

Juvenile psammomatoid ossifying fibroma: An unusual case report

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Abstract

Ossifying fibromas are well-demarcated benign fibro-osseous tumors of craniofacial skeleton most often in the jaws. It has two types, conventional and juvenile ossifying fibroma (JOF). JOF is considered a separate entity from ossifying fibroma due to its locally aggressive behavior and tendency to occur at a young age. Two subtypes of JOF, i.e., juvenile psammomatoid ossifying fibroma (JPOF) and juvenile trabecular ossifying fibroma have been identified on the basis of histology 70% of the JPOF occur in the paranasal sinuses, 20% in the maxilla and only 10% in the mandible. Here, we report a case of JPOF in a 14-year-old girl causing an asymptomatic expansile swelling in the right mandibular posterior region, which is a rare site for this tumor.

Keywords: Fibro-osseous lesion, juvenile, ossifying fibroma, psammomatoid

Introduction

The term “fibro-osseous lesions” refer to conditions in which normal bone architecture is replaced by fibroblasts and collagen fibers and contain various amounts of mineralized material.^[1] It includes entities such as ossifying fibroma, Juvenile ossifying fibroma (JOF), fibrous dysplasia, etc.

Two types of ossifying fibromas are the central type and peripheral type. Central ossifying fibroma is again of two types, the conventional type and juvenile type. JOF is a locally aggressive variant of central ossifying fibroma of the jaws and has a high tendency for recurrence. It most commonly occurs at a younger age. Clinically, it produces large asymptomatic tumor of jaws. The usual site is premolar and molar region with associated teeth being displaced, missing or unerupted.^[2]

Radiographically, the tumor is demarcated from the surrounding bone by a well-defined radiopaque border. Two

microscopic patterns of JOF are juvenile trabecular ossifying fibroma (JTOF) and juvenile psammomatoid ossifying fibroma (JPOF).^[3] Psammomatoid type has the presence of eosinophilic spherical structures called psammoma bodies. The preferred sites for this lesion are paranasal sinuses, maxilla, ethmoid and frontal bone.^[4] The most common clinical manifestations are proptosis, nasal obstruction, headache, swelling and rarely epistaxis. JPOF is considered predominantly a craniofacial lesion with only 10% cases occurring in the mandible.

Case Report

A 14-year-old female patient reported with the chief complaint of large painless swelling on the right lower jaw region since 1 year. The swelling had rapidly grown since 5 months to attain its present size. There was no history of trauma, pain, any discharge and extraction of any teeth. The right side submandibular lymph node was palpable, single in number, about 1 cm in size, mobile, soft in consistency and non-tender on palpation.

Extraorally a diffuse swelling was present on the right lower side of face, approximately 10 cm × 6 cm in size, extending anteroposteriorly 2 cm away from the midline on left side until right angle of mandible and superoinferiorly from a line connecting angle of mouth to angle of mandible until 2.5 cm below the lower border of mandible. Color and temperature of overlying skin was normal. It was non-tender and hard in consistency.

Intraorally lower labial and buccal cortical expansion was present extending from midline until 48 region and minimal lingual cortical expansion was present [Figure 1]. Hard-tissue examination revealed 41 was rotated, 43 was labially erupted and deciduous canine and molars were retained. 44 and 45 were clinically missing. Provisional diagnosis of Dentigerous cyst with 15 was made. Differential diagnosis given was unicystic ameloblastoma, ameloblastic fibroma,

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Figure 1: Intraoral photograph showing buccal expansion

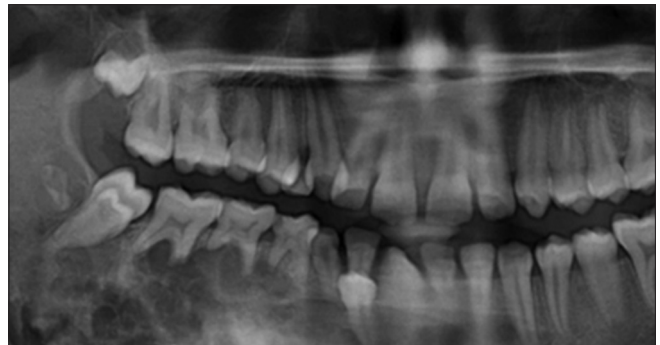


Figure 2: Orthopantomograph showing extensive nature of lesion with ballooning and thinning of lower cortical plate

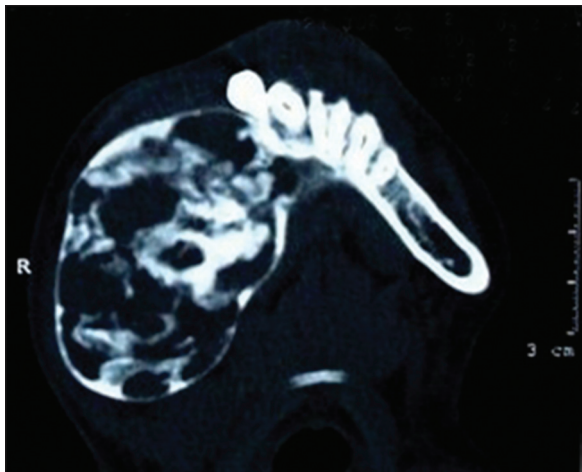


Figure 3: Axial section showing buccal and lingual cortical plate expansion with multiple internal septa

