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ODONTOAMELOBLASTOMA: A RARE CASE REPORT

Oral Pathology

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ABSTRACT

Odontoameloblastoma (OA) is an extremely rare mixed odontogenic neoplasm characterized by ameloblastomatous component and odontoma-like elements in the same tumor mass. It occurs predominantly in chidren and young adults. The majority of cases are associated with unerrupted teeth. Till date very few cases of Odontoameloblastoma have been reported in the literature on base the criteria of the current World Health Organization classification of odontogenic tumors. According to the cases reported in the English literature [9]; among 26 cases, only 5 cases have been reported involving posterior maxillary region. In this article we present extremely rare case of odontoameloblastoma in 8 year old female patient with mild swelling and facial deformity in left maxillary jaw region.

KEYWORDS

Odontoameloblastoma, ameloblastoma, Odontome

INTRODUCTION

The odontoameloblastoma belong to a group of odontogenic tumors that consists of odontogenic epithelium and odontogenic ectomesenchyme with or without dental hard tissue formation which is characterized as being extremely rare.Thoma1 introduced the term odontoameloblastomain 1970.[1]There is simultaneous occurrence of an ameloblastoma and composite odontome. It is unusual in that a relatively undifferentiated neoplastic tissue is associated with a highly differentiated tissue, both of which may show recurrence after inadequate removal. According to WHO, it is a neoplasm that includes odontogenic ectomesenchyme in addition to odontogenic epithelium that resembles an ameloblastoma in both structure and behaviour. Because of the presence of odontogenic ectomesenchyme, inductive changes take place leading to the formation of dentin and enamel in parts of tumor.[5]Here we present a case of Odontoameloblastoma in 8 year old female patient involving left side of maxilla.

CASE REPORT

An eight year old female patient came with chief complain of swelling and asymmetry of face on left side of maxilla [Figure1]. On examination the overlying mucosa was healthy with no ulceration or tenderness. Radiographic examination revealed a radio-opaque mass without any definite morphology with a distinct radio-opaque margin present in the left side of maxilla [Figure 2A]. The tooth germs of permanent teeth were missing in the area. Cone beam computer tomography showed tumor mass in left posterior maxillary region [Figure 2B]. A provisional diagnosis of complex odontoma was made. The lesion was excised under local anesthesia and the tissue specimen was sent for histopathological examination. The gross specimen was roughly spherical; approximately 2x3x 4 cm in size, reddish mixed with white in colour, hard in consistency [Figure 3].The specimen was preserved in 10% formalin and sent for histopathological examination. Hematoxyline and eosin stained sections on light microscope revealed complex odontoma with areas of odontogenic epithelium resembling stellate reticulum like areas of an enamel organ, which is surrounded by single layer of tall columnar ameloblast like cells. There are multiple small cystic degeneration present within the ameloblastic follicle [Figure 4].Based on the clinical, radiological, and histopathological findings, diagnosis of odontoameloblastoma was given.

Figure 1: Clinical photograph of patient

Figure 2A: Orthopantomograph showing radio-opaque mass in the left posterior maxillary region

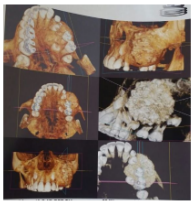

Figure 2B: Cone beam computer tomographyshowing tumor mass in left posterior maxillary regio

Figure 3: Gross tissue specimen of tumor massn

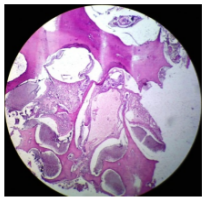

Figure 4: H and E section shows complex odontome with odontogenic epithelium of ameloblast like cells and stellate reticulum like areas

DISCUSSION

Odontoameloblastoma combines the features of ameloblastoma with those of odontoma. Several different synonyms of OA are adamantodontoma, calcified mixed odontogenic tumor, soft and calcified odontome, ameloblastic odontoma, odontoblastoma[8] The pathogenesis of OA is still unknown. About one theory suggests that an hamartamatous proliferation of mineralized dental tissue produced in response to inductive stimuli produced by proliferating epithelium over mesenchymal tissues. Other theories, suggest that both an ameloblastoma and an odontoma develop separately and form a collision tumor. This possibility seems unlikely because of the differences between these tumors with respect to age, location and

symptoms. [7][1]It frequently occurs in younger patient. Odontoameloblastomas have been characterrized as slow progressively growing lesions. They present as expansile, centrally destructive lesions. Symptoms include progressive swelling of the alveolar bone, dull pain, changes in occlusion, and delayed eruption of teeth. [1][1]The majority of tumors are associated with unerrupted teeth and commonly seen in males. [7][1]Radiographically, the OA appears as a well-defined unilocular or multilocular radiolucency containing varying amounts of radiopaque substances. The radiopaque material may be in the form of small particles (denticles representing a compound odontoma like appearance) or of a larger centrally located mass of dental hard structures with the features of a complex odontoma (which may cause divergence of roots of the adjacent teeth). Macroscopy examination shows most OAs are uncapsulated. On cut section, the lesion has a multinodular architecture with soft and hard tissue components. The amount of mineralized tissue may appear as large lobulated masses or as rudimentary teeth scattered within the soft tissue. Microscopically it consists of a great variety of cells and tissue in a complex distribution, including columnar, squamous and undifferentiated epithelial cells, as well as ameloblasts, enamel and enamel matrix, dentin, osteodentin, dentinoid and osteoid material, stellate reticulum like tissue, dental papilla, bone and cementum as well as stromal connective tissue. Many structures resembling normal or atypical tooth germs may be found, with or without the presence of calcified dental tissues. In addition, an outstanding characteristic is the presence of sheets of typical ameloblastoma of one or another of the recognized types- usually basal cell, follicular or plexiform. Isolated small foci of ghost cells occasionally be found. Few mitotic figures are present even though the proliferative tendencies of the epithelial cells are obvious.[2]

CONCLUSION

Although Odontoameloblastoma consists of features of both ameloblastoma and odontoma, its general behaviour is same as that of the ameloblastoma component, and therefore, the same philosophy of management would also apply here as for the ameloblastoma. Along with clinical and radiological, histopathology plays a major role in diagnosis of OA. Early diagnosis can prevent local invasion and neoplastic changes of this tumor which would make treatment more aggressive. This additional rare case adds to the present existing literature among odontogenic tumors of odontoameloblasstoma in posterior maxillary region.

CONFLICT OF INTEREST

None

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