

Juvenile ossifying fibroma of the mandible: A case report

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ABSTRACT

Juvenile ossifying fibroma (JOF) is a rare fibro-osseous entity of the craniofacial skeleton, which poses diagnostic and therapeutic difficulty due to its characteristic behavioral, clinical, and histopathological features. The maxillary involvement of the lesion leads to, nasal obstruction, epistaxis and exophthalmos. The tumor very rarely involves mandible and only a few cases have been reported. It shows slight male predilection with age ranging from 2- 15 years has been reported. Here, we report the case of juvenile ossifying fibroma which came to the department with a chief complaint of swelling in the lower right side of the face since three months. After obtaining the final histopathological diagnosis, the lesion was treated with surgical excision.

Keywords: Computed Tomography, Fibro osseous lesions, Juvenile Ossifying Fibroma, Orthopantomogram.

Juvenile ossifying fibroma (JOF) is a rare fibro-osseous entity of the craniofacial skeleton, which poses diagnostic and therapeutic difficulty due to its characteristic behavioral, clinical, and histopathological features [1]. 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]. JOF (psammomatoid variant) usually involves extragnathic craniofacial region especially in orbital bones and paranasal sinuses (61.6%), jaws, maxilla (19.7%) and mandible (7%) [3].

The maxillary involvement of the lesion leads to nasal obstruction, epistaxis and exophthalmos. It shows slight male predilection with age ranging from 2- 15 years has been reported. In mandible, the tumor occurs more commonly in the ramus than in the body of the mandible. Radiographically juvenile ossifying fibroma appears as unilocular or multilocular

radiolucency with occasional opacification and well defined borders which purely depend upon the stage and time of examination of the lesion.

Cortical thinning, perforation, tooth displacement and root resorption are the features of the JOF [4]. It is seen most commonly in the craniofacial bones of children less than 15 years of age and very rarely been reported in the age range of 3 months to 72 years. Juvenile ossifying fibroma adult variant varies on the basis of the age of occurrence, anatomic site of involvement, high tendency for recurrence, and its locally aggressive behavior [2]. Here, we report the case of juvenile ossifying fibroma in a 5-year-old patient.

CASE REPORT

A 5-year-old male patient reported in our hospital with a chief complaint of swelling in the lower right side of the face for three months. The swelling was initially small in size and had gradually



Figure 1: Extraoral view of the 5-year-old patient.

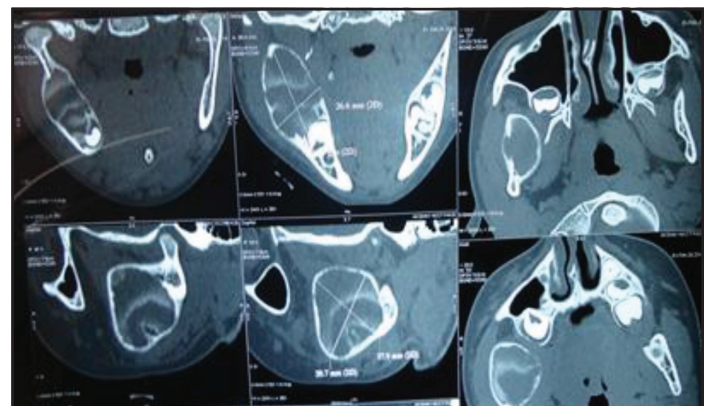


Figure 2: CT of the 5-year-old patient.

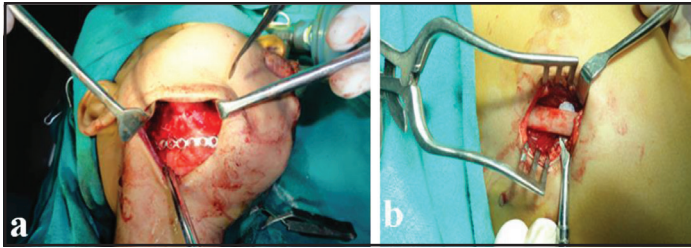


Figure 3: (a) Placement of reconstruction plate extraorally; (b) harvesting of rib bone.

increased to attain the present size and relatively asymptomatic. The patient did not give any history of trauma or any other surgical intervention in the past. The medical and family history of the patient was non-contributory.

On general physical examination, patient was well-nourished (Fig. 1). On extraoral examination, there was gross facial asymmetry caused by the diffused swelling on the right lower side of the face with normal smooth overlying skin and the size of the swelling was approximately 4x4 cm in the greatest dimension. The swelling extended anteroposteriorly from 1 cm below from corner of the mouth till the angle of the mandible and superoinferiorly from an imaginary line drawn from corner of mouth to the tragus till the inferior border of mandible. The swelling was bony hard in consistency not fixed to underlying structures, non-tender and without crepitus (Fig. 1).

The intraoral examination revealed mixed dentition status with a diffused swelling in the right mandibular region and vestibular obliteration in relation to the right first molar till the retromolar region was noticed. The swelling was firm, non-reducible, non-compressible, non-tender and without crepitus on palpation. No evidence of draining fistula or sinuses or mobility or displacement of teeth was noticed.

Computed tomography (CT) revealed a radiolucent region of approximately 4.25 cm anteroposteriorly 2.6 cms buccolingually and 2.35 cms superoinferiorly, in the right mandibular region extending from the 85 region involving the body, angle and ramus of the mandible. Thinning of lingual and buccal cortical plates were evident. The internal structure of the lesion was not completely radiolucent with diffused radiopaque areas within the lesion. There was a displacement

of the mandibular canal more buccally. After the history, clinical examination and radiographic investigation the provisional diagnosis of Juvenile ossifying fibroma was made (Fig. 2).