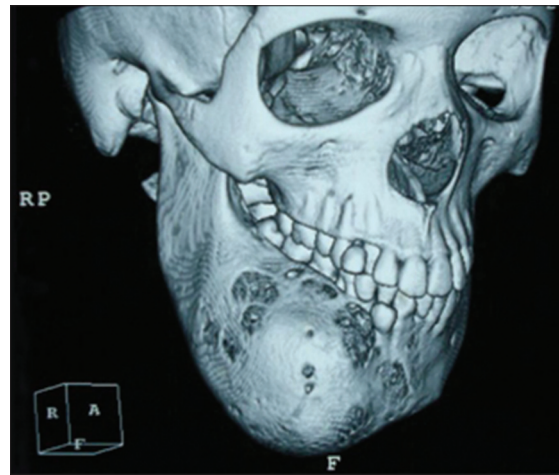


Figure 4: Spiral computed tomography scan showing multiple perforation and internal septa

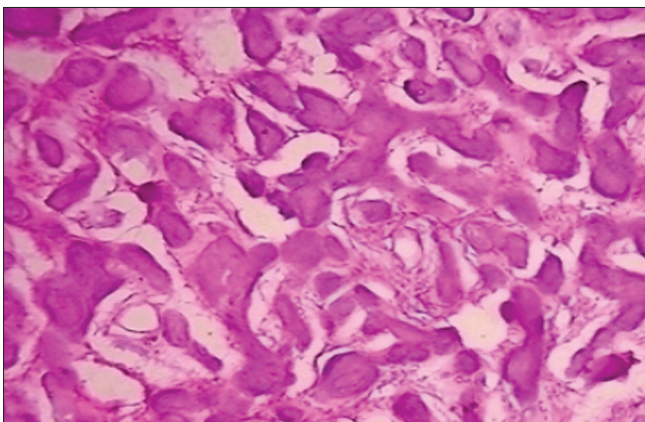


Figure 5: Photomicrograph showing small spherical hematoxyphilic ossicles i.e., psammoma like bodies

calcifying epithelial odontogenic tumor, ossifying fibroma and odontogenic myxoma.

On fine needle aspiration cytology no fluid was aspirated. Orthopantomograph (OPG) revealed mixed radiolucent radiopaque lesion extending from 34 until distal aspect of



Figure 6: Post-operative orthopantomograph of patient



Figure 7: Intraoral post-operative photograph

47 regions with a radiopaque dense mass near the apical aspect of 84 and 85. Borders were well-defined and thinning of lower border of mandible was present. Root resorption of 85, 86 and 46 was present. Inferior alveolar canal was displaced to lower border of mandible [Figure 2].

Axial, coronal and sagittal sections of computed tomography scan showed multilocular lesion with multiple internal septa [Figure 3]. It revealed intact lower border of mandible and the lesion crossing the midline. 3D reconstruction showed formation of internal septa and areas of homogenous calcification [Figure 4].

Histopathological examination showed small spherical hematoxyphilic ossicles i.e., psammoma such as bodies, cellular connective tissue stroma and bony trabeculae [Figure 5]. The final diagnosis was JPOF. The patient was referred to Oral surgery department for surgical removal of the tumor. 6 months post-operative OPG of patient [Figure 6] and intraoral [Figure 7] examination showed no recurrence.

Discussion

According to the new classification of World Health Organization, ossifying fibromas which appear as fast growing mass between 5 years and 15 years of age, are radiologically well-bordered and consistent with ossifying fibroma histologically are referred as JOF.^[5]

JOF is considered a distinct entity from other fibro-osseous lesions because of its locally aggressive behavior and tendency to occur at a young age. It occurs in the craniofacial skeleton and 79% of patients are diagnosed before the age of 15 years. Johnson *et al.* stated that mandibular tumors are more common in girls in 2nd to 4th decades of life.^[5] This is in accordance with our case of a 14-year-old girl.

Clinically, JOF appears aggressive and the first clinical manifestation is swelling of mandibular cortical layer, which produces a marked extra-oral facial asymmetry.^[6] In the present case, an asymptomatic expansile swelling on the right side of jaw causing facial asymmetry was present.

Two distinct histological subtypes are JTOF and JPOF. In a review by El-Mofty (2002), 230 cases of psammomatoid type were reported.^[4]

Some authors suggest a slight male predominance where as others say that males and females are equally affected.^[7] Average age of occurrence for JPOF is 16-33 years and for JTOF is 8½-12 years.^[4] The developing tooth can either be displaced, missing or remain unerupted. In our case, the 1st and 2nd premolars were unerupted.

JTOF mainly occurs in the maxilla, mandible and fronto-ethmoid complex and JPOF occurs in paranasal sinus, calvarium, maxilla and mandible.^[1] JPOF is mostly a craniofacial lesion and rarely occurs in jaws. Johnson *et al.* and Makek in their case series found that 70% of JPOF occurred in the paranasal sinus, 20% in the maxilla and only about 10% in the mandible.^[8] Our case occurred in posterior mandibular area, which is a rare site for this tumor accounting for only 10% of the cases.

Histopathologically JTOF has long, slender and anastomosing trabeculae of osteoid in a fibrocellular stroma and JPOF has a presence of eosinophilic spherical structures dispersed in a fibrous stroma consisting of plump spindle shaped cells that are arranged as strands and whorls called psammoma like bodies.

Radiographically, the tumor is well-demarcated, unilocular or multilocular radiolucency with a variable amount of radiopacity, usually manifesting as fine specks or as ground glass opacification.^[9] Aggressive forms show expansile form of growth pattern, but retain the sharp demarcation from normal adjacent bone. It appears locally aggressive with cortical disruption and involvement of adjacent anatomical structures. Management is surgical with complete resection since partial removal is associated with recurrence.

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