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Recurrent Juvenile Psammomatoid Ossifying Fibroma of the Mandible: A Case Report Mandibular Psammomatoid Juvenile Ossifying Fibroma

Dr. Krachi Agarwal[1*], Prof. Riddhi Jaiswal[2]

[1]Senior Resident, Department of Pathology, King George's Medical University, Lucknow, UP

[2]Department of Pathology, King George's Medical University, Lucknow, UP

[*]Corresponding Author

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ABSTRACT

Background: Juvenile ossifying fibroma (JOF) is a rare benign fibro-osseous neoplasm of the craniofacial bones that typically affects children and young adults. It is characterized by local aggressiveness and a high tendency for recurrence. Two histopathological subtypes are recognized: trabecular and psammomatoid. The psammomatoid variant (PJOF) often arises in the paranasal sinuses or orbits and only infrequently involves the jaws.

Case Summary: A 23-year-old female presented with swelling in the left posterior mandible persisting for 3 years with intermittent pain. Imaging revealed an expansile lytic lesion with a ground-glass appearance and no cortical breach. Differentials included dentigerous cyst and aneurysmal bone cyst. Orthopantomogram showed mixed radiolucent–radiopaque changes. Serum calcium, vitamin D, and PTH were normal. Histopathology revealed a cellular fibrous stroma with concentric calcified ossicles resembling psammoma bodies, consistent with PJOF. One month later, the lesion recurred in the mandibular alveolar bone, showing similar histopathological features without malignant transformation.

Conclusion: PJOF of the mandible is rare but demonstrates locally aggressive behaviour and early recurrence if incompletely excised. Complete resection with clear margins and long-term follow-up is essential for management.

Keywords: Juvenile ossifying fibroma; Psammomatoid variant; Mandible; Fibro-osseous lesion; Recurrence

INTRODUCTION:

Ossifying fibromas are benign fibro-osseous tumours of the jaws and craniofacial skeleton, classified into conventional (adult) types and juvenile variants. Juvenile ossifying fibroma (JOF) is an uncommon but important variant occurring in younger patients, with more aggressive behaviour and a higher recurrence rate [1]. Histologically, two distinct subtypes of JOF are: trabecular JOF (TrJOF) and psammomatoid JOF (PJOF) [1]. The trabecular type affects children of age 8–12 years, whereas the psammomatoid type tends to present in older children and young adults. Despite the term "juvenile," cases have been reported across a wide age range (from infancy up to the seventh decade) [2]. It most often arises in the extragnathic craniofacial bones such as the orbit, frontal, and ethmoid sinuses, accounting for 75% of cases, while only about 25% involve the maxilla or mandible. Due to overlapping clinical and radiologic features with other jaw lesions, a careful correlation of imaging, histopathology, and clinical findings is required for accurate diagnosis [2].

In this report, we present a case of juvenile psammomatoid ossifying fibroma occurring in the mandible of

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a young female. The lesion exhibited rapid recurrence after initial biopsy, emphasizing the lesion's aggressive nature. We discuss the diagnostic workup, differential diagnoses, histopathological characteristics, and the importance of complete surgical management of this rare tumour.

Case Report

A 23-year-old female presented to the oral pathology clinic with a swelling in the left lower posterior jaw region persisting for the last 3 years. The swelling was slowly progressive and was associated with dull pain on and off. There was no history of trauma, and her medical, surgical, and family history were unremarkable. On clinical examination, a hard bony expansion was palpable in the left mandibular body and ramus area. The overlying oral mucosa was intact with no ulceration, and the teeth in the region were vital, although there was slight displacement of the adjacent molar.

