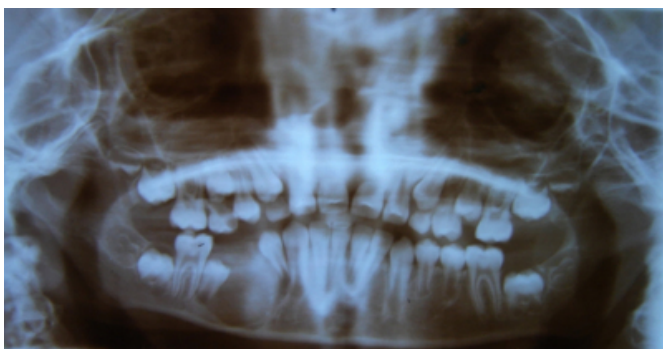
Fig 1: A firm swelling on the right side of the face



Intra-Oral examination revealed a bony hard, non tender swelling of the right mandibular alveolar ridge extending from Permanent right canine to the Permanent first molar. The swelling obliterated the buccal vestibule. An orthopantomograph showed a unilocular radiolucency extending from the distal aspect of erupting mandibular right first premolar to the Permanent mandibular right first molar. The unerupted second Premolar was evident in the radiograph (Fig 2).

Figure 2

Fig 2: Panoramic radiograph shows a large radiolucent area with radio opacities in the right mandible.



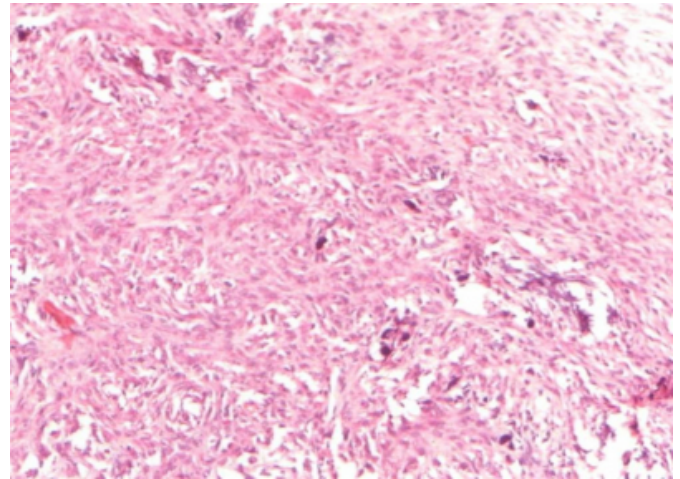
On the basis of the clinical presentation, the lesion was diagnosed as an odontogenic cyst. Preoperative laboratory investigations were within normal limits. Complete surgical excision of the lesion was performed and the tissue was sent for histopathological examination. Following post operative care, the patient was discharge. He has been on regular follow up since then and there is no evidence of recurrence so far.

Microscopic examination of the lesional tissue revealed a

cellular connective tissue stroma. Numerous spindle shaped cells arranged in a fascicular storiform pattern formed the major portion of the stroma (Fig 3).

Figure 3

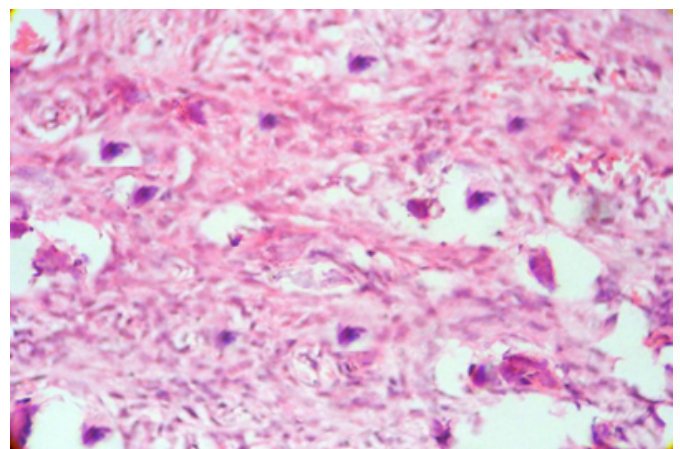
Fig 3: Photomicrograph shows cellular stroma with spindle shaped cells arranged in Strands and whorls and small psammoma- like bodies (Haematoxylin and Eosin 10X)



Irregular strands of osteoid matrix rimmed with plump osteoblasts were scattered in the connective tissue stroma. Many giant cells and areas of hemorrhage were also evident in the stroma. Spheroidal ossicles with a basophilic centre and eosinophilic periphery that resembled Psammoma-like bodies were also dispersed in the connective tissue stroma (Fig 4). The sections were also stained with picrosirius red and the birefringence pattern was observed by polarizing microscopy.

