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Psammomatoid Juvenile Ossifying Fibroma in a Young Adult: Clinical, Radiological, and Histopathological Perspectives

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Abstract

Fibro-osseous lesions of the craniofacial skeleton encompass a broad spectrum of developmental, neoplastic, and reactive conditions wherein normal bone is replaced by a fibrous stroma containing varying degrees of mineralized material. These lesions often present diagnostic challenges due to their overlapping clinical, radiographic, and histopathological features. Among them, Juvenile Ossifying Fibroma (JOF) is a rare but distinct entity, typically occurring in children and adolescents. Known for its locally aggressive behavior and rapid expansion, JOF may cause significant facial deformity, displacement of teeth, and functional complications if not addressed promptly. Histologically, JOF is categorized into two variants: trabecular and psammomatoid. We are presenting a case of Juvenile aggressive ossifying fibroma in a 20-year-old girl who reported to our institute.

Keywords: fibro-osseous lesion, juvenile ossifying fibroma, mandible, ossifying fibroma

Introduction

Juvenile ossifying fibroma (JOF) is a rare fibro osseous entity of the craniofacial skeleton, which poses diagnostic and therapeutic challenges, due to its characteristic behavioral, clinical, and histopathological features.[2] The second edition of the World Health Organization (WHO) Classification of odontogenic tumors defines JOF as a lesion consisting of cell-rich fibrous tissue containing bands of cellular osteoid without osteoblastic rimming with trabeculae of more typical woven bone.[2]

Juvenile ossifying fibroma is a benign bone forming neoplasm, which is defined as a variant of ossifying fibroma occurring in the craniofacial skeleton of young patients.[3] It arises within the craniofacial bones of children under the age of 15 years; however, there are reports of ossifying fibromas in patients with ages ranging from three months to 72 years.[4] JOF differs from the adult variant of ossifying fibroma on the basis of the age of occurrence, anatomic site of involvement, high

tendency for recurrence, and its locally aggressive behaviour.[5]

According to the new edition of the classification of the World Health Organization,[6] ossifying fibromas which appear as fast-growing mass between 5 and 15 years of age, radiologically well bordered, and consistent with ossifying fibroma histologically, are referred as juvenile (aggressive) ossifying fibroma. Juvenile ossifying fibroma (JOF) appears at an early age and in 79% of the patients are diagnosed before the age of 15.[6,7] Males and females are equally affected. JOF originates from periodontal ligament and ranges 2% of oral tumours in children.[8] The JOF is located mainly (85%) in facial bones, in some cases (12%) in calvarium, and very seldom (3%) extracranially.[2] Ninety percent of the lesions located in the face region, involve the sinuses, mainly the maxillary antra.[2.] Mandibular lesions are seen in 10% of the cases.

Histologically, Juvenile Ossifying Fibroma (JOF) is characterized by a highly cellular fibrous stroma composed of plump spindle-shaped fibroblasts arranged in a loosely interwoven pattern. Within this stroma, varying degrees of mineralized material are seen, including immature woven bone, osteoid, and cementum-like deposits. A notable feature is the presence of garland-like strands of immature bony trabeculae and cementicle-like structures. JOF is subclassified into two distinct variants: the trabecular and psammomatoid types, based on the pattern of mineralization. The trabecular variant (TrJOF) is defined by the presence of trabeculae of fibrillar osteoid and immature woven bone, often surrounded by a cellular stroma and occasionally rimmed by osteoblasts. This variant is more commonly found in the jaws, particularly the maxilla. In contrast, the **psammomatoid variant** (PsJOF) demonstrates numerous small, spherical, uniform ossicles resembling psammoma bodies. These ossicles are concentrically laminated and scattered throughout the cellular stroma. PsJOF is reported more frequently than the trabecular type and predominantly involves the sinonasal region and orbital bones, though it can occasionally present in the jaws. Among the two variants, PsJOF is considered more aggressive and has a higher propensity for recurrence, underscoring the importance of accurate histopathological diagnosis and close postoperative follow-up.

Case report

A 20-year-old female patient was reported to the hospital with a chief complaint of swelling in the left back region of the lower jaw for around 18 months. The swelling started slowly to attain the present size. The medical history of the patient was not significant. On extra oral examination, the swelling was oval (Figure 1). Intra-oral examination revealed ovoid swelling present solitary well-defined swelling, bony hard, non-tender swelling extending from mesial surface of the first premolar to first molar posteriorly (Figure 2). Single diffuse swelling was noted in region lateral to the right commissure of the lip. Measuring size 1.6x1.7cm. On palpation swelling is firm in consistency, fixed to underlying tissue, no change in skin colour.

