An atypical adenomatoid odontogenic tumour in the mandible: a report of a paediatric case

Kiran N. Devakalavar, Amit Bali, Jatinder P. Singh Chawla and Pavneet K. Pandher

An adenomatoid odontogenic tumour (AOT) is a rare odontogenic tumour that is often misdiagnosed as an odontogenic cyst. To acquire additional information about AOT, all reports regarding AOT that had been cited in 'Pub Med' since 1990 onwards were reviewed. AOT accounts for about 1–9% of all odontogenic tumours. It is predominantly found in young and female patients, is located more often in the maxilla, and in most cases is associated with an unerupted permanent tooth. The differential diagnosis between AOT and other odontogenic tumours such as ameloblastoma should be well made to avoid extensive ablative surgery. However, AOT frequently resembles other odontogenic lesions such as dentigerous cysts or ameloblastoma. Immunohistochemically, AOT is characterized by positive reactions with certain cytokeratins. For illustration a rare case of an AOT in the mandible is presented that had atypical findings such as buccolingual cortical perforation and resorption with displacement of adjoining teeth. ***Ann Pediatr Surg*** **14:36–38 © 2018 Annals of Pediatric Surgery.**

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Introduction

Odontogenic cysts and tumours form a diverse group of lesions that show deviation from normal odontogenesis. The first case that met the diagnostic criteria was reported by Steensland in 1905 as 'epithelioma adamantinum'. The first description of the lesion was given by Dreibaldt in 1907, who called it a pseudoadenoameloblastoma [1]. Philipsen and Reichart [1] introduced the term adenomatoid odontogenic tumour in 1969, which was adopted by WHO in their 'Histological Typing of Odontogenic Tumours, Jaw Cysts and Allied Lesions' and defined it as 'A tumour of odontogenic epithelium with duct-like structures and varying degrees of inductive change in the connective tissue'. The tumour may be partly cystic, and in some cases the solid lesion may be present only as masses in the wall of the large cyst. An adenomatoid odontogenic tumour is often discovered incidentally during a routine radiographic examination for dental treatments. We have presented a case of a unique adenomatoid odontogenic tumour associated with an impacted mandibular canine with root resorption and cortical bone perforations.

Case report

A 15-year-old girl reported to the department of oral and maxillofacial surgery with a chief complaint of painless swelling in the lower left front region of the face since 1 month. Extraorally a diffuse swelling in the mandibular symphysis area extended from midline to ~4 cm towards the angle of the mandible (anteroposteriorly) and from the lower lip to the inferior border of the mandible (superoinferiorly); the overlying skin was normal (Fig. 1). The swelling was soft to firm in consistency. Intraorally the swelling extended from the lower left central incisor to the left first molar with diffused margins (Fig. 2). The overlying mucosa was normal except in the centre of the swelling due to aspiration of the lesion. The buccal cortical plate was enlarged, leading to obliteration of the buccal vestibule; the lower canine and premolars were displaced lingually. The radiograph (occlusal view) revealed radiolucency associated with an impacted canine with buccolingual cortical expansion and foci of calcifications directing diagnosis towards a dentigerous cyst (Fig. 3). The orthopantomogram revealed a unilocular radiolucency in the left mandible surrounding the impacted canine extending from midline to the mesial root of the first molar. There was displacement of the roots of the adjacent anterior (31,32,73) and premolars (34,35) associated with root resorption of the first premolar along with areas of radiopacities (Fig. 4). Noncontrast computed tomography of the face revealed a cyst of 2.4 cm (superoinferiorly) × 3.6 cm (anteroposteriorly) × 3.2 cm (mediolaterally) with thickened cystic lining and calcifications along the periphery (Fig. 5). There was thinning and erosion of the buccal and lingual cortices with full thickness defect in the buccal and lingual cortices of the mandible (Fig. 5). Pulp vitality test of the teeth involved showed delayed response. The patient was prepared under general anesthesia and the lesion was completely enucleated. Intraorally, the impacted tooth was removed together with the lesion. The retained deciduous canine was extracted and primary closure of the wound was done with mersilk 3-0. The specimen was sent to the laboratory for histopathological examination. Gross pathology revealed creamish white to brown colour, soft to firm consistency, and granular inner surface. Histomorphological examination (Fig. 6) showed the presence of a single large cystic space and odontogenic epithelium in scanty connective tissue stroma surrounded by a thick fibrous capsule. The odontogenic epithelium was in sheets, in ductal and convoluted/whorled patterns. The ductal patterns were peripherally lined by a single layer of ameloblast-like cells with nuclei showing reverse polarization and clear cystic

