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*Case Series*

# JUVENILE OSSIFYING FIBROMA OF THE MANDIBLE A DIAGNOSTIC DILEMMA

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**ABSTRACT**

Juvenile ossifying fibroma (JOF) is classified as a benign fibro-osseous neoplasm characterized by the proliferation of mineralized osteo-fibrous tissue replacing normal bone. JOF are benign yet exhibit locally aggressive behavior with a strong tendency for recurrence. Juvenile ossifying fibroma is predominantly reported in the maxillary region, paranasal sinuses, orbit, and other bones related to paranasal sinuses. JOF is relatively rare in the mandible. In this current case series, we intend to report two cases of JOF in a child's mandible, diagnosis, surgical management, and follow-up. Pediatric dentists should know these entities of maxillofacial pathology so that appropriate diagnosis can be made and prompt treatment can be delivered.

**KEYWORDS**: *ossifying fibroma, fibro-osseous neoplasm, neoplasm*

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## INTRODUCTION

Ossifying fibroma is a benign, fibro-osseous lesion with a similar presentation to Fibrous dysplasia (FD) but differs in the aspect that it is encapsulated; hence, it is more circumscribed and reported in third and fourth decade (1-3).

Juvenile ossifying fibroma (JOF) is a variant of the ossifying fibroma reported in younger children. Even though JOF is benign, it tends to exhibit locally aggressive behavior due to expansion and compression of adjacent tissue (4-8).

Juvenile trabecular ossifying fibroma (JTOF) and Juvenile Psammomatoid ossifying fibroma (JPOF) are two histological variants described in the literature (5). The psammmomatoid variant often presents as a painless growing mass, leading to asymmetry and compression of surrounding tissue (6). Radiographically, JPOF presents itself as an expansile radiographic lesion exhibiting varying degrees of calcification internally (7, 8).

Histopathologic appearance demonstrates a grainy texture that represents calcification and is often called Psammomatoid. Recurrence rates of JPOF vary from 10% to 19% based on the management protocol followed. Recurrence rate is higher when conservative treatment protocol is followed. Clinically, patients with JTOF are often asymptomatic, and early lesions are usually discovered as incidental radiographic findings. Displacement of teeth may be an early sign of this tumor process. JTOF may be aggressive in its growth potential, and as it matures, rapid growth may result in facial asymmetry and jaw deformity (5).

Radiographically, JTOF are usually unilateral, unilocular mixed radiolucent/radiopaque lesions (9). Computer tomographic imaging is required for larger lesions to determine the full extent of the lesion. JTOF tends to expand concentrically from a central point or epicenter outward in all directions, and this expansion may result in the displacement of teeth and the inferior alveolar nerve canal (10). The outer cortical plate remains intact despite significant expansion and thinning. Resorption of teeth is common, and JTOF maintains a well-defined corticated border (10). The primary histologic criterion for JTOF consists of a neoplasm predominantly composed of cellular fibroblastic tissue with thin trabeculae of immature bone, which may anastomose to form a lattice (11).

JTOF is usually well-demarcated but unencapsulated. Of note, there may be considerable variation in stromal cellularity. Plump osteoblastic rimming of bone is a standard feature. Clusters of osteoclastic multi-nucleated giant cells, areas of hemorrhage, and foci of pseudo cystic stromal degeneration may be observed (12). As the lesion proliferates, more aggressive behavior of JOF can be confirmed, usually due to incomplete initial removal or persistence of local irritants. It is seen that incomplete resection causes recurrence in aggressive tumors, and recurrence rates of 30% to 58% have been reported for juvenile trabecular ossifying fibromas.

The World Health Organization (WHO) classifies juvenile ossifying fibroma into two histologic variants: JTOF and JPOF (13). The former is usually seen in children and adolescents (mean age of presentation: 8.5-12 years), whereas the latter usually affects a more comprehensive patient age range (16–33 years). There is no gender preference for either entity. Both entities are relatively rare; however, JPOF is more commonly encountered than JTOF (14).

Localization differs for each subtype, with the maxilla being more common for JTOF and the paranasal sinuses more common for JPOF (15, 16). Here, we describe a case series of two cases with a trabecular variant in the mandible of two young children.

