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A DIAGNOSTIC DILEMMA OF JUVENILE OSSIFYING FIBROMA IN A PEDIATRIC PATIENT – A CASE REPORT

ABSTRACT

Introduction: Juvenile ossifying fibromas(JOF) is an uncommon benign fibro-osseous tumor affecting children under 15 years of age. The aggressive nature added to their high tendency to recur, pose real diagnostic and therapeutic challenges for the dental practitioner and makeslong term postoperative follow-up over the years indispensable[1,2].

Case report: This case report describes a seven-year-old girl who presented with a swelling on the left mandibular region present for over five months. Clinical, radiographic, and histopathological findings confirmed the diagnosis of trabecular juvenile ossifying fibroma (JTOF) and was planned for wide surgical excision.

Discussion: JOF is recognized as a distinct variant of ossifying fibroma and includes two histopathological subtypes: trabecular and psammomatoid. The aggressive nature, potential for recurrence and resemblance to malignant bone tumors often complicate diagnosis and management. Early recognition, precise histopathological evaluationand long-term follow-up are essential to ensure optimal outcome and minimize recurrence[1,3]. This case highlights the importance of distinguishing JOF from other fibro-osseous lesions and underscores the need for vigilant postoperative monitoring in pediatric patients.

Keywords: Juvenile ossifying fibroma,juvenile trabecular ossifying fibroma,juvenilepsammomatoid ossifying fibroma.

INTRODUCTION

The juvenile ossifying fibroma (JOF) is a rare benign tumor and that belongs to the group of fibroosseoustumors[4].Fibro-osseous lesions of the cranial and facial bones are usually benign and tend to grow slowly. Benign fibro-osseous lesions resemble fibrous dysplasia, ossifying fibroma, and cemento-ossifiying dysplasiahistopathologically[5,6].The fibro osseous lesions of the jaws represent a diverse group of entities that are characterized by replacement of normal bone by a fibrous connective tissue matrix with varying amounts of osteoid, immature and mature bone.[7]It is distinguished from other fibro-osseous lesions by factors such as age of onset, clinical presentation, and expected behaviour. The lesion shows aggressive growth and has a high recurrence rate[8]. It most commonly affects children between 5 and 15 years of age and can result in significant facial disfigurement.[9]

Benjamin (1938) first described JOF as an ossifying fibroma with atypical calcification, and the termjuvenile aggressive ossifying fibroma was later introduced by Johnson et al(1952). These lesions account for about 2% of oral tumours in children[10]. JOF is further classified into two types: juvenile psammomatoid ossifying fibroma (JPOF) and juvenile trabecular ossifying fibroma (JTOF). The psammomatoid type commonly affects the bones of the orbit and paranasal sinuses, whereas the trabecular type more often involves the jaws[11].

Juvenile ossifying fibroma typically presents at an early age, with 79% of cases diagnosed before 15 years.[6,12,10] It affects males and females equally and accounts for approximately 2%

of oral tumours in children.[13] JOF is thought to originate from the periodontal ligament.[14] Clinically, the tumour exhibits aggressive behaviour and a considerably higher recurrence rate compared with other fibro-osseous lesions.[15] It is usually localized and well-demarcated, though not encapsulated.[16] Because of its aggressive growth and high recurrence potential, early diagnosis and complete surgical excision are crucial.

Case report

A 7-year-old female patient presented to the Department ofPedodontics, Government Dental College, with a chief complaint of swelling on the lower left side of the face involving the mandible, which had been present for the past five months. She had previously takenmultiple courses of antibiotics prescribed by the Department of Pediatrics, Government Medical College, Thiruvananthapuram, under a provisional diagnosis of bacterial sialadenitis. Her medical and family histories were non-contributory.

On clinical examination, a solitary, well-defined, firm mass was observed extending from the body to the angle of the left hemimandible, producing mild facial asymmetry on the affected side. The lesion caused expansion of both the buccal and lingual cortical plates, resulting in obliteration of the left buccal vestibule in the canine–molar region.The overlying mucosa appeared stretched but intact. On palpation, the swelling was firm with minimal tenderness, and no regional lymphadenopathy was noted. Mouth opening was within normal limits. Intraoral examination revealed that the patient was in the mixed dentition stage, with no evidence of dental caries, tooth mobility, or pathological tooth displacement.