An orthopantomogram (OPG) revealed a well-defined expansile lesion with mixed radiolucent and radiopaque areas in the left posterior mandible (Figure 1a). Notably, the lesion appeared to encompass the area of the impacted third molar, and visible flecks of calcification. Routine laboratory investigations showed serum calcium, 25-hydroxy vitamin D, and parathyroid hormone levels within normal limits, effectively ruling out any metabolic bone disorders (such as hyperparathyroidism) that could produce jaw lesions. Further a non-enhanced computed tomography (CT) scan of the face was obtained, which showed an expansile lytic lesion involving the left hemimandible with a "ground-glass" appearance, patchy radiopacities and cortical thinning; but no evidence of cortical breakthrough, extra-osseous soft tissue extension, or periosteal reaction. The lesion extended from left mandibular first molar to the ramus. The contralateral mandible, bilateral condyles, rami, maxillary sinuses, orbital walls were intact. Based on the clinico-radiological characteristics, the differential diagnoses considered were a dentigerous cyst and an aneurysmal bone cyst. An aspirational biopsy was attempted prior to surgical intervention, which yielded no fluid. Following this, under local anaesthesia, an incisional biopsy from the lesion in posterior mandibular ramus was performed and submitted for histopathological examination.

On gross, the biopsy specimen consisted of multiple soft and hard tissue fragments, together measuring approximately 2 × 2 × 1 cm. Microscopically, the lesion was composed of a highly cellular fibroblastic stroma arranged in interlacing fascicles and a storiform (whorled) pattern. The stromal cells were spindle-shaped to polyhedral with indistinct cell borders and basophilic nuclei. The most striking feature was the presence of numerous calcified ossicles scattered throughout the stroma. These ossicles were of variable size, round to ovoid in shape, and exhibited concentric lamellation closely resembling psammoma bodies (Figure 2a, 2b). In some areas, the periphery of the ossicles showed a brush border blending into the surrounding fibrous stroma, a characteristic feature of psammomatoid ossifying fibroma. No significant mitotic activity or cellular atypia was noted. There were also foci of congested blood vessels within the stroma, but no evidence of the blood-filled cavernous spaces. Based on the above findings, a diagnosis of juvenile psammomatoid ossifying fibroma was established.

The patient was subsequently scheduled for definitive surgical management. However, within about one month of the incisional biopsy, the patient noticed a re-growth in the left mandibular molar area. Clinical examination and imaging confirmed a recurrent mass in the mandibular alveolar bone (Figure 1b). A repeat biopsy of the lesion was performed to evaluate the nature of the recurrence which showed the same features as described previously, a fibro cellular stroma with plentiful psammomatous calcifications, confirming the persistence of PJOF with no evidence of malignant transformation. The absence of any dysplastic cytology or sarcomatous change was reassuring, indicating that the lesion remained a benign ossifying fibroma. The patient and her family were counselled regarding the need for complete surgical resection of the lesion due to its tendency to recur and importance of long-term follow-up was emphasized.

DISCUSSION

Juvenile ossifying fibroma is a distinct clinical and pathological entity within the spectrum of fibro-osseous lesions of the craniofacial bones. It is considered benign but is noted for its potentially aggressive behaviour

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and high propensity for local recurrence [1]. Unlike conventional ossifying fibroma (presents in middle-aged adults and is slow-growing), JOF affects younger individuals and exhibits a more rapid growth pattern [2].

Based on the predominant type of mineralized material present, JOF is histologically subclassified into trabecular variant (contains trabeculae of woven bone, osteoid) and psammomatoid variant (contains the characteristic psammoma body-like ossicles) [3]. It is important to recognize these subtypes, as they have slightly different predilections: TrJOF often occurs in the jaws (especially the maxilla) of younger children, whereas PJOF commonly involves the sinonasal region and present in older patients.

The features of PJOF can overlap with other benign fibro-osseous lesions and cystic lesions of the jaws, making diagnosis challenging without histopathology. The initial differentials of this case included a dentigerous cyst and an aneurysmal bone cyst (ABC). A dentigerous cyst was considered because the lesion was located in the posterior mandible in the region of an impacted tooth. But, the extent of the lesion beyond just the pericoronal space of a tooth and the histologic findings excluded this possibility. The consideration of an ABC was prompted by the expansile, lytic lesion and the patient's history of intermittent pain. However, in this case, a negative aspirate and absence of histopathological blood-filled channels is more consistent with a solid fibro-osseous lesion.