## CASE REPORT AND RESULTS

*Case 1*

A 13-year-old female patient reported to the Department of Oral and Maxillofacial Surgery with a hard swelling on the left side of their face for 3 months. Upon clinical examination, diffuse swelling was on the left lower third of the face. Intraoral examination revealed bony hard expansion of the buccal and lingual cortical plates in the lower left premolar-molar region. The swelling was well-defined, measuring 4 × 3 centimeters and extending from the lower left lateral incisor to the first molar. The mucosa over the swelling looks normal in color and consistency, with no tenderness or visible pulsations (Fig. 1).

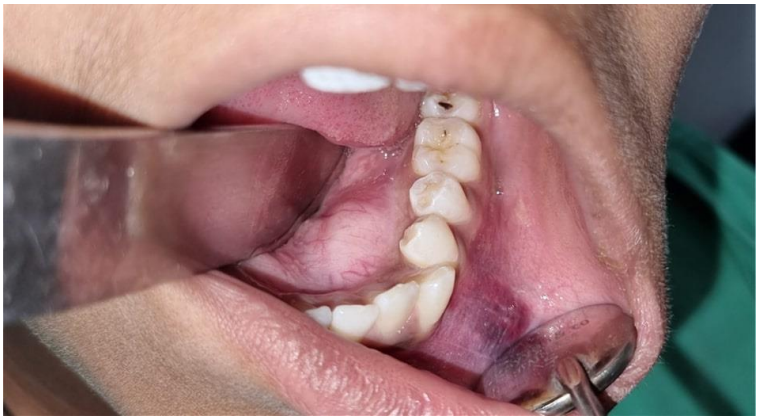

**Fig. 1.** *Clinical intra-oral picture of the expansile swelling.*

No draining sinuses were observed. Tooth mobility was present for the first and second premolars. Teeth vitality tests revealed no response for the first and second premolar and delayed response for the first molar. There was no complaint of paraesthesia or regional lymphadenopathy. Radiographic investigations and orthopantomography revealed a solitary, well-defined irregular radiolucency extending from the central incisor to the mesial root of the second molar. There was the displacement of the canine, first and second premolar, and resorption of the first, second premolar, and mesial root of the first molar. CBCT shows expansion and thinning of buccal and lingual cortical plates measuring 28.43mm buccolingually, 40.9mm anteroposteriorly, and 28.95 superior-inferiorly (Fig. 2).

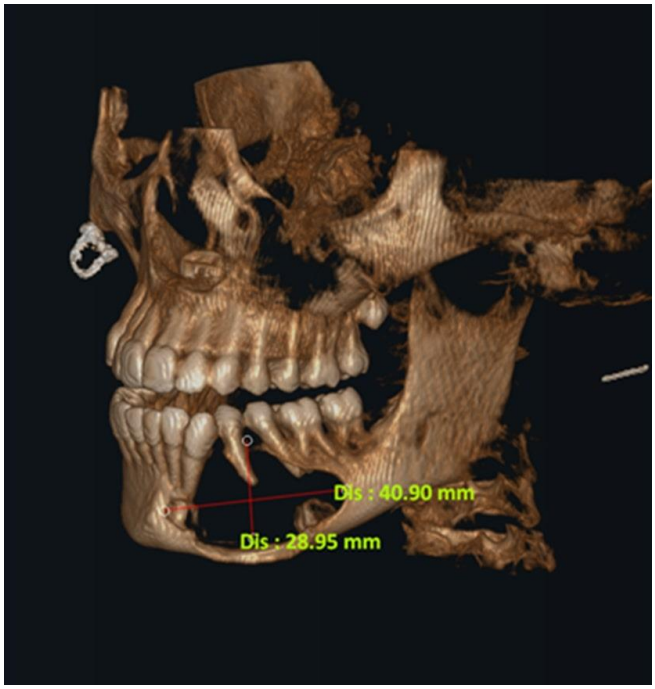


**Fig. 2**. *Pre-operative CBCT image of the lesion.*

The aspiration of the site was negative. Therefore, the cyst diagnosis was ruled out, and an incisional biopsy was done and sent for histopathological evaluation. Section revealed fibrous connective tissue with scattered bony trabeculae and multinucleated giant cells, confirming the diagnosis of JOF. Intentional root canal treatment for the central incisor, lateral incisor, canine, and first molar, followed by surgical enucleation and curettage of the tumor, was planned under general anesthesia.

