

Trabecular Variant: A Rare Entity of Juvenile Ossifying Fibroma of the Mandible

Abstract

One of the rarest entities of fibro-osseous lesions that arise within the craniofacial bones is Juvenile ossifying fibroma (JOF). It is an intraosseous expansile lesion of the jaw that imitate odontogenic lesions. WHO has described two distinct histopathological variants of JOF; trabecular and psammomatoid. Histologically, they are characterized by the presence of fibrous connective tissue stroma along with osteoblastic and osteoclastic cells. Clinical, characteristics show an early age of onset, typical histological patterns, high rate of aggressive behavior and recurrence. This article presents a rare clinical case of the trabecular variant of JOF, its clinical, radiological, histological, and treatment aspects.

Keywords: Aggressive lesion, mandible, trabecular variant

Introduction

Juvenile ossifying fibroma (JOF) is an actively growing aggressive lesion containing cells rich in the fibrous stroma containing cellular osteoid, trabeculae of woven bone without osteoblastic lining, with or without Clusters of giant cells. It is nonencapsulated and well demarcated from surrounding bone (WHO).^[1] In 2002 El-Mofty first identified the two histopathological variants of JOF: Trabecular and psammomatoid.^[2] Rare cases of mandibular JOF have been reported.^[3] Clinical examination reveals it as asymptomatic slow growing swelling causing facial asymmetry, extending to a substantial size and behaving as an aggressive lesion.^[4] Pain and paraesthesia are less commonly noted. Depending on the site of involvement, exophthalmos, and epistaxis is sometimes observed. It has higher male predominance with the mean age of occurrence ranging from 2 to 15 years.^[4] The radiographic features show unilocular or multilocular radiolucency with well-delineated borders and few opacification in the center.^[4] The suggestive features of the aggressive variant are the presence of the perforation, cortical thinning, tooth displacement, and root resorption. The advanced imaging studies show more invasive and destructive features apart from conventional radiographic features.^[4] Histopathologically both the entities are nonencapsulated, well demarcated from the surrounding bone

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consisting of neoplastic cellular stroma formed by spindle shape fibroblast.^[5] The trabecular variant shows irregular strands of cellular osteoid and irregular osteocytes. The osteoblast encloses the focal areas of the multinucleated giant cell. In contrast, the psammomatoid pattern forms concentrically laminated ossicles that vary in shape and having peripheral eosinophilic osteoid rims with basophilic centers typically.^[5] The overall clinical features, histopathological features, and advanced imaging were indispensable to provide the diagnosis as JOF. A nonaggressive form of this lesion is managed by curettage and local excision whereas aggressive form is managed by complete surgical excision, *en-bloc* resection or hemimaxillectomy to prevent recurrences.^[6] Reported here is a case of a trabecular variant of JOF in a 17-year-old female patient.

Case Report

A 17-year-old female represented with a chief complaint of pain and slowly progressive swelling of the left side of face along with reduced mouth opening for the last 2 months. Initially, the swelling was smaller in size and increased progressively to attain final size causing facial asymmetry with reduced mouth opening. There was no relevant medical and dental history. Extraoral examination revealed an ill-defined, diffuse, and bony hard swelling of approximately measuring about 6 cm × 4 cm on the left side of the face,

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Figure 1: (a and b) preoperative extraoral view of the patient showing swelling on the left side of face

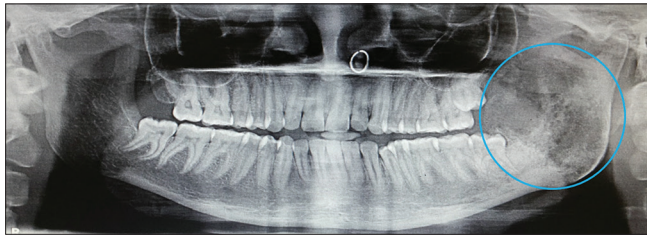


Figure 3: Preoperative panoramic view of the patient

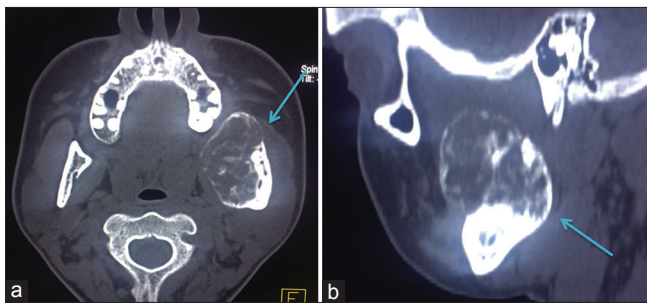


Figure 5: (a) Axial view of computed tomography, (b) sagittal view of computed tomography

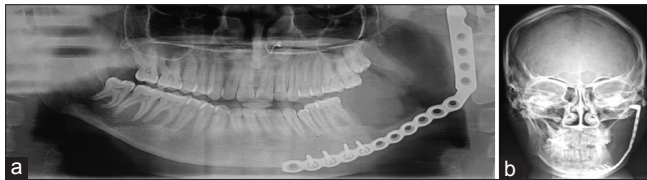


Figure 7: (a) Postoperative panoramic view of the patient, (b) postoperative posteroanterior view of the patient



Figure 8: (a and b) Postoperative extraoral view of the patient with intact facial symmetry



