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CASE REPORT

Trabecular Juvenile Ossifying Fibroma of Mandible

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ABSTRACT

Juvenile ossifying fibroma is classified as a rare fibro-osseous neoplasm that usually but not exclusively occurs in children under 15 years of age, affecting the craniofacial bones. It is a locally aggressive lesion and spreads quickly. Histologically, it is composed of cellular, fibrous stroma as well as loose myxoid foci, areas of pseudocystic degeneration and small clusters of multinucleated giant cells. Based on the type of mineralized component, two histological variants are recognized, psammomatoid and trabecular, with the psammomatoid variant containing concentric lamellated ossicles and trabecular variant containing irregular strands of highly cellular osteoid encasing plump and irregular osteocytes. Reported here is a case of trabecular juvenile ossifying fibroma involving the mandible of a 19-year-old boy.

Keywords: Fibroma, Ossifying, Fibrous dysplasia of bone, Odontogenic tumors.

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INTRODUCTION

Juvenile ossifying fibroma (JOF) is described in literature as a rare variant of ossifying fibroma, occurring in the craniofacial skeletal of young individuals.[1,2] It occurs in two distinct histopathological variants referred to as psammomatoid juvenile ossifying fibroma (PsJOF) and trabecular juvenile ossifying fibroma (TrJOF).[1,3,4] The term JOF was initially introduced by Johnson et al[3] in 1952 and included in the second edition of the World Health Organization classification of odontogenic tumors.[5] It manifests usually as an asymptomatic swelling exhibiting aggressive behavior. The lesion is nonencapsulated but well demarcated from surrounding bone.[6-8] Histologically, JOF is characterized by a cell rich fibrous stroma, pseudocystic degeneration, areas of hemorrhage, small clusters of multinucleated giant cells and a variable mineralized component.[4,6,7] The two histological variants of JOF exhibit different patterns of mineralized components. The entity designated as PsJOF exhibits concentric lamellated ossicles having typically basophilic centers and peripheral eosinophilic osteoid rims, while the TrJOF shows irregular strands of highly cellular osteoid incorporating plump and irregular osteocytes.[1,4] In this article, a case is reported of a TrJOF involving the right mandible of a 19-year-old boy. This article seeks to highlight the diagnostic dilemma posed by the present case due to its atypical presentation.

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CASE REPORT

A 19-year-old boy presented with an asymptomatic gradually expanding swelling over the right side of lower jaw from one and half years. The past medical and dental history as well as family history was not significant. Upon clinical examination, a diffuse, hard swelling measuring 5 × 3.5 cm was observed over the right mandibular body region (Fig. 1). The swelling was nontender and hard in consistency. The overlying skin appeared normal. There were no palpable cervical or submandibular lymph nodes. Intraorally, obliteration of labial sulcus on the right side was seen. No abnormality was observed with the teeth. Orthopantomograph (OPG) revealed a mixed radiolucent/radiopaque expansile mass affecting the right body of mandible (Fig. 2).

A provisional diagnosis of fibrous dysplasia was made. An incisional biopsy specimen was submitted for histopathological examination which revealed a nonencapsulated lesion showing infiltration of surrounding bone structures and reactive new bone formation at the periphery. The lesion consisted of uniformly fibrocellular connective tissue containing irregular strands of highly cellular osteoid encasing plump and irregular osteocytes and a few areas of pseudocystic degeneration (Fig. 3). On the basis of these findings, a diagnosis of trabecular juvenile ossifying fibroma was made. A total excision of the lesion was performed under general anesthesia. Follow-up of the case is not available.

DISCUSSION

The term ossifying fibroma also known as cemento-ossifying fibroma and cementifying fibroma is described as an odontogenic neoplasm arising from the periodontal ligaments and affecting the tooth bearing areas of the jaws.[9] It occurs

in adults, being more common in women. The lesion is usually encapsulated and can be surgically shelled out easily from the surrounding bone. Histologically, it is composed of trabeculae of immature/woven bone or cementum like tissue or both in a fibrous stroma.[1,4,9] The entity under discussion here, JOF trabecular variety is a benign tumor of craniofacial skeleton occurring predominantly but not exclusively in children below 15 years of age. In a review published by Slootweg et al,[4] the mean age of onset was 11.8 years. There was a slight male predilection with male to female ratio of 1.5:1. Clinically, this lesion has a more aggressive growth compared to ossifying fibroma.[10] Most cases of JOF are asymptomatic as was in our case. The first clinical manifestation is a gradual, painless swelling of the affected bone, usually the maxilla, although the trabecular variant of JOF seems to show more aggressive growth[1,6-8,10] If the orbital bones or paranasal sinuses are involved, then patient may develop exophthalmos, bulbar displacement, nasal obstruction and epistaxis. The radiological features depend upon the location of tumor and amount of calcified tissue produced by the tumor. Accordingly, the lesion may show varying degrees of radiolucency.[2,10]