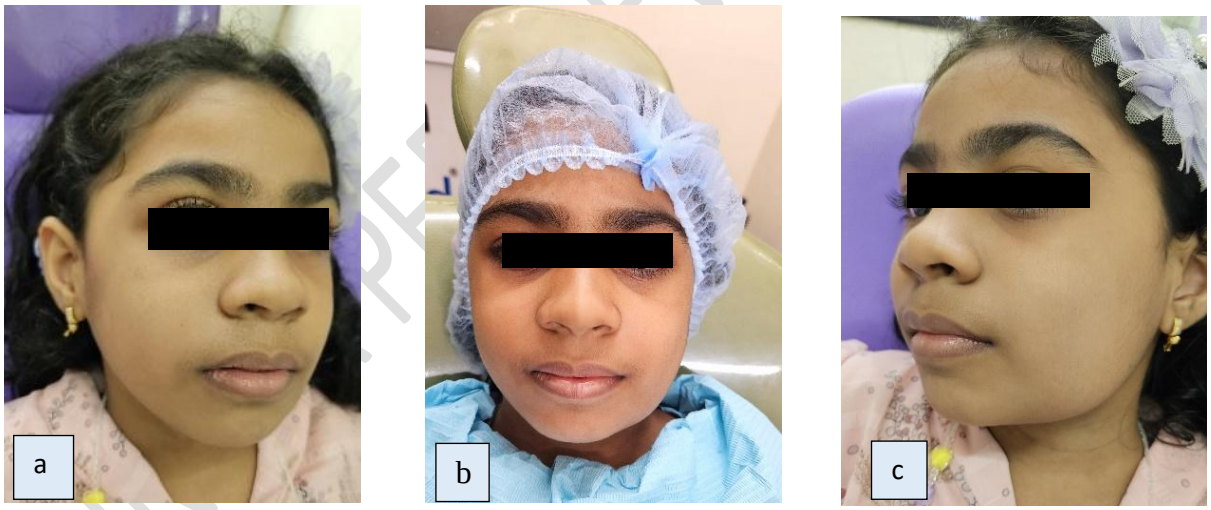


Figure1:(a) Lateral view of the normal right side of mandible.(b) Frontal view shows the facial asymmetry.(c) Lateral view of the extraoral swelling in the left mandibular region

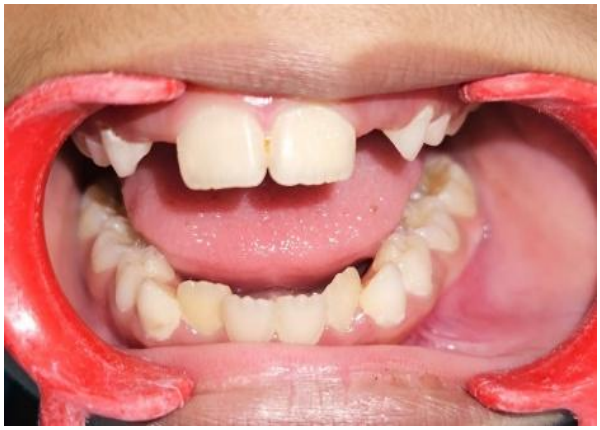


Figure 2: Intraoral view showing obliteration of the lower left buccal vestibule from the 73 to 36 region

Radiographic investigations included intraoral periapical radiography (IOPAR), orthopantomography (OPG), topographic views, and cone-beam computed tomography (CBCT). The IOPAR and OPG findings were within normal limits, whereas the topographic radiograph showed periosteal new bone formation. CBCT revealed a mixed radiolucent–radiopaque lesion with an altered trabecular pattern and a characteristic ground-glass appearance involving the left body, angle, and ramus of the mandible. Multiple small radiolucent areas with irregular margins were present within the lesion, suggesting a chronic inflammatory process or primary chronic osteomyelitis in the affected region. Anteriorly, the lesion extended from the distal aspect of tooth 75, while posteriorly it involved the entire ramus of the mandible, extending posterosuperiorly to the condylar and coronoid processes. Routine hematological investigations were unremarkable, except for an elevated erythrocyte sedimentation rate (ESR).