*Surgical management*

Under strict aseptic conditions, general anesthesia was administered under naso-endotracheal intubation. First and second premolars were extracted, and enucleation was done through an intraoral crevicular incision extending from the lower right canine to the left second molar (Fig. 3).

Enucleation of the entire lesion and peripheral ostectomy of 1cm bone was done, followed by reinforcement with a titanium reconstruction plate and primary closure with 3-0 synthetic resorbable sutures (Fig. 4).

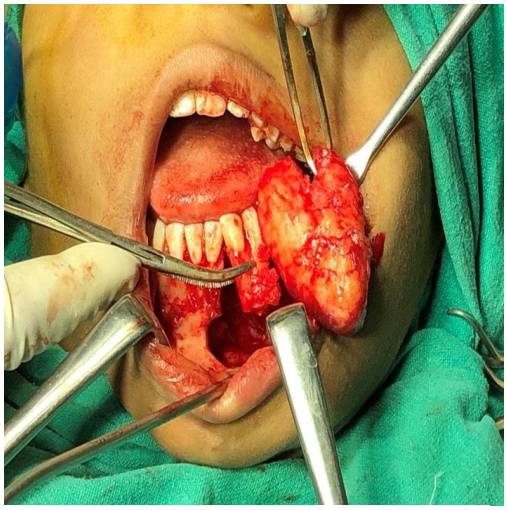

**Fig. 3**. *Surgical resection of the lesion.*

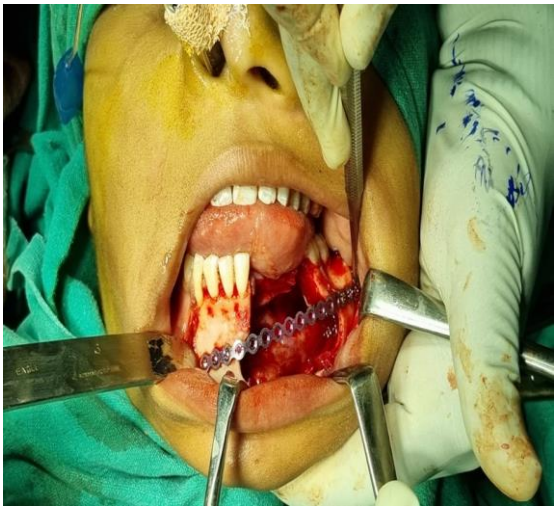

**Fig. 4**. *Post-resection stabilisation of the defect with a titanium mesh.*

The surgery was uneventful. The histopathological report confirmed the diagnosis of a trabecular variant of JOF. Postoperatively, the patient was put on Cefixime 1gm I.V. 12th hour, Metronidazole 500mg I.V. every 8th hour, and Diclofenac 75mg every 12th hour for 5 days. The healing was satisfactory. The patient has been followed up for 6 months. There are no complaints of swelling or paraesthesia, and gradual bone remodeling has occurred. Follow-up OPG showed reduced radiolucency and good amounts of bone regeneration (Fig. 5).

