CASE REPORT

Adenomatoid Odontogenic Tumour

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INTRODUCTION

Adenomatoid odontogenic tumour was first described by Dreiblatt, in 1907, as a pseudoadenoameloblastoma.1 In 1948 Stafne2 considered it a distinct entity, but it was classified by others as a variant of ameloblastoma. Philipson and Binn3 proposed the name adenomatoid odontogenic tumour in 1969 and suggested that it not be regarded as a variant of ameloblastoma because of its different behaviour. This term was adopted by the World Health Organization (WHO) classification4 in 1971. Max & Stern5 coined the name Adenomatoid Odontogenic Cyst in 2003.

There are 3 variants of adenomatoid odontogenic tumour6–8 the follicular type (accounting for 73% of cases), which has a central lesion associated with an embedded tooth; the extrafollicular type (24% of case), which has a central lesion and no connection with the tooth; and the peripheral variety (3% of cases).

CASE REPORT:

A 19-year-old girl was referred by her orthodontist for evaluation of a mandibular radiolucent lesion noticed on a panoramic radiograph. (Fig. 1) to the department of oral pathology, Kamineni Institute of Dental Sciences. The medical history was insignificant and patient was in good general health. Intraoral examination showed that the labial vestibule was obliterated by expansion of the buccal cortical plate from the lower left canine and premolar region. (Fig. 1). the permanent first premolar was missing. A panoramic radiograph revealed a, well-circumscribed radiolucency around
impacted lower left first premolar (Fig. 2). The lesion produced an expansion of tissue and extended into the alveolar processes, disrupting the usual orientation of the anterior teeth. On the basis of the clinical and radiographic findings, the differential diagnosis was adenomatoid odontogenic tumour, ameloblastic fibrous odontoma, calcifying odontogenic cyst, calcifying epithelial odontogenic tumour, infected dentigerous cyst, and unicystic ameloblastoma.

The patient underwent surgery with local anesthesia. A mucoperiosteal flap in the left premolar region was reflected to expose the labial aspect of the tumour. The labial cortex was very thin and had several areas of complete resorption. The tumour was enucleated along with the impacted lower permanent first premolar (Figs. 3 and 4). The areas between the roots of the involved teeth were curetted well and the flap was sutured in place. Healing was uneventful, and there was no evidence of recurrence 1 year after the surgery.

Histopathological examination revealed sheets of polygonal cells throughout the fibrous connective tissue stroma (Fig. 5). The ductal lumina were surrounded by columnar epithelial cells and filled in some areas with eosinophilic material (Fig. 6). In other places amorphous calcified material was present. The histopathological report confirmed the diagnosis of adenomatoid odontogenic tumour.

DISCUSSION:

Adenomatoid odontogenic tumour is a slowly growing lesion, with a predilection for the anterior maxilla (ratio of cases 2:1 relative to mandible) of young females. They are diagnosed in the second decade of life, and more than half occur during the teenage years. The female to male ratio for all age groups and all variants is close to 2:1. The lesions are typically asymptomatic, but growth of the types with central lesion results in cortical expansion, as in the case reported here. The involved teeth are commonly impacted, and adjacent teeth may be slightly displaced. It can cause a painless hard swelling, as in the case reported here and can be found on routine radiographic examination. Adenomatoid odontogenic tumours, accounting for approximately 3% of all odontogenic tumours, are less frequent than odontoma, cementoma, myxoma and ameloblastoma. It has been suggested that this tumour may be a hamartoma rather than a true neoplasm, but there is currently no evidence to resolve this dispute. For cases in which the lesion appears to surround an unerupted tooth and has no radiopaque component, dentigerous cyst may also be considered in the differential diagnosis. However, an adenomatoid odontogenic tumour often appears to envelop the crown as well as the root, whereas dentigerous cysts do not envelop the roots.

The origin of adenomatoid odontogenic tumours is controversial. Some believe they originate from the odontogenic epithelium of a dentigerous cyst. In addition to the anterior maxilla, the tumour has been reported in other areas of the jaw, such as the angle of the mandible. Therefore, dental lamina remnants likely represent the progenitor cells for this benign odontogenic tumour. According to this hypothesis, the lesion grows (sometimes while forming a cystic space) next to or into a nearby dental follicle, leading to the “envelopmental theory.” In the case reported here, the lesion surrounded a fully formed premolar, which suggests “envelopmental” pathogenesis.

Recent reports indicate that the cells of an adenomatoid odontogenic tumour usually differentiate toward an apparent ameloblastic phenotype but fail to achieve further functional maturation. WHO has described the histologic features of the tumour as follows: "A tumor of odontogenic epithelium with duct like structures and with varying degree of inductive changes in the connective tissue. The tumor may be partly cystic and in some cases the solid lesion may be present only as masses in the wall of a large cyst. It is generally believed that the lesion is not a neoplasm. "The histologic appearance
of all variants is identical and exhibits remarkable consistency. At low magnification the most striking pattern is that of various sizes of solid nodules of columnar or cuboidal epithelial cells forming nests or rosette-like structures with minimal stromal connective tissue. Between the epithelial cells of the nodules and in the centre of the rosette-like configuration is found eosinophilic amorphous material, often described as tumour deposits. Conspicuous within the cellular areas are structures of tubular or duct-like appearance. A third characteristic cellular pattern consists of nodules of polyhedral, eosinophilic epithelial cells with squamous appearance and exhibiting well-defined cell boundaries and prominent intracellular bridges. These islands may contain pools of amorphous amyloid-like material and globular masses of calcified material (thus the suggestion of a combination of calcifying epithelial odontogenic tumour and adenomatoid odontogenic tumour). This same pattern was observed in our case. Another epithelial pattern has a trabecular or cribriform configuration. Occasional foci of mitotic activity can be traced. Induction of hyaline, dysplastic dentinoid material or calcified osteodentin has been described. The connective tissue stroma is very loosely structured and contains thin-walled congested vessels characteristically showing marked degenerative (fibrinoid) changes of the endothelial lining, vessel wall and perivascular connective tissue. It has been suggested recently that the tumour droplets represent some form of enamel matrix. Immunohistochemical studies of the lesion suggest expression of keratin and vimentin in the tumour cells at the periphery of the ductal, tubular or whorled structures. Amelogenin and enamelin in small mineralized foci are found in the tumour cells and in hyaline droplets.

Since all variants show identical benign biological behaviour and almost all are encapsulated, conservative surgical enucleation or curettage is the treatment of choice. Recurrence has been reported in very few cases.

**SUMMARY:**

The present case is a rare report of an adenomatoid odontogenic tumor presented in mandible of 19 year old female involving impacted first premolar and showing hybrid tumor like histopathological picture and its subsequent management.

**REFERENCES**


Figure 3: Intraoral photograph shows surgical exposure for lesion which has obliterated the vestibule.

Figure 4: Intraoral photograph shows surgical exposure for lesion which has obliterated the vestibule.

Figure 5: Histopathological slide shows features typical of adenomatoid odontogenic tumour, including sheets of polygonal cells throughout the fibrous connective tissue stroma.

Figure 6: Histopathological picture shows the ductal lumina were surrounded by columnar epithelial cells and filled in some areas with eosinophilic material.

Figure 7: Duct-like spaces seen with eosinophilic material.