Microscopically, two variants are recognized, each differing in the appearance of their mineralized component. Both types of JOF are characterized by the proliferation of a highly cellular, fibroblastic spindle cell stroma which may show areas that are loose and myxoid. Myxoid regions may be associated with pseudocystic degeneration. Other common features of JOF include areas of hemorrhage, focal collections of multinucleated giant cells and mitotic figures which are not numerous.[11] The mineralized component in the two patterns is different. The TrJOF demonstrates anastomosing trabeculae of immature, woven bone incorporating plump eosinophilic osteoblastic cells. On the other hand, the PsJOF shows concentric, lamellated ossicles that typically have basophilic centers with peripheral eosinophilic rims.[1,4,11]

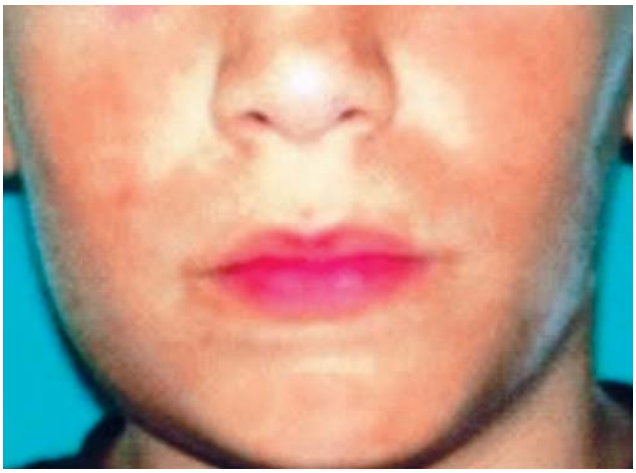

Fig. 1: A diffuse swelling over the right body of mandible

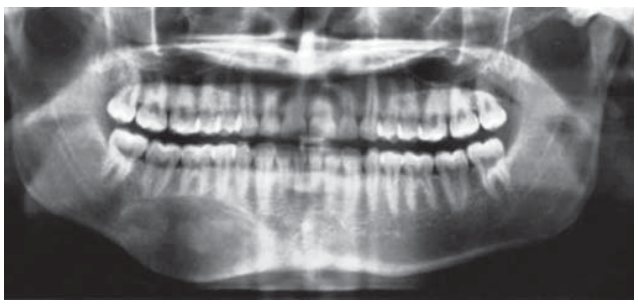

Fig. 2: Orthopantomograph showing a prominent mixed radiolucent/radiopaque expansile mass affecting the right body of the mandible

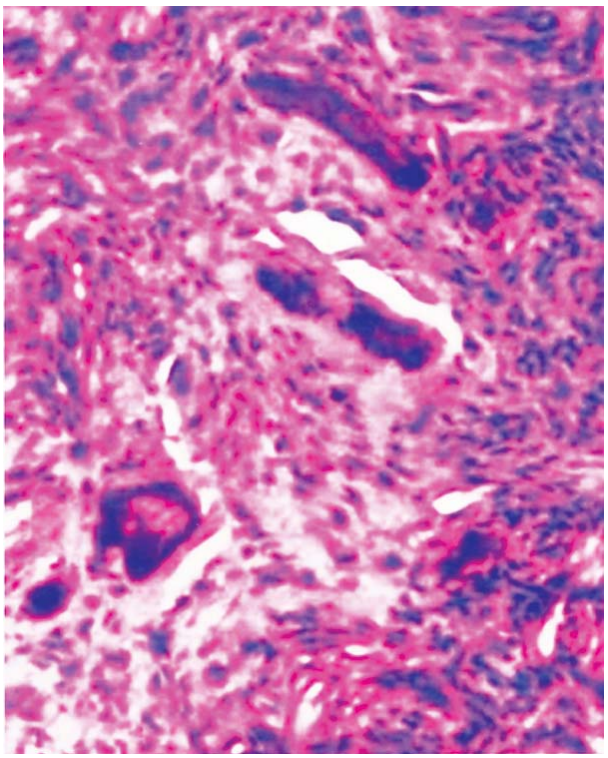

Fig. 3: Fibrocellular connective tissue composed of irregular strands of highly cellular osteoid encasing plump and irregular osteocytes (H & E, ×45)

Despite reported differences from ossifying fibroma on the basis of age of patients, most common sites of involvement and clinical behavior, the TrJOF can still be misdiagnosed as an ossifying fibroma. As aforementioned, ossifying fibroma is well encapsulated, occurs predominantly in women in third and fourth decades of life and affects primarily the mandible. Also, the tumor can be shelled out from its bed with ease.[1,3,4,6,9] Histologically, it is composed of cementum like particles that are not uniform in size and are more often than not, admixed with immature bone trabeculae.[3,6,8,9]

CONCLUSION

We have reported a case of a slowly enlarging swelling in the mandible of a 19-year-old boy. A provisional diagnosis of fibrous dysplasia was made. Biopsy and histological examination revealed uniformly fibrocellular connective tissue containing highly irregular strands of highly cellular osteoid encasing plump and irregular osteocytes. On the

basis of histological findings, a diagnosis of trabecular juvenile ossifying fibroma was made. This report emphasizes the importance of recognizing juvenile ossifying fibromas as they are clinically and histologically distinct from other fibro-osseous lesions in that JOF is more aggressive clinically and needs a more radical surgical treatment than that employed for fibrous dysplasia or ossifying fibroma and has a recurrence rate of 30 to 56%, thereby necessitating a more radical approach to treatment as compared to other fibro-osseous lesions.

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