Figure 1: Extraoral photograph showing swelling in right mandibular region

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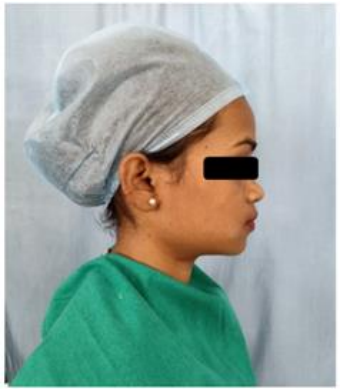

Figure 2: Intraoral photograph showing swelling in the posterior mandible from premolar region to molar area. Radiographic examination revealed Well-defined roughly oval-shaped unilocular radiolucency with scalloped borders noted at right mandible extending super inferiorly from the alveolar crest of 44,45,46 (Figure 3). Multiple radiopacities were noted within the well-defined radiolucent lesion. Partial loss of labial cortical plate Thinning of the lingual cortical plate. A displaced root of 45 was noted.

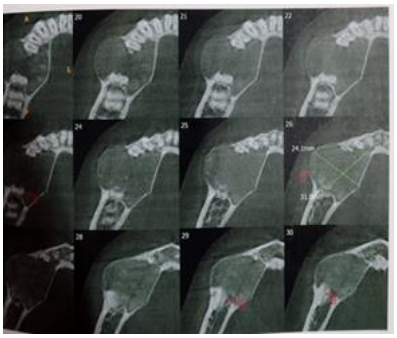

Figure 3: CBCT showing a Well-defined roughly oval-shaped unilocular radiolucency with scalloped borders noted at right mandible extending super inferiorly from the alveolar crest of 44,45,46.

Considering the extent of the lesion, an Excisional biopsy was performed which revealed multiple bits of soft tissues Measuring about -4x2x2 cm approx. 1.5x1.5x0.5 cm measuring approx. irregular in shape brownish white in colour, soft to firm in consistency (Figure 4).

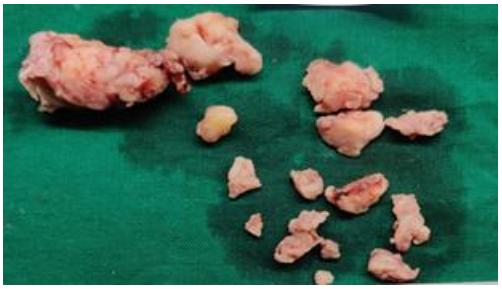

Figure 4: Macroscopic view of the tissue.

Later, H&E-stained studied sections show presence of connective tissue only. The connective tissue is composed of numerous plump proliferating fibroblasts interspersed by delicate collagen fibre bundles and a few endothelial-lined capillaries. Lamellar & woven bone rimmed by osteoblast present along with mineralized component. At places, concentric lamellated and spherical asides that vary in shape and have basophilic centers and show varying degrees & stages of calcification. At the periphery reactive bone formation is noted. Histological features were consistent with Psammomatoid juvenile ossifying fibroma (Figure 5).

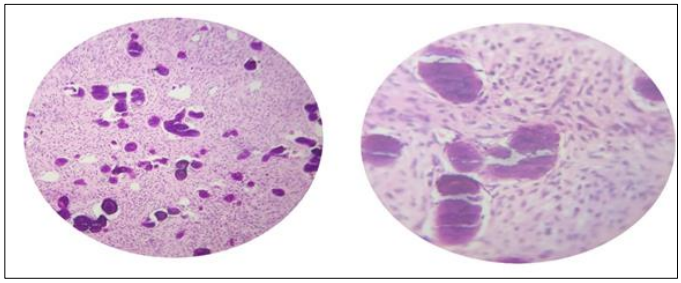

Figure 5 A & B: Spherical calcified ossicles resembling psammoma bodies are embedded in a cellular fibrous stroma. The ossicles have irregular margins in contrast to psammoma bodies and are variably calcified (under magnification 10x and 40x)

Discussion

Terminology

The term "psammos" originates from the Greek word *psammos*, meaning "sand," reflecting the characteristic microscopic appearance of small, round, calcified structures resembling sand particles. Over the years,

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various terminologies have been used in the literature to describe this lesion, highlighting its histopathological features and clinical behavior. A summary of these terminologies is provided below:

Table 1:

