ADENOMATOID ODONTOGENIC CYST: A CASE REPORT

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Introduction

The Adenomatoid odontogenic tumor (AOT) is not a tumor but rather a cyst that has an hamartomatous intraluminal proliferation of the epithelial cells derived from Hertwig epithelial root sheath. While at some times this proliferation may fill the lumen to give the impression of a solid tumor, a close inspection will reveal its emergence from an epithelial lining. The calcifications seen in these cysts, which represent attempts of the root sheath epithelium to induce root dentin, have been identified as dentinoid material. Therefore, the more appropriate term is Adenomatoid Odontogenic Cyst, AOC.[1]

The AOT was first described by Stafne in 1948 as an 'epithelial tumour associated with developmental cysts of the maxilla. He reported three cases, occurring in the anterior mandible, two related to impacted teeth (follicular AOT) and a third lesion unassociated with an unerupted tooth (extrafollicular AOT).[2]

In 1967, the term odontogenic adenomatoid tumour was suggested by Abrams et al. In 1969, Philipsen and Birn presented a review based on 76 cases of AOT, which showed the tumour to be an entity clearly distinguishable from the solid or multicystic ('classical') ameloblastoma.[3]

In 1990 Philipsen reviewed 499 cases of AOT and provided a comprehensive database on this tumour. The AOT has been divided into peripheral (extraosseous) and central (intraosseous) groups. The central lesion is further divided into follicular (related to an unerupted tooth) and extra-follicular (not related to an unerupted tooth).[4,2] In their series the central variant was the most common type, accounting for 97.2% of all lesions, 73% of which were follicular.[2] Philipsen and Reichart (1998) surveyed the literature of AOTs and identified 112 cases in which 394 (95.6%) were central and 18 (4.4%) peripheral.[5]

Clinical features

The cyst presents as an expansile lesion usually in the anterior jaws. Two thirds occur in the maxilla, two thirds occur in young women, two thirds are associated with an unerupted tooth and two thirds of those teeth are canine teeth. The discovery of the cyst on the radiograph is often incidental. At other times it may enlarge and cause pain.[1]

The appearance on a panoramic radiograph is that of a well-demarcated, unilocular radiolucency usually associated with an impacted tooth. 50% of cases will have large calcifications to appear on the radiograph. Displacement of roots of adjacent teeth is seen and only rarely is there any resorption of roots.[1]

Differential diagnosis

The cysts that appear without radiographic evidence of calcification will be more suggestive of the Dentigerous cyst. Other lesions to consider in the early years are Odontogenic keratocyst, Ameloblastic fibroma, Odontogenic myxoma and Central giant cell tumor or Ameloblastoma as the age increases beyond 14 years.

Cysts in which calcifications are seen resemble calcifying odontogenic cysts. Mixed radioopaque radiolucent lesions may include in their differential diagnosis Ameloblastic fibroodontoma, Odontogenic fibroma and beyond 14 years of age, Calcifying epithelial odontogenic tumor may be considered even though a CEOT is uncommon at any age and even less common in individuals below 25 years.[1]

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Gross morphology

The lesion readily separates from its bony crypt because it has a thick connective tissue capsule. The cut surface may show a lumen and the thick cystic lining will be rough textured like carpet piles.[1]

Histopathology

These cysts typically appear with intraluminal masses. They are very cellular with a scant fibrous stroma. Epithelial cells are bimorphic: spindle shaped cells lie in streams , whorled nodules and rosettes. Within these rosettes, eosionphilic PAS positive material is seen.

In addition, there are cuboidal to columnar cells that usually line small duct like spaces or larger cyst like spaces.

When the nuclei are polarized, it is towards the basement membrane.

Calcifications are present in varying quantities. There may be Liesegang ring formations and sometimes the calcifications resemble dentin or cementum.[1]

Treatment

Enucleation. Recurrences are not seen. The resultant bony cavity is closed in a primary fashion without using drains etc. the bony regeneration in young patients is complete in about 9-12 months.[1]

Case report

A 15 year old female reported with a painless swelling on the left side of the face which was present from 6-7 months.

Extraorally, a small diffuse swelling was seen around the ala of the nose of left side measuring approximately about 2cm x 2cm.

Intraoral examination revealed the presence of a well-defined, oval swelling in 22, 23 and 25 region. Tooth 24 was missing. The swelling was firm, non tender and measured approximately 2 cm × 2.5 cm × 2 cm. It was of same color as that of the adjacent mucosa and the surface was smooth.