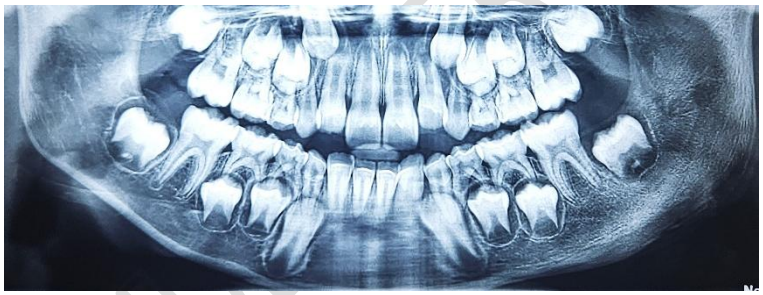


Figure3: Orthopantomograph showing mixed radiolucent–radiopaque lesion in the left second mandibular premolar– ramus region.

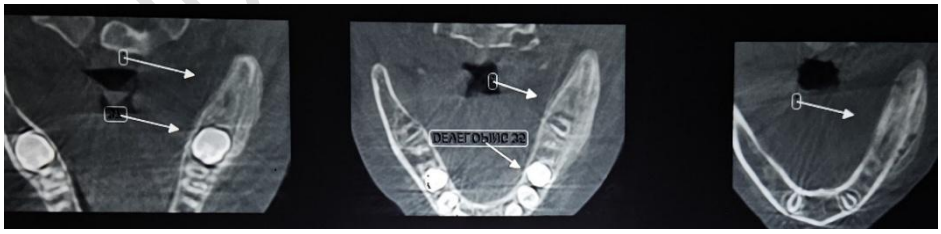


Figure4: Axial section CBCT view showing expansion of both the buccal and lingual cortical plates

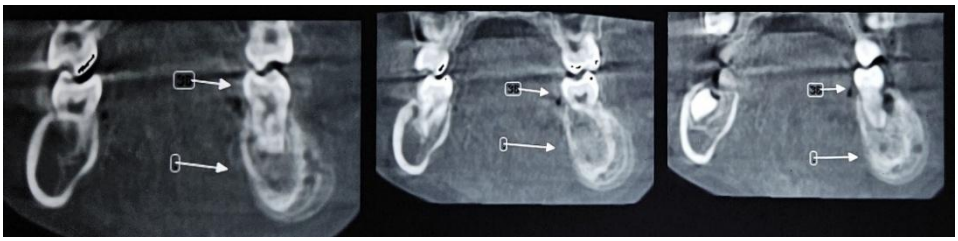


Figure 5 :coronal section CBCTview shows thickening of cortical plate

An incisional biopsy was performed under local anesthesia (2% lignocaine with adrenaline). A vestibular incision was made extending from the canine to the molar region, followed by reflection of a subperiosteal flap. A bony specimen was obtained using a chisel and mallet and fixed in 10% neutral buffered formalin for histopathological examination. Hematoxylin and eosin (H&E)–stained sections showed trabeculae of fibrillary osteoid and woven bone fragments embedded within a highly cellular, storiform stroma composed of spindle-shaped and polyhedral cells with minimal collagen production. The osteoid formations appeared as characteristic paint-brush–like strokes. Based on the correlation of clinical, radiographic, and histopathological findings, a final diagnosis of juvenile trabecular ossifying fibroma was made. The patient was subsequently scheduled for wide surgical resection of the affected mandibular region under general anesthesia.

