CASE REPORT

Adenomatoid odontogenic tumour in a 20-year-old woman

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SUMMARY

Adenomatoid odontogenic tumour is a relatively rare and distinct odontogenic tumour that is exclusively odontogenic epithelium in origin. It comprises 3% of all odontogenic tumours. This report describes the surgical therapy, clinical course and morphological characteristics of an adenomatoid odontogenic tumour that developed in the left maxilla of a 20-year-old patient.

BACKGROUND

Adenomatoid odontogenic tumour, rightfully called the master of disguise, was first documented in the literature by Steen lands epithelioma adamantium.¹ Adenomatoid odontogenic tumour with its simple abbreviation AOT is the most widely accepted terminology proposed by Philipsen and Birn.² According to WHO histological typing of odontogenic tumours, jaw cyst and allied lesions, AOT is defined as a tumour of the odontogenic epithelium with duct-like structures and with varying degrees of inductive changes in the connective tissue. The differential diagnosis of AOT is crucial in terms of surgical management. Local excision is adequate to free the patient from AOT. AOT is estimated to constitute about 2.2–7.1% of odontogenic tumours, as reported in a recent study.³ This report describes the surgical therapy, clinical course and morphological characteristics of an adenomatoid

odontogenic tumour that developed in the maxilla of a 20-year-old patient.

CASE PRESENTATION

A 20-year-old female patient was referred to our clinic with a nine-month history of swelling on the left side of her face. The swelling was slow in onset and gradually increased in size leading to disfigurement of the face. The patient gave a history of bleeding from left nose associated with pain and nasal obstruction. There was no history of trauma, fever or pus discharge related with swelling. The medical and family history was unremarkable. There was no sign of general lymphadenopathy. Extraorally, there was facial asymmetry due to a solitary, diffused nodular swelling of about 4×6 cm on the left maxillary region of the face (figure 1A). On palpation of the affected area, the swelling was slightly tender, firm to hard in consistency, fixed to the underlying bone, free from the overlying skin.

Intraorally, a solitary swelling was situated in the left maxillary region measuring about 6×5 cm, extending from marginal gingiva of the left lateral incisor to distal to the first molar on the same side, obliterating the vestibule mediolaterally. The margins were diffuse and the surface mucosa was normal. On palpation, swelling was non-tender, hard in consistency towards the gingival margin and firm towards the buccal vestibule (figure 1B). There was expansion of the buccal cortex in

Figure 1 Extraoral photograph showing a diffuse swelling (left side of image) and intraoral lesion in left canine–premolar area (right side of the image).

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Figure 2 Orthopantomogram showing small radiopaque masses with missing permanent canine in the left side of the maxilla.

relation to the second quadrant with no sign of expansion of cortex on the palatal side. Upper left premolars and first molar were depressible in the socket with grade III mobility and were vital. The deciduous canine was over-retained and had no signs of eruption of permanent canine.

INVESTIGATIONS

Considering all clinical details such as missing teeth, patient's symptoms, radiographs such as orthopantomogram (OPG), Water's view and CT were performed to locate the missing permanent maxillary canine and to know the nature and extension of the lesion. Initially OPG was performed to locate the missing tooth and OPG revealed a large unilocular radiolucency measuring about 5×6 cm extending from distal surface of maxillary left lateral incisor to the distal surface of maxillary second molar of the same side. It was irregular in shape, with well circumscribed and corticated margins (**figure 2**). The findings of OPG were inconclusive; Water's view was advised by the radiologist. Interestingly, Water's view showed that the permanent canine was displaced just below the floor of the orbit placed mediolaterally and completely enveloped by the tumour (**figure 3A**). The extent and the magnitude of the lesion could not be determined by both radiographs; CT was advised by the operating oral and maxillofacial surgeon. The lumen of the lesion revealed numerous fine snowflake-like calcifications in the CT (**figure 3B**). The roots of the premolars and first molar showed signs of severe root resorption.

Routine blood and urine investigations were advised and found to be normal.

Incisional biopsy of the lesion revealed scanty connective tissue stroma composed of duct-like structures having a central

Figure 4 Photograph showing surgical specimen along with impacted tooth.

space surrounded by a layer of columnar to cuboidal epithelial cells. The cells also showed a rosette pattern of arrangement.

DIFFERENTIAL DIAGNOSIS

On the basis of the clinical findings a provisional diagnosis of AOT was given and the following other lesions were thought of while listing the differential diagnosis: calcifying odontogenic cyst, central ossifying fibroma, ameloblastoma, calcifying epithelial odontogenic tumour, central cementifying fibroma.