In the present case, the “ground-glass” radiopacity observed on CT is a feature often associated with fibrous dysplasia [FD] of bone [2]. However, FD presents as an ill-defined bony enlargement blending with normal surrounding bone. In contrast, JOF are usually well-circumscribed and are separated from surrounding bone. Additionally, FD is often a monostotic or polyostotic developmental bone dysplasia and would not ordinarily present with such rapid recurrence after biopsy or partial removal. It is also important for pathologists to distinguish PJOF from an extracranial meningioma with psammoma bodies, a rare entity in the craniofacial bones, by using immunohistochemical markers if needed [2]. Differential diagnosis is summarised in Table 1.

One remarkable aspect of this case was the early recurrence of the tumour just one month after the initial biopsy. This can be attributed to the lesion’s aggressive nature and the fact that only an incisional biopsy (sampling) was performed initially, leaving the bulk of the tumour in situ. The literature reports local recurrence rates of about 30% to 56% if incompletely excised [4, 5]. Malignant transformation has not been documented, but an untreated or recurrent JOF can cause significant local destruction, deformity, and functional impairment due to its growth [6].

The standard treatment for JOF is complete en bloc surgical resection (segmental mandibulectomy or maxillectomy) with a margin of normal bone, wherever feasible [7]. Smaller lesions may be excised by enucleation or curettage with a cuff of surrounding bone [8]. In some reports, adjunctive therapies (cryotherapy or pharmacotherapy) have been attempted, but surgery remains the mainstay. Reconstructive procedures (like bone grafting or microvascular free fibula flap in cases of segmental mandibulectomy) may be required to restore form and function. Long-term follow-up is crucial, as recurrences can occur even years later.

This case also highlights the importance of a multidisciplinary approach in diagnosing and managing fibro-osseous lesions of the jaws. The initial evaluation involved radiologists, oral surgeons, and oral pathologists. Advanced imaging (CT, and if needed MRI) can characterize the extent of the lesion and its effects on surrounding structures. Pathological examination is definitive for distinguishing among the various fibro-osseous lesions. Close cooperation between the surgical team and pathologists ensured that once the diagnosis of JOF was rendered, the patient could be directed to appropriate definitive surgical treatment.

Table 1: Differential diagnosis of Juvenile psammomatoid ossifying fibroma:

S. No.	Differential diagnosis	Clinico-radiological features	Histopathology
1.	Dentigerous cyst	Occurs in posterior mandible in an impacted tooth; unilocular radiolucency without internal calcifications	Fibrous tissue with squamoid epithelium
2.	Aneurysmal bone	Rapid growth; expansile, lytic lesion;	Cavernous blood-filled channels or

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	cyst	positive aspiration of fluid/blood	hemosiderin-laden stromal cells
3.	Fibrous dysplasia	Upto 2nd decade; Slow-growing; diffuse enlargement of bone that blends with normal surrounding bone	Curvilinear trabeculae of woven bone without lamellated ossicles
4.	Psammomatoid Meningioma	6-7th decade; Slow growing; Soft tissue tumour enhancement	Meningothelial cells or whorls with haphazardly arranged psammoma bodies, Epithelial Membrane Antigen positive

CONCLUSION

Juvenile psammomatoid ossifying fibroma is a rare benign tumor of the craniofacial bones presenting in the jaws of young adults. It should be considered in the differential diagnosis of persistent jaw swellings, especially when imaging shows a well-demarcated fibro-osseous lesion with mixed radiodensity or ground-glass appearance. Histopathology reveals pathognomonic psammoma body-like calcifications within a cellular fibrous stroma. Though, malignant transformation is not known to occur, it behaves in a locally aggressive manner with a high recurrence rate if inadequately excised. Optimal management requires complete surgical resection, reconstruction and diligent long-term follow-up. This case report adds to the limited literature on PJOF of the mandible in young adults and underscores the importance of recognizing this entity to ensure timely and effective patient care.