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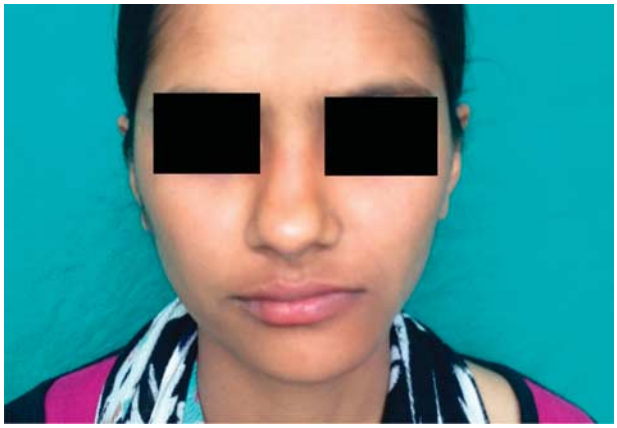
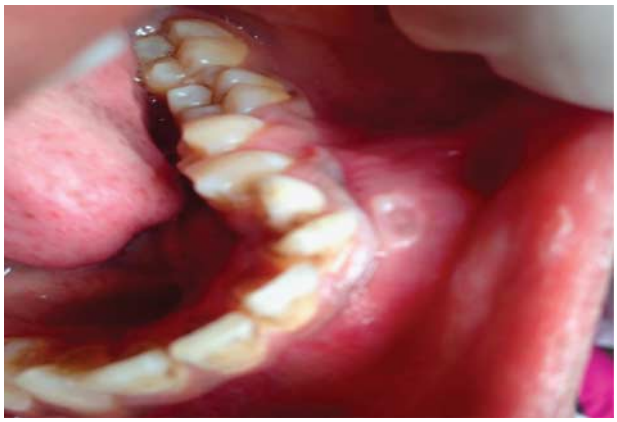
Fig. 1



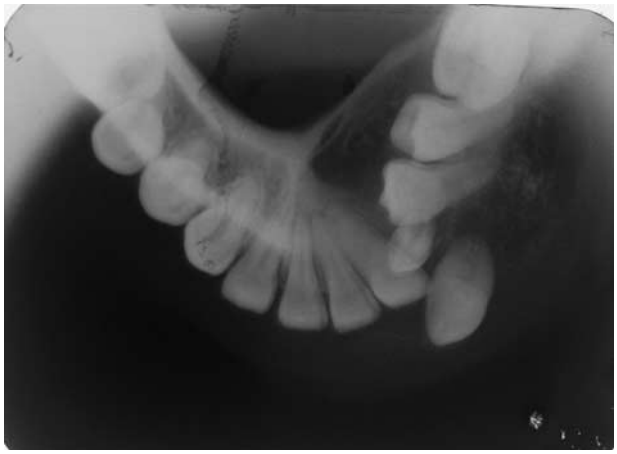
Front profile showing swelling in the left parasymphysis region.

Fig. 2



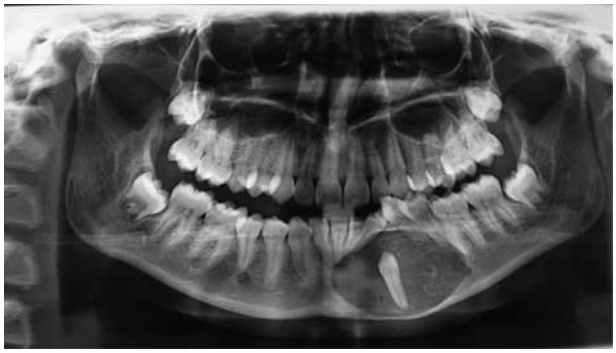
Clinical presentation of the intraoral swelling, revealing displacement of adjacent teeth, absence of the left permanent canine with retained deciduous canine, and mucosal changes associated with aspiration of fluid.

Fig. 3



Radiograph (occlusal view) revealing radiolucency associated with impacted canine with buccolingual cortical expansion and foci of calcifications directing diagnosis towards a dentigerous cyst.

Fig. 4



Orthopantomogram showing a well-defined intraosseous radiolucent lesion enclosing the canine completely, displacing roots of the mandibular central, lateral incisors and left first and second premolar with occasional root resorption of the first premolar. Calcifying nodules can also be distinguished within the radiolucent area.

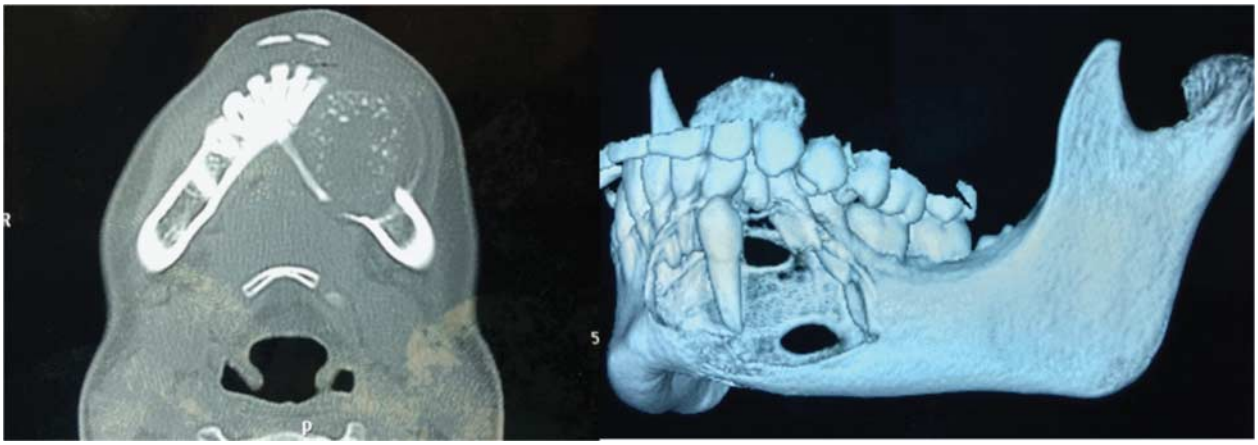
spaces. The convoluted patterns show spindle-shaped cells surrounded by amorphous eosinophillic material. The histopathological diagnosis was adenomatoid odontogenic tumour.