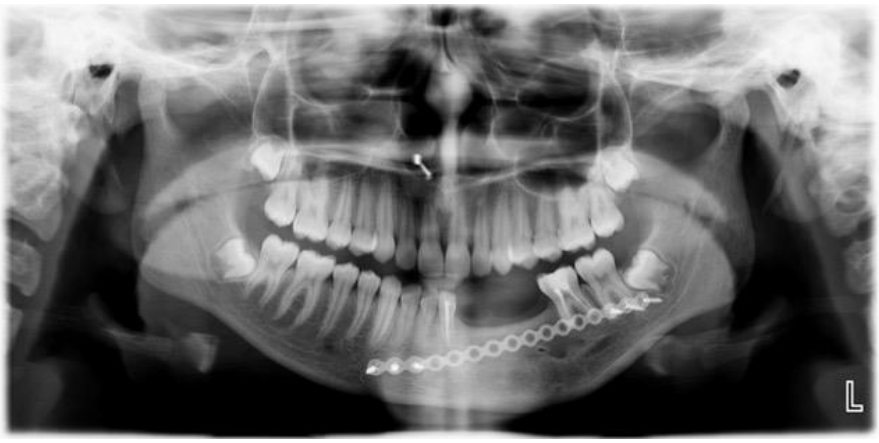

**Fig. 5**. *18 months follow-up visit.*

*Case 2*

A 5-year-old child reported to the Department of Oral and Maxillofacial Surgery with diffuse swelling in the left lower jaw for 10 months. The growth was rapidly increasing in size. On examination, an oval-shaped swelling measuring around 4x2 cm was seen. The swelling was well-defined; swelling extends from the deciduous canine to the second molar, causing expansion of buccal and lingual cortical plate expansion and obliteration of buccal vestibule concerning tooth numbers #74 and #75. The overlying attached gingiva and mucosa are normal. On palpation, the swelling was painless, hard in consistency, and without any pulsations or discharge. There was no paraesthesia over the lower lip or chin. Lymph

nodes were nonpalpable. Orthopantomography revealed a solitary, well-defined radiolucency surrounded by a radiopaque border (Fig. 6).

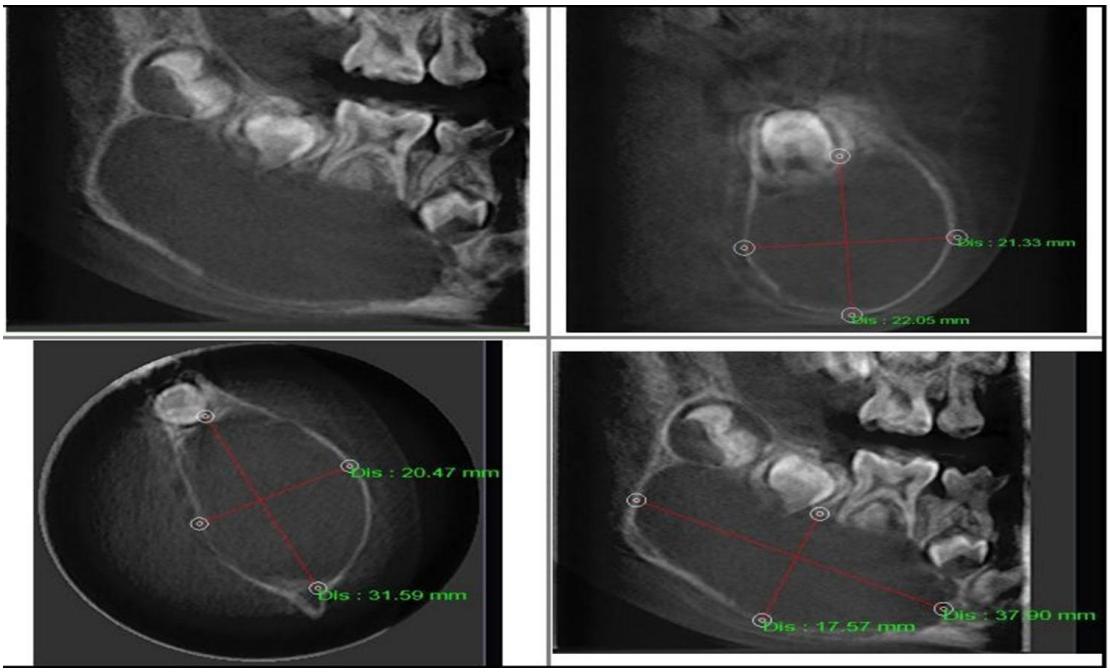


**Fig. 6**. *Pre-operative OPG of the lesion.*

Cone beam computed tomography showed expansion and thinning of buccal and lingual cortical plates measuring 21.33mm buccolingually, 37.9 mm anterior-posteriorly, and 17.52 mm superior-inferiorly. The inferior border was intact. The aspiration of the site was negative. An incisional biopsy was done and sent for histopathological evaluation. Section revealed fibroblastic stroma admixed with psammoma bodies.