An incisional biopsy was performed under local anesthesia providing a diagnosis of trabecular Juvenile ossifying fibroma. The histopathological picture revealed cell-rich fibrous stroma containing strands of cellular osteoid without osteoblastic rimming along with few trabeculae of typical woven bone.

Surgical management was done after receiving the histopathologic diagnosis. After the general anesthesia, complete excision of the tumor was done with 5 mm safety margins. Intraoperatively, the tumor was well-encapsulated. After the excision of tumor we prepared the chest for non-vascularized graft (Fig. 3b). Preserving the periosteal envelope, 5th and 6th ribs were harvested of appropriate length through a chest incision. The bone graft was then kept in a solution of 300mg clindamycin/500ml normal saline. Careful hemostasis of the donor site was achieved and closer was done with 3-0 vicryl and skin sutures were taken with 5-0 proline and pressure dressing was applied. The site of the facial lesion was then open and prepared to receive the bone graft. The graft was prepared to fit the defect and secured with reconstruction plates and screws (Fig. 3a, 4, 5), operated site was closed with 3-0 vicryl and intermaxillary fixation was applied for 4 weeks postoperatively after that patient was followed up for 8 years without recurrence (Fig. 6).

DISCUSSION

Juvenile ossifying fibroma shows slight male predilection with age ranging from 2- 15 years. Very rarely, it has been reported in the age range of 3 months-72 years. Juvenile ossifying fibroma adult variant varies on the basis of the age of occurrence, anatomic site of involvement, a high tendency for recurrence, and its locally aggressive behavior [2]. Blacks are more prone to ossifying fibromas than whites. Mostly, fibro-osseous lesions are asymptomatic and progress slowly but it can show aggressive and destructive growth when seen in patients below 15 years of age [5]. JOF arises as a result of the differentiation of mesenchymal cells of periodontal ligament, the multipotential precursor cells, into cementum, osteoid, or fibrous tissue [5].



Figure 4: Placement of reconstruction plate with rib graft.



Figure 5: Postoperative OPG of the patient.

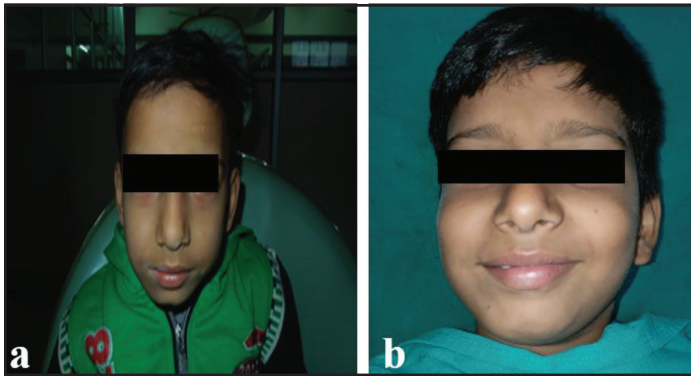


Figure 6: (a) 6 months postoperative frontal profile of the patient; (b) follow-up after eight years

JOF is a heterogeneous entity with two distinct histopathological types: Ps-JOF and Tr-JOF [3]. Accurate identification and diagnosis of these two different entities include the cemento-ossifying fibroma and fibrous dysplasia, are important, to make correct therapeutic management [3]. Ps-JOF and Tr-JOF can be distinguished from each other by the age in which it appears, the site that is involved and its clinical behaviour [6]. Ps-JOF is described predominantly in children in the literature and is not exclusive to those below 15 years of age. A review by Cicciù *et al* showed that the patients ranging in age from 3 months to 72 years, with 62.8% of cases occurring between 5 and 15 years of age [3]. On comparing, Tr-JOF and Ps-JOF, it occurs in older and wider age groups with unclear sex prediction because of conflicting studies.

The majority of the cases of Ps-JOF are seen affecting the orbital bones and paranasal sinuses. However, it is least commonly present in the mandible with an incidence of only 8%. In the mandible, the most frequently involved site is the ramus followed by anterior mandibular lesions are rare. But when seen in contrast to Tr-JOF, it predominantly involves the maxilla, followed by the mandible [6].

JOF can be discovered clinically by the presence of bony expansion but in early cases, it can often be incidental due to routine radiographic examinations. The radiographic presentation varies, it can be unilocular as well as multilocular but the most common presentation is the circumscribed unilocular lesion that may show central radiopacities with ground glass appearance. In aggressive lesions, there is evident cortical thinning and perforation [6].

The primary modality of treatment of JOF is surgical management. JOF is not radiosensitive to radiotherapy however an adjuvant interferon Therapy has been reported in the literature. The management of JOF also includes enucleation and curettage.

JOF has a high recurrence rate of almost 30-56% in cases of surgical excision. Incomplete excision and difficulty in surgery due to access to the location of the lesion result in recurrence due to the infiltrative nature of the tumor. So as to reduce the recurrence rate the authors have suggested segmental resection with a 5 mm margin as a definitive treatment of JOF. Autogenous graft and oral rehabilitation are used as reconstructive materials to restore oral function and aesthetics. Immediate reconstruction is not advised because of its highly recurrent nature [7]. In our case, the reconstruction was done by both bone graft as well as reconstruction plate and the results were excellent.

CONCLUSION

Management of aggressive juvenile fibroma is evolving continually. Generally, an aggressive type of fibroma requires complete surgical excision. Large or recurrent cases especially involving maxillary region needs en-bloc resection. In the above-discussed case, there was no recurrence even after the period of eight years. Hence, we would like to conclude that JOF especially aggressive type should be carefully managed surgically and the patient should be under long term follow up for any recurrences.

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