Author	Year	Terminology Used
Benjamins	1938	Osteoid fibroma with atypical ossification
Gögl	1949	Psammomatoid ossifying fibroma
Johnson et al.	1952	Juvenile active ossifying fibroma
Makek	1983	Psammous desmo-osteoblastoma (a variant of osteoblastoma)
Slootweg et al.	1994	Juvenile ossifying fibroma with psammoma-like ossicles
Wenig et al.	1995	Aggressive psammomatoid ossifying fibroma
Hartstein et al.	1998	Psammomatoid ossifying fibroma
World Health Organization (WHO)	2005	Juvenile psammomatoid ossifying fibroma

The term “active” was originally employed to describe lesions demonstrating marked cellular proliferation and a tendency toward aggressive clinical behavior, including bone destruction, invasion into surrounding anatomical areas, and, in rare cases, life-threatening progression. However, Wenig et al. argued against the continued use of this term, noting that histological assessment alone does not provide a reliable measure of the lesion's duration or biological behavior. Additionally, labels such as “juvenile” or “young” were deemed inappropriate, since such lesions can manifest across a wide age range and are not confined to pediatric populations.[7]

Johnson et al. suggested that these lesions might begin developing during adolescence and remain undetected until adulthood, only becoming apparent when they enlarge sufficiently to produce clinical symptoms. This reasoning, they argued, supports the continued use of the term “juvenile.” On the other hand, Wenig et al. did not report such delayed manifestations and instead proposed the designation “aggressive psammomatoid ossifying fibroma,” emphasizing the lesion’s unique histopathological features and its locally invasive potential.[8]

Subsequently, Hartstein et al. recommended adopting the term "psammomatoid ossifying fibroma" as a more precise and unbiased descriptor, avoiding any assumptions regarding the typical age of onset or clinical behavior. However, the World Health Organization (WHO), in its most recent classification of odontogenic and maxillofacial bone tumors, continues to use the term "juvenile psammomatoid ossifying fibroma", reflecting both its characteristic microscopic features and its relatively common occurrence in younger individuals.[9]

Slootweg et al. identified two distinct variants of juvenile ossifying fibroma (JOF), differentiated by their histological features and typical age of onset. These subtypes are the WHO-type JOF, and the psammomatoid-type JOF (PO-type), the latter being marked by the presence of psammoma-like calcified structures. The WHO-type generally presents at a younger age, with a mean age around 11.8 years, whereas the PO-type is more commonly seen in young adults, with a mean age of about 22.6 years.[10]

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El-Mofty introduced a classification system for juvenile ossifying fibroma (JOF) based on histological differences, identifying two major variants: trabecular JOF (TrJOF) and psammomatoid JOF (PsJOF). These forms also demonstrate distinct age distributions. TrJOF is typically found in children around 8.5 to 12 years old, while PsJOF tends to present in adolescents and young adults, usually between 16 and 33 years of age.[11]

Psammomatoid Juvenile Ossifying Fibroma (PsJOF) often manifests with symptoms such as proptosis, ptosis, visual impairment, nasal blockage, and headaches, particularly when the lesion involves the orbit or paranasal sinuses. In certain instances, the lesion may behave aggressively, invading adjacent structures and causing bone erosion, potentially resulting in facial asymmetry, optic nerve compression, or vision loss. On rare occasions, meningitis may arise due to direct communication between the sinuses and the cranial cavity. While PsJOF typically affects the orbit, sinonasal tract, and cranial bones, approximately 25% of cases involve the jaws. In contrast, trabecular JOF (TrJOF) is most often found in the maxilla, presenting as a slow-growing, painless swelling. Nonetheless, atypical presentations of PsJOF in the maxilla or mandible have been reported, occasionally demonstrating multifocal and aggressive features.[11]

The development of this lesion may be associated with disturbances in root formation, and genetic alterations, particularly chromosomal translocations at Xq26 and 2q33, have been suggested as contributing factors in its etiopathogenesis.[12]

On radiographic examination, juvenile ossifying fibroma (JOF) typically presents as a well-circumscribed lesion that may appear unilocular or multilocular, with radiolucent, mixed, or radiopaque features, depending on the extent of mineralization or cystic transformation. Characteristically, these lesions may show fine internal radiopacities, often described as having a ground-glass appearance, and are frequently bordered by a thin, sclerotic rim, sometimes resembling an eggshell outline. In more aggressive cases, expansile bone remodeling is evident, though the lesion typically maintains clear boundaries from the adjacent bone. There may also be signs of cortical thinning or localized cortical breach. As the lesion matures, its radiographic features may evolve. JOF can resemble other fibro-osseous or osseous lesions, such as fibrous dysplasia, cemento-ossifying fibroma, aneurysmal bone cyst, osteosarcoma, and osteoblastoma. However, it can be differentiated based on its well-defined borders, which contrast with the ill-defined margins of fibrous dysplasia, and by the lack of periosteal reaction, which helps distinguish it from osteosarcoma.[13,14]