Key Clinical Message

- Juvenile psammomatoid ossifying fibroma (PJOF) is a rare, aggressive fibro-osseous tumor that typically arises in paranasal sinuses or orbits but only infrequently involves the mandible.
- Accurate diagnosis relies on correlating clinical- radiological features with the pathognomonic histological finding of psammoma body–like ossicles.
- Additionally, this case underlines the need for clinicians to be aware of rare entities like psammomatoid JOF when evaluating jaw swellings in young patients, as misdiagnosis or under-treatment can lead to rapid recurrence. Also, an early diagnosis and treatment are essential to prevent undue morbidity.
- The case highlights that mere curettage or partial removal is inadequate. Instead, a more aggressive surgical approach is warranted.

Limitations

A long-term follow-up of the case remains the limitation. Also, a detailed outcome of the definitive complete surgery is beyond the scope of this case report, as the focus here is on the presentation, diagnosis, and pathology of the lesion.

Declarations

- Patient Consent: Written informed consent was obtained from the patient for publication of this case report and accompanying images.
- Conflict of Interest: The authors declare no conflict of interest.
- Funding: None.
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Photomicrographs:

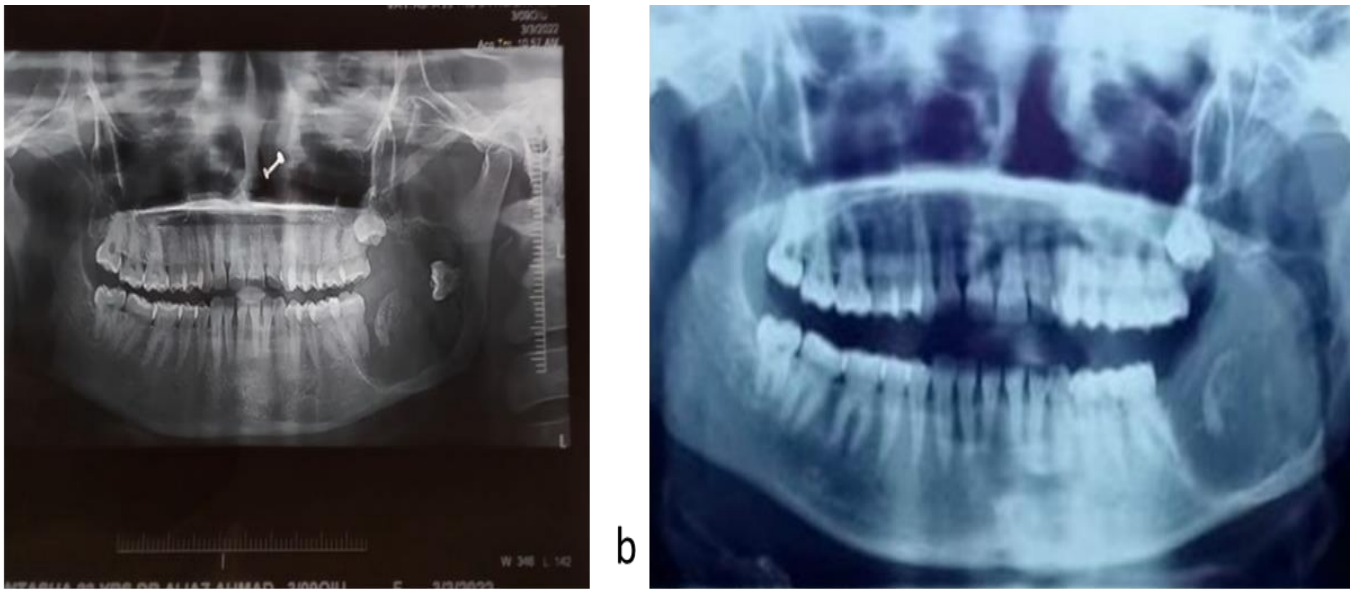


Figure 1 a) Orthopantomogram (OPG) showing a well-defined expansile lesion in the left posterior mandible with mixed radiolucent–radiopaque areas and a "ground-glass" appearance. The lesion causes cortical expansion without cortical breach or periosteal reaction, suggestive of a fibro-osseous lesion, **b)** Orthopantomogram (OPG) obtained one month after the initial biopsy showing recurrence of the lesion in the left mandibular alveolar bone. The lesion demonstrates similar mixed radiolucent–radiopaque changes with cortical expansion, consistent with recurrent psammomatoid juvenile ossifying fibroma.

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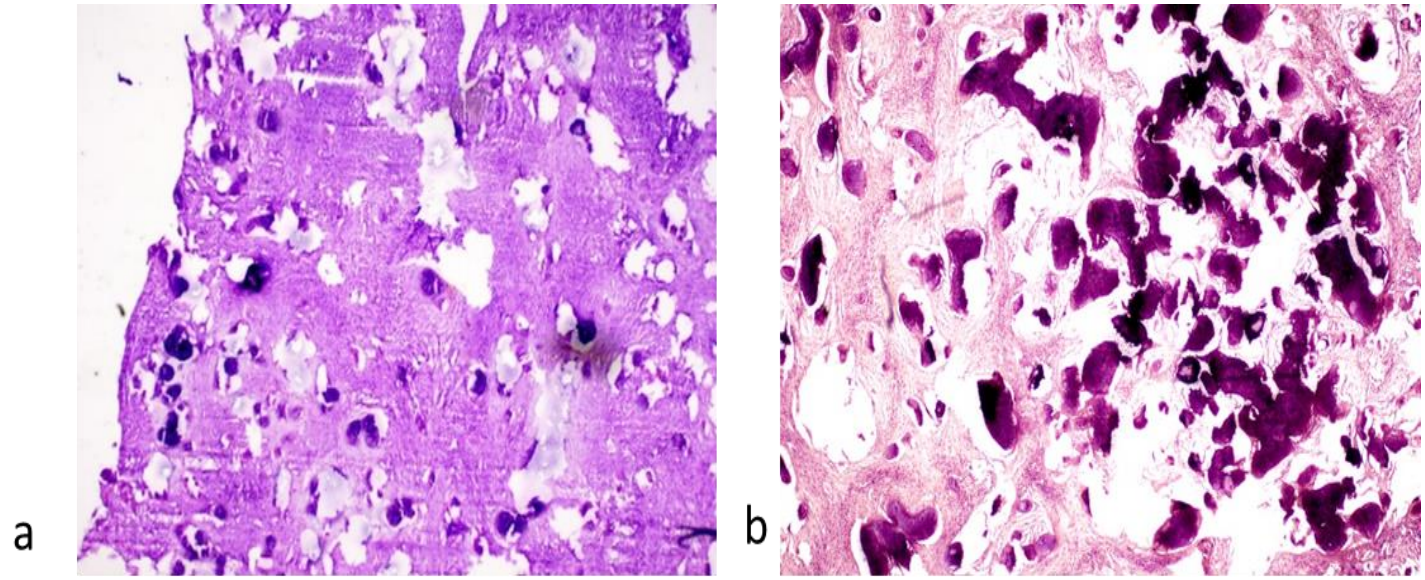


Figure 2 a) Photomicrograph of the lesion (H&E stain, 100×) showing a highly cellular fibrous stroma with concentric calcified ossicles; **b)** Photomicrograph of the lesion (H&E stain, 400×) showing a fibro cellular stroma with numerous well-formed calcified ossicles resembling psammoma bodies, confirming the diagnosis of psammomatoid juvenile ossifying fibroma.