Discussion

Adenomatoid odontogenic tumour is often associated with impacted teeth, frequently canines or lateral incisors [1–3]. It shows a female predominance, and occurs most often during the second and third decades of life. The maxilla is the preferred site, almost twice as often as the mandible, and the anterior region is affected more frequently than the posterior area [1–3]. There are three variants of adenomatoid odontogenic tumour: follicular, extrafollicular, and peripheral. The first two are intraosseous and account for 96% of all adenomatoid odontogenic tumours [1–3]. The more common variant of the two is the follicular type, which involves an impacted tooth and is often mistaken for a dentigerous cyst [1]. The extrafollicular type is not associated with an impacted tooth. The rare peripheral type occurs extraosseously and often appears as a fibroma or an epulis on the gingival tissues [1,4].

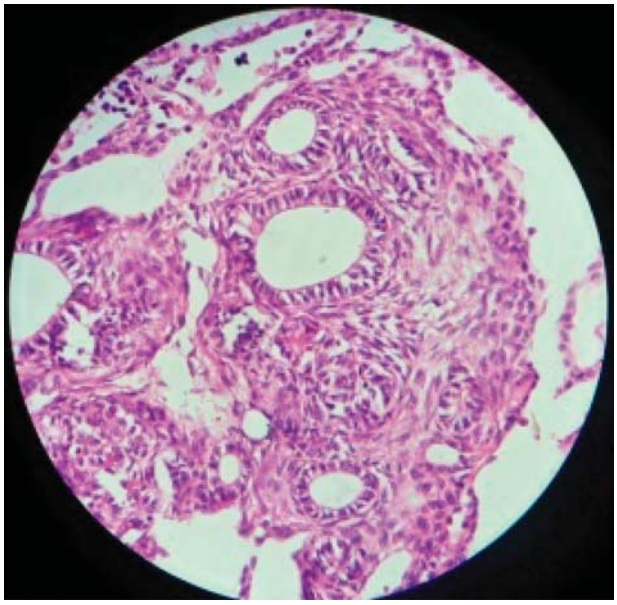
The current case of the 15-year-old girl was concordant with the occurrence of most adenomatoid odontogenic tumours, which occur during the second decade of life (69% of cases), usually in the 13–19-year age group [4], or sometimes during the third decade [2]. The current case was typical of adenomatoid odontogenic tumour with respect to age and sex. Also, in our case the tumor was located in the left mandibular anterior region, which is a rare though previously described location. Radiographically, the adenomatoid odontogenic tumour is a well-defined lesion, associated with the crown of an impacted tooth, and commonly extends in the apical direction along the route. This feature is useful to differentiate the adenomatoid odontogenic tumour from a dentigerous cyst, as the dentigerous cyst often covers only the crown of the affected teeth; further, in about 50% of adenomatoid odontogenic tumours, radiography reveals some calcified foci, whereas dentigerous cysts are completely radiolu-

Fig. 5



Noncontrast computed tomography of the face, revealing cyst with thickened cystic lining and calcifications along the periphery. There was thinning and erosion of the buccal and lingual cortices with full thickness defect in the buccal and lingual cortices of the mandible. Note the impacted mandibular canine in the cavity.

Fig. 6



Histopathological microscopic view showing duct-like structures composed of regularly single-layered or double-layered cuboidal cells and duct-like structures composed of columnar epithelial cells characteristic of adenomatoid odontogenic tumour.

cent [5]. In the present case, panoramic radiography showed a radiolucency around the impacted canine with buccal and lingual bone perforation; this association with buccal and lingual cortical perforation has been reported in the literature in very rare cases [5,6]. The lesion was associated with divergence of adjacent roots, and their resorption, which is not present normally [4].

The treatment proposed for this case was conservative surgical enucleation and removal of impacted tooth; this procedure has been described in other studies as well [3,5,7,8]. More invasive treatment, such as partial block resection, should be indicated only in exceptional cases of large tumours or when there is a risk for bone fracture [5]. Marsupialization of adenomatoid odontogenic tumours associated with dentigerous cysts is another option, which permits preservation of the teeth involved, followed by orthodontic repositioning [2,4,6,9]. Conservative surgical enucleation or curettage is the treatment of choice for all types of adenomatoid odontogenic tumours. The prognosis for adenomatoid odontogenic tumours is good, and recurrence is extremely rare after complete excision.

Conflicts of interest

There are no conflicts of interest.

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