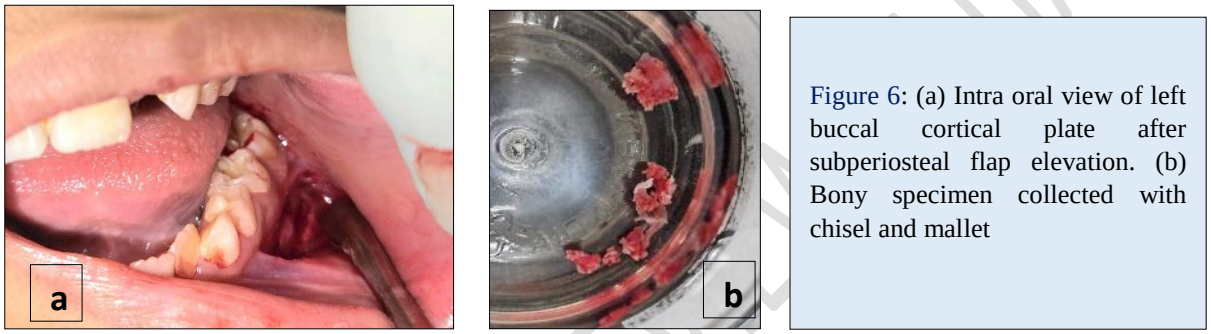
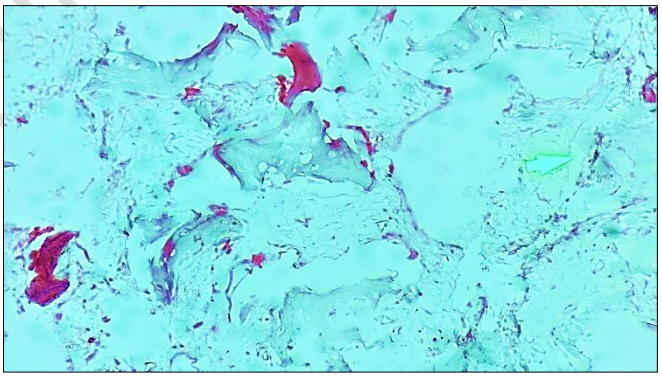


Figure 6: (a) Intra oral view of left buccal cortical plate after subperiosteal flap elevation. (b) Bony specimen collected with chisel and mallet

Figure 7: H&E stained serial sections of cellular stroma which is delicately collagenous & numerous trabeculae of bone with osteocytes and some of the trabeculae show osteoblastic rimming. Scanty inflammatory infiltrate is present.



DISCUSSION

Benign fibro-osseous lesions of the head and neck region are uncommon and include several entities with overlapping clinical and radiographic features. These lesions comprise fibrous dysplasia (FD), ossifying fibroma (OF), and cemento-osseous dysplasia (COD).[17] According to the World Health Organization (WHO) classification of odontogenic tumors (2005), OF can be categorized into conventional ossifying fibroma and juvenile ossifying fibroma (JOF).[11] JOF is a rare neoplasm characterized by replacement of the normal bone matrix in children. It is typically observed in individuals under 15 years of age, with a slightly higher incidence reported in males. These tumors are generally large and expansile, frequently extending into the ethmoid and sphenoid sinuses, nasal cavity, orbital walls, and maxillary

bone, although mandibular lesions are also documented. JOF is further subdivided into juvenile psammomatoid ossifying fibroma (JPOF) and juvenile trabecular ossifying fibroma (JTOF).[11] Both variants occur at considerably younger ages compared with other fibro-osseous lesions. The trabecular type tends to present in individuals aged 8.5–12 years, whereas the psammomatoid variant appears at a slightly older mean age of 16–33 years.[18] The psammomatoid variant is more common and more aggressive, demonstrating a higher recurrence rate than the trabecular form.[19] In its trabecular form, JOF may resemble osteosarcoma in terms of aggressiveness.[20] In the present case, despite the lesion's relatively slow progression and the intact appearance of the mucosa, its underlying pathology was significant.