Focal areas show mature bony trabeculae surrounded by fibrous stroma, confirming the diagnosis of JPOF. Considering factors like an intact inferior border, no involvement of adjacent structures, no paraesthesia of the lip, and the age of the patient, this case was planned for excision and curettage without resection.

*Surgical management*

The patient was operated under general anesthesia under naso-endotracheal intubation. The intraoral crevicular incision was given from the distal aspect of the deciduous canine to the first molar. Enucleation of the entire lesion along with peripheral ostectomy was done and sent for biopsy, followed by reinforcement with a titanium reconstruction plate, and primary closure was done with synthetic resorbable sutures (Fig. 7, 8).

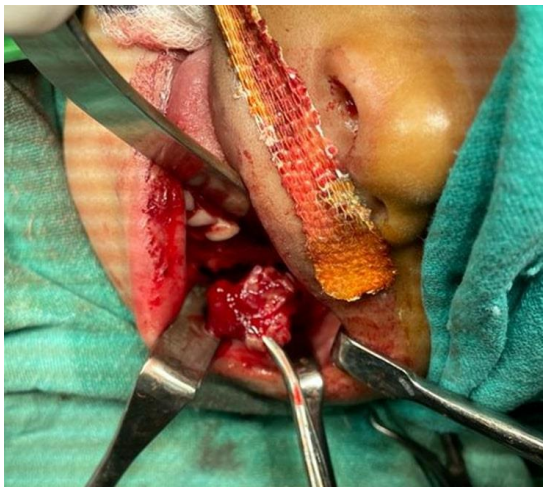

**Fig. 7**. *Surgical resection of the lesion.*

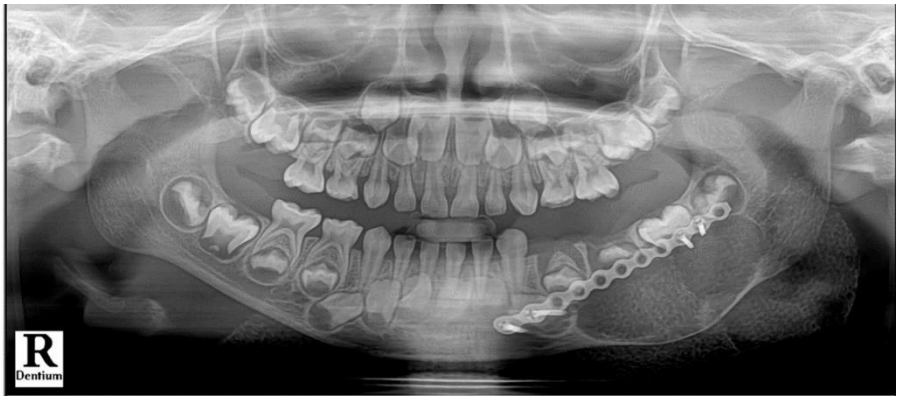


**Fig. 8**. *Post resection stabilization of the defect with a titanium mesh.*

The surgery was uneventful. The histopathological report confirmed the diagnosis of JPOF. A post-operative follow-up visit after 12 months showed a filling up of bone defect (Fig. 9).

