

Trabecular Type of Juvenile Ossifying Fibroma – A Rare Case Report

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Abstract: Juvenile ossifying fibroma (JOF) is a rare controversial fibroosseous lesion affecting the craniofacial skeleton and occurring commonly in children and young adults. It is highly aggressive and has a high tendency to recur. It is distinguished from the adult variant of ossifying fibroma on the basis of age, site, clinical behaviour and microscopic appearance. Because of high recurrence rate, which is 30-58% complete excision is essential. We report a rare case of trabecular JOF of maxilla which was confirmed histologically.

Keywords: Juvenile ossifying fibroma; Fibro-osseous lesions; Ossifying fibroma, craniofacial skeleton, Trabecular

1. Introduction

Juvenile ossifying fibroma (JOF) term is used for a rare actively growing lesion that mainly affects individuals younger than 15 years of the age [1]. This lesion behaves in an aggressive fashion, reaching massive proportions with extensive cortical expansion. Over time, lesions with this morphology have been variously described as juvenile ossifying fibroma [2], active juvenile ossifying fibroma [3], aggressive ossifying fibroma [4], reticular desmooosteoblastoma [5] or active fibrous dysplasia [6]. This lesion most commonly involves the paranasal sinuses and periorbital bones, where it may cause exophthalmos, proptosis, sinusitis and nasal symptoms. This rare tumor behaves in a more aggressive fashion than does ossifying fibroma, may be mistaken as low grade osteosarcoma, which may alter the treatment. Juvenile ossifying fibroma is a well defined clinical and histological entity that has been separated from other central fibro osseous lesions, including the cemento ossifying fibroma [4]. It is described in WHO classification [1] as “an actively growing lesion consists of a cell rich fibrous stroma, containing bands of cellular osteoid without osteoblastic rimming together with trabeculae of more typical woven bone. Small foci of giant cells may also be present, and in some parts there may be abundant osteoclasts related to woven bone. Usually no fibrous capsule can be demonstrated, but like the ossifying fibroma (and unlike fibrous dysplasia), the JOF is well demarcated from the surrounding bone”. In the jaw, JOF is considered to develop from undifferentiated cells of the periodontal ligament, usually in the premolar and molar region. The differential diagnosis with other fibro-osseous lesions of the jaw, such as cemento-ossifying fibroma, should be made [7]

2. Case Report

A 15-year-old male presented with swelling in the upper back region of jaw since two months. The swelling had gradually increased in size over two months. There was no associated pain or pus discharge. Extra oral examination revealed a single, non-tender, bony-hard, diffuse swelling over the right maxilla. Intra-orally, a bony-hard, non-tender, swelling involving vestibule and palate in relation to the teeth 13, 14, 15, and 16 was observed (Fig. 1). No dental

focus of infection detected. OPG revealed mild ground-glass like radiographic area with a partially lining sclerotic border, between and overlapping the roots of 15 and 16. The roots of 15 and 16 were diverged. Results of blood investigations were within normal limits. Serum ALP was normal. CT scan revealed a large rounded mass of the right maxilla, infiltrating the right maxillary antrum (Fig. 2) The lesion was debulked and submitted for microscopic evaluation. Histological examination of hematoxylin and eosin stained slides demonstrated a bony trabeculae of varying size in fibrocellular stroma. Trabeculae consist of lamellar and predominantly woven bone with osteoblastic rimming (Fig. 3). One area showed accumulation of few multinucleated giant cells (Fig. 4). Cellular osteoid strands were also seen in loose fibroblastic stroma. Stromal degeneration and cystic spaces were also seen. The lesion was diagnosed as trabecular type of juvenile ossifying fibroma.

3. Discussion

The JOF, as the name suggests, has its higher incidence in children and young adults [2,3]. Among the many classification systems for this lesion, the classification by Slootweg et al. [4] had divided JOF into two distinct groups, the JOF-WHO type and JOF-PO (psammoma-like ossicles) type, based primarily on the difference in the age of occurrence: the mean age of occurrence of JOF-WHO is 11.8 years and that of JOF-PO is 22.6 years [4]. El-Mofty [5] recent classification identified two categories, trabecular JOF (TJOF) and psammomatoid JOF (PJOF), based on histologic criteria. However, the two categories also have a distinct predilection for specific age-groups: the average age of occurrence of TJOF is 8½–12 years, whereas that of PJOF is 16–33 years [5]. Although JOF can occur anywhere in the skeleton, its highest incidence is in the facial bones, most commonly the maxilla [2,6]. One clinical feature that helps differentiate TJOF from PJOF is the site of involvement, with PJOF occurring mainly in the paranasal sinuses and TJOF occurring mainly in the maxilla [5]. Mandibular and extracranial involvement are rare [2]. Gender predilection has been a matter of controversy, with some authors claiming no predilection for either gender, whereas Johnson et al. found a higher incidence in females