Figure 4

Fig 4:: Photomicrograph shows Psammoma-like bodies in the cellular stroma (Haematoxylin and Eosin stain 40X)



DISCUSSION

The spectrum of fibro-osseous lesions includes a variety of

developmental, reactive or dysplastic lesions and neoplastic entities. Juvenile Ossifying fibromas are benign, potentially aggressive fibroosseous lesions of the craniofacial bones. PsJOF is an unique variant of JOF that has a predilection for the sinonasal tract and the orbit [5689]. The lesion has demonstrated a potential to proliferate, invade and destroy tissues until the eyes and the cerebrospinal space are affected [9]. In addition to its aggressive behaviour this lesion also has a very strong tendency to recur, and recurrence rate as high as 30%-56% have been reported [5681011].

PsJOF are predominantly craniofacial lesions and jaw lesions are very rare. The pathogenesis for these jaw lesions are related to the maldevelopment of basal generative mechanism that is essential for root formation. The developing tooth can either be displaced, missing or remain unerupted [5]. In the current case the first premolar was slightly displaced and the second premolar was unerupted

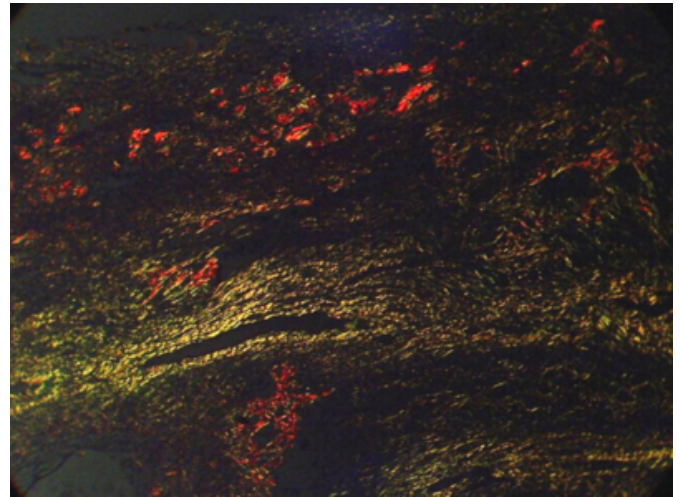
The pathognomonic feature of this fibroosseous lesion is the presence of eosinophilic spherical structures dispersed in a fibrous stroma consisting of plump spindle shaped cells that are arranged as strands and whorls. Gogl [4] was the first to term these unique spherical structure as Psammoma- Like bodies. These particles vary in appearance, but usually have a central basophilic area and a peripheral eosinophilic fringe [9] (Fig 5). Ultrastructurally, Psammoma-like bodies in PsJOF were found to possess a dark rim of crystals from which small spicules and needle like crystalloids project toward the periphery [12].

It has been established that Picrosirius red staining followed by polarizing microscopy can selectively demonstrate collagen in tissue sections [13] and the different birefringent colour in tissue sections is related to the distinct physical aggregation of collagen fibers [14]. The color profile ranging from green- greenish yellow is related to poorly packed collagen fibers, while orange red- red color profile represents well packed collagen fibers [15].

The birefringence pattern of both the osseous trabeculae and the psammomatoid bodies in the lesional tissue of PsJOF was intense red. Thus, psammoma like -bodies and osseous trabeculae contain mature collagen fibers that are well-packed and hence give an intense red colour profile. On the other hand, the collagen fibers in the lesional tissue gave a green to greenish- yellow birefringence (Fig 5).

Figure 5

Fig 5: Polarizing colours of picrosirius red stain: Psammoma-like bodies and osseous trabeculae show intense red birefringence while the collagen fibers show green- greenish yellow color profile (Picrosirius Red stain 10X)



Hirschfield et al in their study on the birefringence pattern of the connective tissue wall of odontogenic cysts found that collagen fibers in the stroma of keratocyst had a green-greenish-yellow color profile. These poorly packed collagen fibers were presumed to be composed of procollagen intermediates or pathological collagen [16].

Odontogenic Keratocysts are entities known to possess an aggressive biological behaviour and has a potential to recur. The biological behaviour as well as the picrosirius red staining color profile of collagen fibers of PsJOF appears to be identical to that of keratocysts. Hence, predominance of these green -greenish yellow collagen fibers may signify an aggressive behaviour of the lesion. A higher ratio of green to greenish -yellow and orange- orange red fibers may also be an indicator of the aggressive potential of the lesion. Thus, the neoplastic cells of PSJOF may be proliferating immature cells that are capable of inducing the formation of a functional stroma of poorly packed pathological collagen fibers.

CONCLUSION

Psammomatoid Juvenile ossifying fibromas are unique lesions that occur commonly in children. Psammoma- Like bodies are the hallmark of this neoplasm. Mandibular lesions are uncommon and can be mistaken for an Odontogenic cyst. Clinical, radiographic histopathological correlation will be beneficial in arriving at the accurate diagnosis. Picrosirius red staining followed by polarizing microscopy enhances the histological assessment of the lesion and can provide additional information on the behaviour of this fibro osseous

entity that can influence treatment. In addition the high recurrence rate of this fibro osseous lesion warrants continued patient follow-up.

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