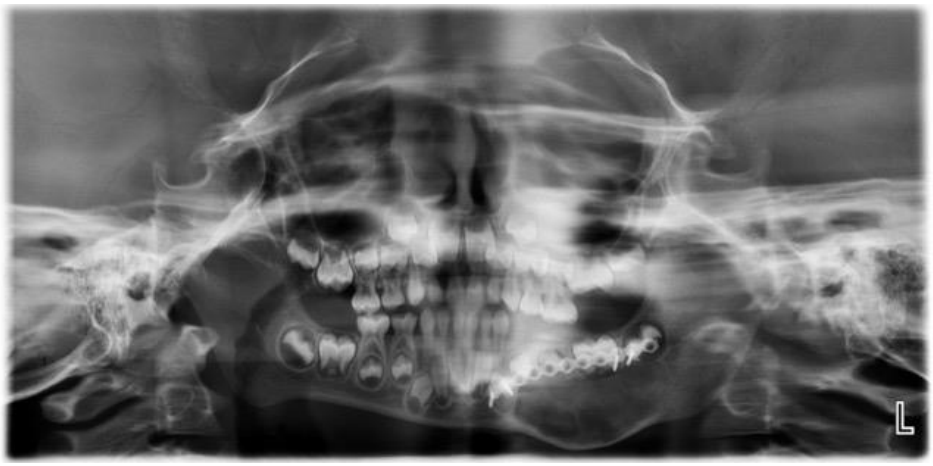


**Fig. 9.** *12-month follow-up visit.*

## DISCUSSION

Previous literature has published multiple case reports on JTOF and JPOF (Table I). Most of the jaw's fibro-osseous lesions are benign, asymptomatic, and grow slowly. Ossifying fibromas fall within the group of benign fibro-osseous lesions. As previously believed, they are not of odontogenic origin but instead made of osteogenic calcified matrix.

**Table I**. *The table shows a list of previous case reports.*

| Sno | Author | Type | Age/Gender | Variant | Location | Management |
|---|---|---|---|---|---|---|
| 1 | Adham et al 2023 (17). | Case report | 12/Male | Psammomatoid | Maxilla:<br>Left nasal bone, left paranasal sinus, left nasal cavity, nasal septum, and pushing the medial wall of the left eye. | Subtotal maxillectomy and reconstruction in one stage. Reconstruction surgery was made with titanium mesh and rib cartilage. |

| | | | | | | |
|---|---|---|---|---|---|---|
| 2 | Kim et al. 2023 (18) | Case report | 4/Male | Psammomatoid | Zygomaticomaxillary area on the left side | Excision |
| 3 | Nnko et al 2022 (19) | Case report | 8/ Female | Trabecular | Right maxillary area | Hemi maxillectomy and reconstruction of maxilla with a rib |
| 4 | Arfaj et al. 2022 (20) | Case report | 15/Female | | Aggressive juvenile active ossifying fibroma of the ethmoid sinus with orbital and intracranial extension | endoscopic debulking of the lesion followed by lateral rhinotomy, and finally, frontal craniotomy with reconstruction |
| 5. | Gotmare et al 2017 (21) | Case report | 8/Male | Psammomatoid | Expansile lesion in the left ramus of the mandible. | enucleation and curettage with standard hemi-mandibulectomy along with the fixation of construction plate. |
| 6. | Sarode et al 2018 (22) | Case report | 10/Male | Psammomatoid | Anteriorly up to the right canine region, posteriorly up to the left side zygomatic bone, superiorly up to left infraorbital margin and inferiorly up to the alveolus | Excision with palatal prosthesis |
| 7 | Titinchi et al 2021 (23). | Retrospective study | 3-31 years | Trabecular-10 Psammomoid-7 | Different regions in the mandible and maxilla. | Enucleation for small well defined lesions in mandible Curettage with peripheral ostectomy for medium to large neoplasms Resection with reconstruction for large infiltrative recurrent neoplasms |
| 8 | Acosta et al 2023 (24) | Case Report | 14/Male | | Lesion involving the left nasal cavity, left maxillary ethmoid and sphenoid sinus, and left side of the nasopharynx | Multidisciplinary assessment and care were done by NS and ENT to debulk the base of the tumor via transnasal and sublabial access endoscopically |
| 9 | JamesI J Green et al 2023 (25) | Case Report | 4/Male | Trabecular | Entire maxilla | Maxillary resection |
| 10 | Saad et al 2019 (26) | Case Report | 9/Female | Trabecular | Right impacted molar area to the left first molar area causing perforations in the buccal and lingual cortical plates of mandible | Surgical excision |
| 11 | Sultan et al | Case Report | 8/Male | Trabecular | Right mandibular | The tumor was |