TREATMENT

The surgery was performed under general anaesthesia. The left antrum of Highmore was exposed via a fenestration of the facial aspect of the maxilla. The bone appeared to be thinned to a parchment-like layer but was intact. Below the bone, a reddish, bulky layer of a cystic tumour became evident. The cyst filled the sinus completely and was consecutively excavated. There were no apparent infiltrations of the surrounding bones. The retained canine was completely embedded in the cyst (**figure 4**). Histological examination of the excisional tissue showed islands of odontogenic epithelium with duct-like and sheets of spindle-

Figure 3 Water's view and CT scan showing impacted maxillary permanent canine (arrows) with multiple, small calcifications.

Figure 5 Photomicrograph showing sheets of spindle-shaped cells arranged in a ductal pattern (H&E stain $\times 50$).

shaped cells arranged in a rosette-like pattern (figures 5 and 6). The underlying connective tissue showed collagen fibres arranged in bundles with areas of foci of calcification scattered in the stroma between the tumour cells which may be regarded as attempts of enamel /dentin/cementum formation (figure 7). From the clinical, radiological and histological findings, a final diagnosis of AOT was made.

OUTCOME AND FOLLOW-UP

The oral defect was closed by primary intention. The healing was uneventful without any complications and the patient is disease free after 6-month follow-up (figure 8). The patient reported swelling and bleeding from left nose associated with nasal obstruction were solved after the surgery (figure 9).

DISCUSSION

AOT is a benign, hamartomatous, non-aggressive lesion with slow but progressive growth. The pathogenesis of AOT is unknown. The cell of the origin was suggested as entrapped epithelium in the line of fusion by Stafne (1948); odontogenic epithelium, outer enamel epithelium, inner enamel epithelium, cell rests of Malassez, dental lamina or its remnants.^{4 5}

Figure 6 Photomicrograph showing tumour cells in ductal and rosette pattern.

Figure 7 Photomicrograph showing areas of foci of calcification scattered between the tumour cells (inset for different area of calcification).

It is mainly of intra-osseous nature of about 96% and has two variants, the follicular type (71%) and extra-follicular type (29%). In the follicular type, permanent teeth are invariably involved. Anterior maxillary gingiva is the most common site for peripheral variant. Radiographically, AOT presents as a radiolucent area which is presented by a well-defined/circumscribed and generally associated with a canine tooth in the anterior maxillary and the presence of calcifications within the lesion. The follicular subtype of AOT appears as a radiolucent lesion associated with the crown and often seen with part of the root of an unerupted tooth. A large central lesion results in cortical plate expansion and displacement of neighbouring teeth. Root resorption is rare. The peripheral variant shows only slight erosion of the alveolar bone crest.⁶⁻⁹

Figure 8 Postoperative photograph showing the disease-free lesional site.

Figure 9 Extraoral photograph showing the nasal obstruction (left side of the image,(A)) before the surgery and normal appearance left nostril (right side of image,(B)).

Microscopically AOT is composed of spindle epithelial cells forming nests, cords or cell masses in a little fibrous stroma. Rosettes can be formed from the epithelial cells around a central space that may be empty or containing eosinophilic material. Duct-like structures can be prominent, sparse or even absent and constitute a central space, surrounded by cuboidal or columnar cells with peripheral nuclei. Small focus of calcification may be scattered throughout the tumour, being interpreted as abortive formation of enamel/dentine/cementum.^{1 6}

As the tumour is well encapsulated and benign in nature, conservative surgical enucleation is the treatment of choice.^{4 6} In

large lesions, the adjacent teeth may have to be extracted because of the lack of supporting bone. In cases of lesions associated with the tooth favouring orthodontic movement, marsupialisation or decompression of the lesion is recommended as the initial modality of treatment. Recurrence is very rare (0.2%).¹⁰

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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Learning points

- The adenomatoid odontogenic tumour (AOT) shows a slow and progressive growth, asymptomatic, usually accompanied by varying degrees of facial asymmetry associated with an unerupted tooth.
- In the present case, rare presenting features of epistaxis, nasal obstruction, resorption of roots of adjacent teeth with calcifications radiographically mimicking a fibro-osseous lesion were noted.
- The definitive diagnosis of AOT was given only after histological examination of the lesion.
- The benign behaviour, presence of capsule or clear demarcation of the lesion allow easy and complete removal, and low rates of recurrence justify a conservative surgical approach with complete enucleation of the lesion as the treatment of choice.

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