Figure 2: (a) Preoperative mouth opening, (b) postoperative mouth opening

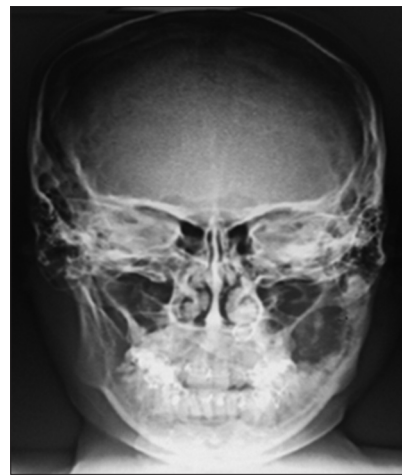


Figure 4: Preoperative posteroanterior view of the patient

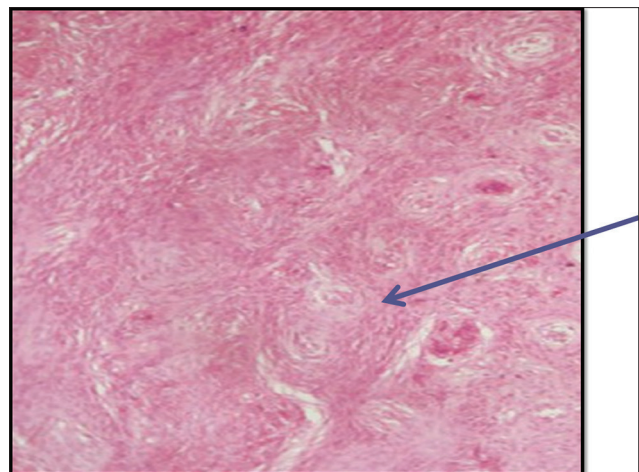


Figure 6: Histopathological section

extending superior-inferiorly from left zygomatic arch till angle of the mandible and anteroposteriorly from nasolabial fold till tragus of the left ear with normal overlying skin [Figure 1a and b] Intraorally, swelling was solitary, slightly tender with the expansion of buccal and lingual cortical plates with no pathological changes to the overlying mucosa.

There was reduce mouth opening measuring about 15 mm in the greatest dimension [Figure 2a]. Orthopantomogram revealed a large well-bordered, well-defined unilocular, circular, mixed lesion (calcification specks) approximately, 2 cm in size involving the ramus, coronoid, and angle of the mandible [Figure 3] posteroanterior view of mandible showed a well-defined, mixed lesion involving the coronoid process, ramus and the angle of the left side of the mandible [Figure 4]. Axial and sagittal view of computed tomography confirmed the routine radiographic features and further showed a heterogeneously enhancing mixed lytic-sclerotic lesion with a sclerotic rim extending into adjacent soft tissue measuring 5.12 cm × 3.83 cm with exterior and superior extension into the left infratemporal fossa and medial extension up to pterygoid muscle [Figure 5a and b]. Biopsy was taken from the involved site, and the H and E section showed the presence of trabeculae of fibrillar osteoid and woven bone. In few areas, multinucleated giant cells were also noted. Peripherally bony trabeculae lined by osteoblast were also seen [Figure 6]. Based on clinical, radiographic, advanced imaging studies and histopathological report provided the diagnosis of the trabecular variant of JOF affecting left mandible with differential diagnosis of cement-ossifying fibroma, osteoblastoma, and osteogenic sarcoma. The lesion was excised by segmental resection using transmandibular approach and reconstruction was done [Figure 7a and b]. The closer was done using 3.0 vicryl sutures. Postoperative healing was uneventful; mouth opening was increased to the value of 30 mm [Figure 2b]. Postoperatively, the esthetics, oral function of the patient such as speaking, and chewing were intact and normal [Figure 8a and b]. The patient is still under regular follow-up as the lesion has high recurrence rate.

Discussion

JOF mostly strikes children with slightly male predominance with a mean age of 11 years. Clinically, it is asymptomatic, however rarely it is aggressive and symptomatic.^[7] In the present case, the patient was symptomatic showing its rare entity. According to Slootweg and Müller, both maxilla and mandible are affected with more prevalence for maxilla.^[8] In the present case, the duration of swelling noted was 2 months. The characteristic radiographic features reveal a central opacification with well-defined unilocular to multilocular pattern giving it a ground glass appearance.^[9] This radiographic appearance depends on duration, stage, and histology of the lesion. The 50% of cases of JOF are recorded in multiple sinuses with few occurring in the single sinus.^[9] The duration, clinical and radiographic features and extension of the lesion into the adjacent soft tissue favors the aggressive type of JOF as previously reported.^[3] The features like resorption and perforation of cortices, tooth displacement and aggressive growth of the lesion favor the diagnosis as an aggressive variety of JOF. In this case, except for tooth resorption all other features like perforation of the cortices and extension into the surrounding tissue was noted. Because of the high recurrence rate, there is no follow-up protocol

mentioned in the literature and immediate reconstruction is not advised. The smaller lesions are treated by enucleation and curettage successfully.^[10] Resection is considered where there is recurrence, invasion into surrounding tissues, or when preserving the inferior border is not feasible.^[11]

Conclusion

JOF is an uncommon clinical entity, because of its aggressiveness and high recurrence rate it is necessary to arrive at an early diagnosis, applying the appropriate clinical, diagnostic approaches, and appropriate treatment for effective results and long-standing follow-up.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed

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Nil.

Conflicts of interest

There are no conflicts of interest.

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