| | | | | | | |
|---|---|---|---|---|---|---|
| | 2018 (27) | | | | ramus. | enucleated, an ostectomy was performed, and the bone cavity heat-treated with electrosurgical coagulation |
| 12 | Aslan et al 2018 (28) | Case Report | 13/Female | Psammomatoid | Maxillary sinus | Complete surgical resection |
| 13 | Solyman et al 2020 (29) | Case Report | 4/Male | Psammomatoid | Left maxillary sinus and encroachment of left orbit | Debulking surgeries |
| 14 | Mouna Lyoubi et al 2021 (30) | Case Report | 14/Female | Psammomatoid | polylobed mass filling the right nasal cavity, | Endoscopic transnasal approach with image-guided neuro-navigation system and complete surgical removal |
| 15 | Ashwan Paranthaman et al 2017 (31) | Case Report | 13/Female | Trabecular | right side of the face, extending from right alar region to the infraorbital margin superiorly, and extending laterally to the right cheek region | Maxillary resection |

Since they refer to the same thing, cementifying and cemento-ossifying fibroma are no longer used. Their epidemiology is poorly understood because they have long been mistaken for cemento-osseous dysplasia. Their distribution is primarily restricted to the craniofacial bones.

Ossifying fibroma is a rare fibro-osseous tumor composed of fibrous tissue and mature bone. It is subdivided into conventional and juvenile subtypes. JOF is an uncommon variant of ossifying fibroma that behaves in a more aggressive pattern than the conventional type (32).

Adults older than 30 to 40 years are typically affected with conventional form in their jaw. In 80% of cases, JOF develops in children and young adults before age 15. The JOF has been identified as a distinct histopathological entity among the fibro-osseous group of disorders because of its distinctive histological characteristics. Both variants exhibit distinct histopathological patterns, and hence, the diagnosis is more predictable. Age is an important factor in the diagnostic criteria of juvenile ossifying fibroma.

The clinical presentation of JOF is a slow-growing, asymptomatic, expansile, spherical or ovoid swelling, causing significant facial asymmetry, and depending upon the anatomical site of involvement, there can be nasal obstruction, epistaxis, exophthalmos, and proptosis. In most patients (85%), the tumors are located in the facial bones, but they also involve the calvaria (12%) and extracranial sites (4%). In the facial bones, 90% of the tumors arise from the

maxilla and paranasal sinuses, and the remaining 10% arise from the mandible (33). Rarely, pain and paraesthesia associated with the swelling are present. Root resorption and displacement of the involved teeth are observed. In the mandible, the angle and ramus are the most common sites of involvement (34).

Juvenile ossifying fibroma can be radiographically divided into 3 stages. Initial stage/Stage I: it appears as a well-defined radiolucent lesion. Mixed-stage/Stage II: radiolucent lesion with areas of internal calcification. Mature stage/Stage III-Completely radio-opaque mass. On CT, it appears as a well-demarcated lesion with a sclerotic rim and less dense fibrous central core. Three patterns have been described in CT. Pattern-1: Radiolucent central core with a thick outer mantle. Pattern-2: Ground glass mural nodule. Pattern-3: Homogeneous solid radio dense lesion (37).

Histologically, JOF is characterized by a cellular fibrous stroma, garland-like bony strands, and cement particles. A recent study by El-Mofty identified two histopathological variants: JTOF and Psammomatoid JOF (PJOF). The average age of occurrence of JTOF is considerably younger than JPOF, with the average being eight-and-a-half to twelve years compared to a 16- to 33-year range of PJOF (34).

## CONCLUSIONS

Complete surgical excision is preferred to conservative curettage in both juvenile ossifying fibroma. Long-term follow-up is necessary because incomplete resection increases the risk of recurrence, which is more aggressive than primary and occurs 6 months to 19 years after surgery. For complete resection, an open surgical rather than an endoscopic approach may be required, depending on the degree of disease and invasion. Due to the tumor's radioresistance, high risk of malignant transformation, and late radiotherapy-related adverse effects in children, radiotherapy is not recommended. Juvenile ossifying fibroma should be managed as a locally aggressive neoplasm due to its high recurrence rate (30-50%) and aggressive nature.

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