Intraoral periapical radiographs of the region showed a radiolucency with a well circumscribed radio-opacity in the 24 region and the presence of an impacted tooth within it. (Fig.1)

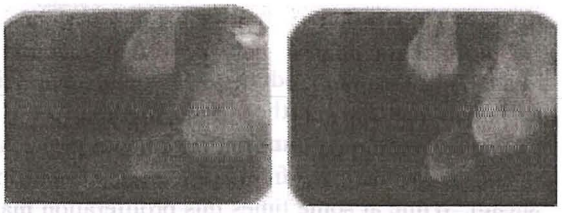


Fig.1. IOPA Radiograph showing a radiolucent lesion associated with impacted 24 tooth.

The occipitomental radiograph of skull revealed a radiolucent swelling with circumscribed radio opacity and the presence of an impacted premolar tooth within the radiolucency. (Fig.2).



Fig. 2 Occipitomental view of skull showing a cystic lesion with some calcified masses (?) in relation to tooth 24.

Aspiration done with wide bore needle was negative.

Surgery

Enucleation was performed with primary closure of the surgical site and no bone graft was used to close the defect. (Fig.3)

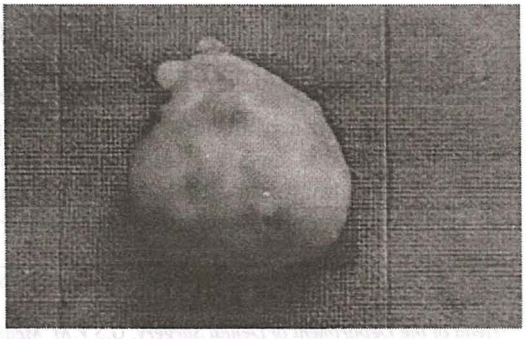


Fig. 3 Enucleated cyst.

The mass was cut open to reveal a cystic lesion with many long projections from the cyst wall into the lumen almost occluding it. The tooth present in it was identified as the first premolar. (Fig.4)

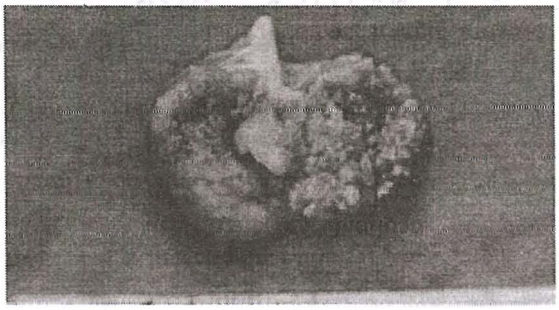


Fig.4 cut section of the enucleated cyst showing numerous intraluminal projections and tooth 24.

Histopathology report

Hematoxylin and eosin stained sections of the excised mass showed the presence of cystic cavity along with fibrous connective tissue wall. The proliferation of tumour cells was seen in the cystic cavity. Tumour cells were polyhedral to spindle in shape and arranged in varying patterns from solid islands, nests to plexiform pattern. Duct-like structures with lumina of varying size with some basophilic non-homogeneous material were present. Calcification was also present within the tumour parenchyma.

Based on the clinical, radiological and histopathological findings, the case was diagnosed as "Adenomatoid Odontogenic Cyst".

Discussion

Some authorities feel that, having the slow growth and circumscribed growth pattern of the lesion, it is best classified as a hamartoma rather than a true neoplasm. Although there is evidence that the tumor cells are derived from enamel organ epithelium, investigators have also suggested that the lesion arises from remnants of dental lamina.[6]

The presence of the impacted tooth within the radiolucent lesion and circumscribed sclerosed bone led to the differential diagnosis of Dentigerous cyst and Unicystic ameloblastoma which were negated by the absence of any aspirated fluid.

For the most part, it is not until the tumor has been excised and examined histologically that the true nature of the lesion is known.[7]

AOCs macroscopically appear as a soft, roughly spherical mass with a distinct fibrous capsule. Upon gross sectioning, the cut surface shows a variegated appearance with small areas of hemorrhage in a grayish-white tissue. Small or large cystic spaces may be present and these may contain yellowish gelatinous material or blood-stained fluid. In some cases, the tumor may be almost entirely cystic. A tooth or teeth may be embedded in the tumor or attached to it.[8]

There is little variation in the histopathology of a peripheral AOC compared with an intraosseous AOC- proliferation of odontogenic epithelium with variable-sized duct-like structures and solid nodules of cuboidal or columnar cells that are encapsulated by thin connective tissue, with a vascular stroma and diffuse calcifications- although there is some evidence to suggest that extraosseous lesions are nonencapsulated more frequently. There is no evidence to suggest that these peripheral neoplasms arise from surface epithelium. In the present case, the lesion was well encapsulated

The treatment recommended for all AOCs is enucleation or excision. As reported in the literature, in the present case, the lesional tissue was also easily separated from the overlying mucosa. There have been no reports of recurrences, with the exception of one case reported by Takigami, in a 49-year-old female patient with a number of recurrences of a maxillary AOT.[2]

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