The hallmark characteristics of JOF include early age of onset, localized tumor growth, distinct clinical presentation, aggressive behavior, and a high recurrence potential.[11,13,9] Clinically, JOF may present as an asymptomatic, slowly or rapidly expanding bony swelling that results in facial asymmetry. The tumor may attain considerable size and often demonstrates aggressive features such as rapid enlargement, cortical thinning, cortical perforation, and invasion of adjacent anatomical structures.[10,8] Reported symptoms vary and may include facial swelling, a progressively enlarging hard mass, sinusitis, nasal obstruction, tooth displacement, root resorption, cortical perforation, ocular proptosis, and epistaxis. Pain and paraesthesia are uncommon. The tumor may erode bone partitions and invade adjacent orbital, nasal, and cranial compartments, resulting in facial deformity, displacement of orbital structures, and obstruction of sinus drainage.[21]

JOF is believed to arise from the differentiation of mesenchymal cells of the periodontal ligament or multipotent precursor cells that form fibrous tissue, cementum, or osteoid. Controversy exists regarding its predilection site; while some studies suggest the maxilla is more frequently involved, others report a higher incidence in the mandible.[9,22] Johnson et al. reported a higher incidence in females, whereas Bertrand et al. noted equal distribution between genders.[20,13]

Radiographically, JOF may appear as a unilocular or multilocular radiolucency, or as a mixed radiolucent–radiopaque lesion with well-defined borders. Root displacement and resorption may occur but are uncommon.[22] Most lesions are well-defined and demonstrate mixed radiodensity, as also reported by Chrcanovic and Gomez.[23] Although JOF is not encapsulated, it is usually separated from surrounding bone by a radiopaque border—an important feature distinguishing it from fibrous dysplasia. It typically exhibits a concentric or centrifugal pattern of growth and may show a characteristic "ground-glass" appearance on radiographs.

Histologically, the present case demonstrated classic features of trabecular JOF (TrJOF), including spindle-shaped fibroblastic cells arranged in a whorled pattern, osteoid trabeculae, and multinucleated giant cells. These findings align with those described by Slootweg and Panders,[10] who emphasized the highly cellular nature of JOF, with abundant osteoid and the presence of multinucleated giant cells. These cellular characteristics are common to both variants; however, they are more pronounced in the trabecular type, where the stroma is densely packed with fibroblasts arranged in a storiform pattern.[3] Additionally, the presence of psammomatous calcifications supports the diagnosis of PsJOF when present, as these structures are considered pathognomonic for the psammomatoid variant, as described by Eversole et al.[24]

Regarding treatment, Slootweg and Müller[10] reported no significant differences in outcomes between limited surgical procedures and more extensive surgeries, whereas Waldron et al.[25] advocated local excision and curettage as preferable treatment options, also recommending local excision for recurrent cases. Incomplete resection is associated with recurrence, particularly in aggressive tumors. Therefore, some authors recommend en bloc resection as the most adequate treatment. Curettage combined with peripheral osteotomy, or in some cases segmental mandibular resection with reconstruction, is suggested for extensive or recurrent lesions. Long-term recurrence may lead to sarcomatous transformation.[26],[3]

It is widely accepted that JOF behaves as a locally aggressive lesion with a high recurrence rate when inadequately treated. The recommended management is en bloc resection with free surgical margins[15],[3] Radiotherapy is contraindicated, and a "wait-and-see" approach is generally not advised.[27],[10] Marginal resection is recommended for large lesions with cortical perforation or severe cortical thinning. Total resection or partial mandibulectomy is reserved for cases in which the lower border of the mandible cannot be adequately identified.[27] Nonetheless, for both trabecular and psammomatoid variants, conservative surgical excision remains an acceptable treatment approach, despite reports of multiple recurrences. The extent of surgical management should be tailored to the patient's age, tumor location, and involvement of adjacent vital structures.[28]

CONCLUSION

The aggressive nature and rapid growth of juvenile ossifying fibroma necessitate early diagnosis, careful histopathological evaluation, and comprehensive management with long-term follow-up due to its high recurrence rate. This case highlights the importance of identifying the trabecular variant of juvenile ossifying fibroma, which presented in the mandible of a female patient in the mixed dentition stage. It also emphasizes the value of conservative treatment to preserve the developing permanent tooth germs, in contrast to more aggressive surgical approaches.

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