[2] and El-Mofty reported a male predilection [5]. JOF clinically manifests as an asymptomatic bony-hard swelling, the duration and extent of which may vary depending on the site and aggressiveness of the lesion. It can expand the involved bones, causing facial asymmetry. Depending on the site, symptoms such as pain, paresthesia, malocclusion, sinusitis, proptosis, etc., can also occur due to the swelling [6,8].

Radiographically it can be radiolucent, mixed, or radiopaque, like any fibrous lesion depending on the degree of calcification [4], with root displacement commonly and resorption rarely [4,6,8]. The lesion can cause expansion as well as perforation [5]. A 'ground-glass' appearance on radiographs has been reported which can be mistaken for fibrous dysplasia [5]. It usually has a concentric or centrifugal growth pattern, which can lead to an erroneous clinical diagnosis of cemento-ossifying fibroma [6,9].

The microscopic features of the lesion are distinctive and include a cell-rich fibrous stroma containing bands of cellular osteoid without osteoblastic lining, osteoid strands, and trabeculae of woven bone [4,5,9]. Osteoid matrix may develop incorporated plump eosinophilic osteoblastic cells. Multinucleated giant cells are commonly seen. JOF-PO (psammoma-like ossicles) type is slightly more cellular than JOF-WHO type. Due to the resemblance of the psammoma-like ossicles seen in JOF-PO type to the The aggressive nature of this entity, along with the reported high rates of recurrence (30–58%), [5,6] suggests that JOF should be treated like a locally aggressive neoplasm. Surgical resection, rather than conservative curettage, is therefore the preferred line of treatment [3,6]. cementicles in cemento-ossifying fibroma, it has been argued that JOF-PO a type of cemento-ossifying fibroma [5]. However, the marked cellularity of JOF-PO is in sharp contrast to the usually stroma-rich appearance of the latter group of lesions.

4. Conclusion

Juvenile ossifying fibroma is an aggressive lesion which needs to be differentiated clinically and histologically from other fibro-osseous lesion so that appropriate treatment can be given in accordance with the age of the patient, keeping in mind the young age of patient. It is very important to have minimum five year follow up.

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Figure 1: Intra-orally, a bony-hard, non-tender, swelling involving vestibule and palate in relation to the teeth 13 to 16.

Figure 2: CT scan revealed a large rounded mass of the right maxilla, infiltrating the right maxillary antrum.

Figure 3: Revealed numerous bony trabeculae with prominent osteoblastic rimming, interspersed in dense fibrous connective tissue stroma.

Figure 4: Area shows accumulation of few multinucleated giant cells.



Figure 1



Figure 2

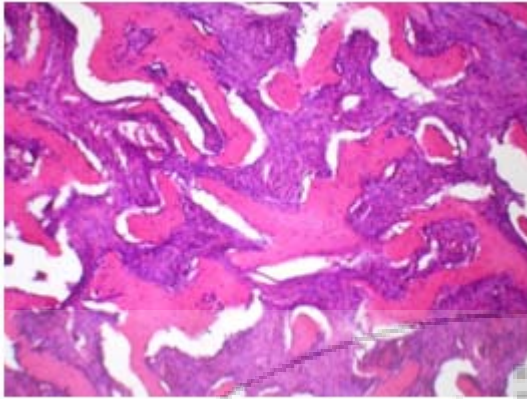


Figure 3

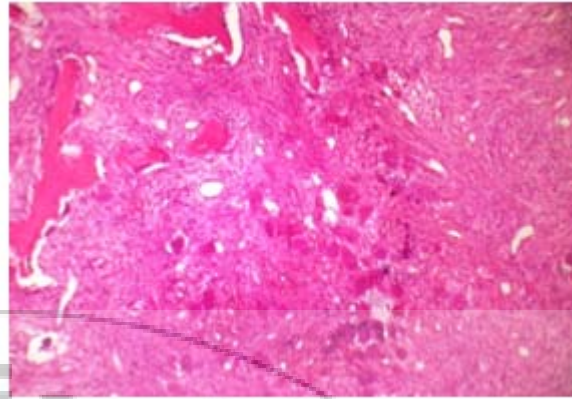


Figure 4

