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CASE REPORT

Juvenile Active Ossifying Fibroma of the Mandible: A Case Report

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ABSTRACT

Introduction: Juvenile ossifying fibroma (JOF) is a benign fibro-osseous lesion containing various amounts of calcified masses scattered in a fibro cellular stroma. Distinguishing feature of JOF from other fibro-osseous lesions is by its propensity to recur.

Case report: Herewith, we report a case of Juvenile ossifying fibroma in the mandible of a 10-year-old female girl. Based on the clinical, radiographic and histopathological evaluation, the present case was diagnosed as juvenile active ossifying fibroma

Management: The lesion was treated by partial mandibulectomy using transmandibular approach under antibiotic coverage. Oral functions of the patient remained adequate. The patient was kept under one year of follow up and did not show any sign of recurrence.

Conclusion: Juvenile active ossifying fibroma is considered to be a distinctive entity due to its predilection for a specific age group, complex histopathological appearance, and its aggressive clinical behavior. High recurrence rates have been reported which warrant a need for long term follow-ups.

Keywords: Fibro-osseous lesion, Ossifying fibroma, Psammomatoid, Trabecular.

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INTRODUCTION

Juvenile ossifying fibroma (JOF) is an uncommon benign fibro-osseous lesion containing variable amounts of calcifications, scattered in a cellular fibrous connective tissue stroma.[1] It is also known as 'Aggressive ossifying fibroma' due to its high potential to recur and aggressive nature. The most characteristic feature of JOF is its higher incidence of occurrence in children and young adults.[2] JOF is considered as a variant of cement-ossifying fibroma but unlike it, JOF can be characterized by a highly cellular stroma with cellular osteoid, woven bone, small foci of giant cells and abundant osteoclasts.[3]It has two distinct histological subtypes, trabecular and psammomatoid.[4] Whether both variants are component of a single lesion or entirely separate lesions is a subject of debate.[5] The treatment of JOF is surgical excision, but recurrence rates of up to 30% and 50% have been encountered.[1]

CASE DESCRIPTION

A 10-year-old female patient reported to the Department of Oral Medicine and Radiology with a chief complaint of swelling in the left back region of the lower jaw for 1 year. The swelling started slowly to attain the present size. Medical history of the patient was not significant. On extra oral examination, the swelling was oval measuring about 6 × 5 cm in diameter extending from the angle of mandible anteriorly to about 1 cm beyond the ramus of mandible posteriorly. Superiorly, swelling extended to the level of the ear lobe and inferiorly 2 cms beyond the lower border of mandible with the overlying skin being normal. On palpation, the swelling was non-tender and nonpulsatile. It was soft in the superior part while firm in the inferior part and was not attached to the underlying structures. Intraoral examination revealed an ovoid swelling present in the left posterior region of the mandible, extending from mesial surface of the first premolar to the retromolar area. It also involved buccal mucosa and measured 4 × 5 cms in diameter with an irregular smooth surface (Fig. 1).

Radiographic examination revealed an oval, circumscribed radiolucency with sclerotic margins with cortical expansion and resorption of the roots of the 1st molar (Fig. 2). Hence, a provisional diagnosis of ameloblastoma and the primordial cyst was given.

Considering the extent of the lesion, an incisional biopsy was performed which revealed a non-encapsulated lesion exhibiting highly cellular stroma. Within the stromal tissue, numerous ossicles of varying shapes with central mineralization and peripheral osteoid rimming

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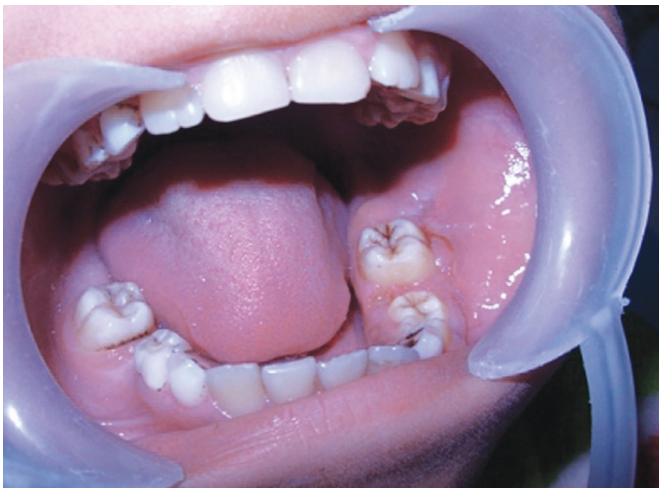

Fig. 1: Intraoral photograph showing swelling in the left posterior side of the mandible from premolar to the retromolar area

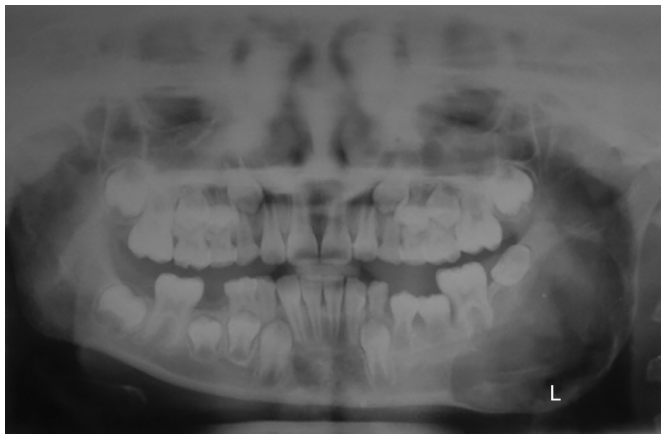

Fig. 2: Radiographic examination revealed an oval, circumscribed radiolucency with sclerotic margins, cortical expansion and resorption of the roots