Histopathological examination is essential for confirming the diagnosis of Psammomatoid Juvenile Ossifying Fibroma (PsJOF). A key diagnostic characteristic is the presence of numerous spherical calcified structures, commonly known as psammoma-like ossicles, dispersed within a highly cellular fibrous stroma. These ossicles are typically eosinophilic, with a central basophilic zone encircled by an eosinophilic rim, creating a concentric or laminated appearance. These distinct structures were first identified and described by Gögl, who termed them "psammoma-like bodies," derived from the Greek term *psammos*, meaning "sand," due to their granular resemblance.[11]

The stromal component of Psammomatoid Juvenile Ossifying Fibroma (PsJOF) typically consists of plump, spindle-shaped fibroblastic cells, which may be organized in fascicles, whorled patterns, or interlacing strands. The amount of collagen within the stroma is variable, ranging from loosely arranged fibrous tissue to

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regions that are highly cellular with scant intercellular matrix. In some cases, the lesion also contains trabeculae of immature woven bone or mature lamellar bone. Additional findings may include pseudocystic spaces, areas of hemorrhage, or features suggestive of aneurysmal bone cyst-like transformation.[14]

On ultrastructural examination, the psammoma-like ossicles in Psammomatoid Juvenile Ossifying Fibroma (PsJOF) display a dense, mineralized outer rim composed of crystalline material, from which fine spicules or needle-like projections radiate outward. These calcified components are useful in distinguishing PsJOF from other fibro-osseous lesions. A notable microscopic feature of these ossicles is the presence of a "brush border" pattern, characterized by hair-like extensions at the periphery. These radiating mineralized structures extend outward from a central core, creating a fringed or brush-like appearance under light or electron microscopy.[16]

The presence of this brush border around the psammoma-like ossicles is a useful diagnostic clue for identifying Psammomatoid Juvenile Ossifying Fibroma (PsJOF). When assessed alongside other characteristic features—such as a densely cellular fibrous stroma and spindle-shaped fibroblastic cells arranged in whorls or fascicular patterns—this ultrastructural detail can aid in differentiating PsJOF from other fibro-osseous lesions.[17]

Under microscopic examination, it is important to differentiate Psammomatoid Juvenile Ossifying Fibroma (PsJOF) from other lesions such as ossifying fibromas (OFs) and extracranial meningiomas. In ossifying fibromas, the psammoma-like ossicles typically exhibit a thin and smooth collagenous border, whereas in PsJOF, these ossicles show a thicker, more irregular rim. Although extracranial meningiomas can contain genuine psammoma bodies, they characteristically express epithelial membrane antigen (EMA), a marker that is usually absent in PsJOF.[18]

It is also important to distinguish Psammomatoid Juvenile Ossifying Fibroma (PsJOF) from central cementifying fibroma, which is a benign odontogenic tumor. Central cementifying fibromas predominantly affect adult females and are characterized histologically by a dense fibrous connective tissue stroma containing cementum-like calcifications called cementicles. In contrast to PsJOF, these lesions typically have a less cellular stroma, do not exhibit the psammoma-like ossicles, and generally have a low recurrence rate following surgical removal.[19]

Treatment of Psammomatoid Juvenile Ossifying Fibroma (PsJOF) primarily involves complete surgical removal, which may be achieved through enucleation with curettage for smaller lesions or en bloc resection for larger, more extensive tumors, depending on their size and anatomical location. While conservative surgery may be sufficient for limited lesions, aggressive excision is often necessary to reduce the high risk of recurrence, which has been reported between 30% and 60%. This emphasizes the importance of early diagnosis, thorough surgical management, and long-term monitoring. The use of radiotherapy is generally avoided due to its lack of effectiveness and the potential to induce malignant transformation. Although the lesion can behave aggressively locally, metastasis has not been documented.[20]

Conclusion

Psammomatoid Juvenile Ossifying Fibroma (PsJOF) is a rare but distinct fibro-osseous lesion, often presenting with aggressive growth and potential for recurrence. Early diagnosis and comprehensive surgical management are crucial to achieving favourable outcomes. The presented case underscores the

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importance of differentiating PsJOF from other similar lesions through detailed histopathological analysis. Long-term follow-up is essential to monitor for recurrence and ensure effective treatment. This report contributes to the understanding of PsJOF, emphasizing the need for awareness of its unique clinical and histopathological features to facilitate timely diagnosis and management.

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