Case Report

Adenomatoid odontogenic tumour in the maxilla: a rare benign tumour in an uncommon site

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ABSTRACT

We report a case of adenomatoid odontogenic tumor (AOT) in the maxilla in a boy aged 17 years. AOT is an odontogenic tumour arising from the enamel or dental lamina. AOT is rare and it represents 3–7% of all odontogenic tumors. This lesion affects young girls and is associated with an impacted tooth, usually canine. This case is presented to highlight the presentation of the tumour in a male patient and in an uncommon site, the left upper 1st molar. Differentiating this benign tumor from other lesions is difficult before surgical management and histopathological examination is important in accurate diagnosis.

Keywords: Adenomatoid odontogenic tumor, 1st molar, Follicular type

INTRODUCTION

Adenomatoid odontogenic tumour (AOT) is a rare benign neoplasm of odontogenic epithelial origin. Philipsen and Birn proposed the name Adenomatoid odontogenic tumor, a term that was adopted by the first edition of the World Health Organization classification of odontogenic tumors in 1971.^{1,2} It is a relatively uncommon, benign and slow growing tumour.³ AOT accounts for 3-7% of all the odontogenic tumours.⁴ It is usually found in young patients, especially in the second decade of life.⁵ Females are more often affected than males. AOT commonly occurs in the maxilla especially in the anterior part of the jaw than the posterior part.² The current WHO classification of odontogenic tumors defines AOT as being composed of odontogenic epithelium in a variety of histo-architectural patterns, embedded in a mature connective tissue stroma and characterized by slow but progressive growth.¹ Adenomatoid odontogenic tumor can be associated with an impacted tooth. The origin of the tumour remains controversial but according to the

envelopmental theory they originate from the odontogenic epithelium of a dentigerous cyst.⁶

CASE REPORT

A 17 year old boy presented with a painless swelling on the left side of face for the last 6 months which was progressively increasing in size for the past 2 months. There was no history of trauma, pain or discharge from the lesion. He also had complaints of ipsilateral and persistent nasal obstruction for 2 months.

The swelling was of size 4 × 3 cm, on the left infra orbital and malar region and was hard in consistency. The swelling extended from the lower part of left naso-labial fold to middle of zygomatic arch horizontally and from the inferior orbital margin to the level of line of nares vertically. The mass had elevated the dorsum of the nose and medialised the lateral wall on the ipsilateral side leading to nasal obstruction. The swelling was hard and smooth with no local rise of temperature. It was not

tender. Skin over the swelling was normal and pinchable with no punctums or scars. The margins of the swelling were ill defined. Examination of the oral cavity showed no abnormalities.

Figure 1: Preoperative picture of the patient.

Figure 2: CT scan showing radiolucent lesion.

Figure 3: Operative procedure. a) Caldwell Luc incision b) mucoperiosteal flap elevation c) infraorbital nerve identification d) anterior wall of maxillary sinus opened e) releasing attachment on all sides f) cyst curetted out after removal of cyst g) cavity h) gross specimen.

Radiography showed a unilocular cystic, mass in the left superior alveolar margin encroaching the maxillary sinus with an unerupted 1st molar tooth within. The margins were well defined and osteosclerotic. A 3.7 × 2.5 cm sized radiolucent lesion containing a tooth within noted arising from the left anterolateral superior alveolar margin encroaching into the left maxillary sinus. There was resultant mass effect over the left nasal cavity with partial obliteration of anterior naris and anterior part of inferior meatus and soft tissue bulge caused by the lesion in the left maxillary region.

The surgical treatment given was enucleation of the cyst and curettage under general anaesthesia through a Caldwell Luc approach. The cyst was removed in toto from the maxillary sinus along with the teeth within.

The gross specimen was a single globular tissue mass of size 4×3×1.5 cm. It was grey brown in colour and a tooth was seen embedded in the soft tissue mass. The cut section showed firm solid areas in between.

Figure 4: Cut section of the specimen.

Histopathological examination showed a well circumscribed benign epithelial tumor with strands, sheets, whorls & ductular patterns around ductular lumina and tumor cells which were round to oval with scanty cytoplasm, uniform nuclei and bland chromatin. Epithelial tumor cells surrounded by polyhedral to spindle shaped cells, scattered homogenous hyaline material and calcified deposits, congested blood vessels & areas of stromal hemorrhage were seen confirming the diagnosis of adenomatoid odontogenic tumour. The follow-up examination after one year revealed no local recurrence.

Figure 5: Histopathological examination. a) circumscribed tumour, b) cells in ductular pattern, c) Scant cytoplasm, bland chromatin.

Figure 6: Postoperative photograph.

DISCUSSION

The AOT is a benign non-invasive rare odontogenic tumour, showing slow growth which causes jaw swelling.¹² There is a slight female over male predilection which is almost 2:1 and it appears most often in the second decade of life.¹³ 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.”^{14,15}

Our patient presented in the second decade of his life which is very characteristic of this tumour. The tumors are usually unilocular and 1.5 to 3 cm in size, but larger multilocular lesions has been reported.⁶ It is debatably thought to be a hamartoma or benign neoplasm. It is seen twice more commonly in maxilla⁷ than mandible and 40% are related to the crowns of unerupted maxillary canine teeth. In our case the patient was a male and impacted tooth was an upper molar which is rare.

AOT can occur as intraosseous and peripheral forms. Intraosseous AOT may be radiographically divided into 2 types- follicular (pericoronal) and extrafollicular (extracoronal) types.⁸ This type is usually discovered during routine radiographic examination.⁵ The extraosseous (peripheral/gingival) type of AOT is rarely detected radiographically, but there may be slight erosion of the underlying alveolar bone cortex.^{2,8}

The tumour here, presented as intraosseous, follicular type lesion which is the commonest type characterised by tumor manifests as a well-defined unilocular radiolucency surrounding the crown and partly the root of an unerupted tooth.

The clinical features and radiological findings are more or less the same for different slow lesions affecting the area. A differential diagnosis of dentigerous cyst, odontogenic keratocyst, ameloblastoma, odontogenic myxoma was considered. The cut section can be used to

assess the contents. It is mucoid in myxomas, paste like in odontogenic keratocyst and dentine in odontomas. In this case, differentiation is mainly based on histology especially the nature of epithelium.

The most striking pattern is various sizes of solid nodules of columnar or cuboidal epithelial cells forming nests or rosette-like structures with minimal stromal connective tissue. Between these characteristic eosinophilic amorphous material is found. Within the cellular areas tubular or duct-like appearance, nodules of polyhedral, eosinophilic epithelial cells with squamous appearances, pools of amorphous amyloid-like material and masses of calcified material are also seen.

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.^{9,10} This type is often mistaken clinically as dentigerous cyst or odontogenic keratocyst because of the predominant cystic appearance here.¹¹ Conservative surgical treatment is enucleation and curettage. It is curative and recurrence even after incomplete removal is rare.

CONCLUSION

Adenomatoid odontogenic tumour (AOT) is a benign odontogenic jaw lesion. The occurrence of tumour in male patient from the upper first molar is noted. CT scan is mandatory for the diagnosis of such lesions which can easily be mistaken for a dentigerous cyst or odontogenic keratocyst. Definitive diagnosis is mostly postoperative after histopathological diagnosis. The treatment given in this case, enucleation and curettage is satisfactory and recurrence is rare.

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