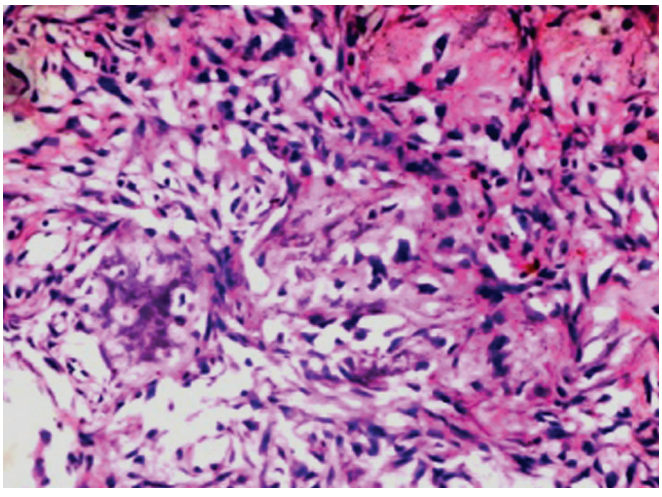

Fig. 3: H&E stained section of Juvenile ossifying fibroma highly cellular connective tissue stroma with numerous osseous trabeculae (40X)

were observed. Plump osteoblasts were seen lining the ossicles. Thus histological findings were suggestive of Juvenile ossifying fibroma. After histopathologic interpretation; the patient was advised for surgical excision of the lesion.

Grossly, the specimen was irregular in shape, firm to soft in consistency and brownish-white in color, measuring 2.5 × 2 cm in diameter. Later multiple H&E stained sections showed highly cellular connective tissue stroma with numerous osseous trabeculae and osteoid in a discrete pattern. Trabeculae were lined by plump osteoblasts which appeared normal in morphology. Chronic inflammatory cells and focal areas of hemorrhage were also present (Fig. 3). Histological features were consistent with juvenile trabecular active ossifying fibroma.

Treatment, Management and Follow-up

Considering the extent and size of the lesion, the patient underwent partial mandibulectomy using transmandibular approach under antibiotic coverage. The patient was kept under one year of follow up and did not show any sign of recurrence.

DISCUSSION

In 1938, Sir Benjamin was first to describe juvenile ossifying fibroma as an 'osteoid fibroma with atypical calcification'. Later in 1952, Sir Johnson et al. coined the term 'juvenile active ossifying fibroma'.[6] Juvenile Active Ossifying Fibroma (JAOF), a variant of Ossifying Fibroma (OF), is considered to be a distinctive entity because of its propensity to develop in younger age, more complex histopathology and has a potential for locally aggressive growth.[7] It is considered to be highly aggressive lesion and has a strong tendency to recur.[2] Makek in 1987 suggested two variants of JOF: 'trabecular desmo-osteoblastoma and psammomatous desmo-osteoblastoma'. According to the author, these lesions are of osteoblastic origin that arises in the intramembranous bones of the jaws, and skull.[8] Slootweg et al. in 1994 regarded them as different entities and preferred the term 'Juvenile ossifying fibroma' to these lesions that correspond to the WHO definition.[8]

The age range in JOF varies from 6 months to 70 years of age with a mean age of trabecular juvenile ossifying fibromas as 11 years whereas patients diagnosed with psammomatoid variant aged around 22 years.[7] Slight predilection in males has been noted with a male-to-female ratio of 1.3:1 which was in contrast to our study. The maxilla is the most frequent site (54%), followed by the mandible (35%) and the fronto-ethmoid complex (9%). JOF is slow-growing and is usually well-circumscribed.[8] Both the trabecular and psammomatoid JOF are distinguished because of their age predilection, clinical behavior, and histological features.[9]

Radiographically, the lesion appears as circumscribed radiolucency with cortical expansion. The trabecular variant of JOF is common in jaws. The psammomatoid

variant frequently appears in the orbital and frontal bones and also in paranasal sinuses. Nasal obstruction, exophthalmos or proptosis may be seen. Rarely, temporary blindness occurs.[6]

Histopathological features show the tumor with cellular fibrous stroma that exhibits areas that are loose and other regions are highly cellular that the individual cell cytoplasm is hard to discriminate due to nuclear crowding.[7] Hemorrhagic areas and clusters of multinucleated giant cells are usually seen.[3] calcification in the two variants is different. The trabecular variant has irregular strands of highly cellular osteoid lined by plump osteoblasts and multinucleated osteoclasts. In contrast, the pathognomonic feature of the psammomatoid pattern forms is the presence of eosinophilic spherical structures scattered in a fibrous stroma consisting of plump spindle-shaped cells that are arranged as strands and whorls.[10]

Management and prognosis of the juvenile ossifying fibroma are not certain. Smaller lesions are treated with complete local excision or thorough curettage but for rapidly growing lesions, wide surgical resection is required. Thirty to fifty percent of recurrence have been reported for JOF.[11]

CONCLUSION

A juvenile active ossifying fibroma is regarded as a specific entity due to its predilection for a specific age group and its aggressive clinical behavior. It has a complex histopathological appearance. Clinical management usually includes complete local excision or curettage, but for rapidly growing lesions, wide surgical resection is required. High recurrence rates have been reported which warrant a need for long term follow-ups.

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