CASE REPORT

Adenomatoid odontogenic tumour in mandible in a 14-year-old boy

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SUMMARY

A 14-year-old boy reported with a painless swelling over the right anterior mandible with missing right canine tooth. The lesion was diagnosed as 'central follicular adenomatoid odontogenic tumour' and excised surgically under general anaesthesia. The patient was on a year-long clinical and radiographical follow-up.

BACKGROUND

Adenomatoid odontogenic tumour (AOT) is an uncommon distinct neoplasm accounting only for 2.2–7.1% of all odontogenic tumours.[1] It is a benign (hamartomatous), non-invasive lesion with a slow but progressive growth. In 1999, Philipsen and Reichart[2] presented a review based on reports published until 1997, which showed some interesting aspect about AOTs. However, AOT was first described by Steensland[3] in 1905. Variety of names like adenoameloblastoma, ameloblastic adenomatoid tumour, adamantinoma, epithelioma adamantinum or teratomatous odontoma have been used in the literature for AOT.[4] According to Philipsen and Reichart,[2] AOT appears in three clinical-topographic variants: follicular (71%), extra-follicular (23%) and peripheral (4%), both follicular and extra-follicular occur intrabony.[1]

The lesion usually presents as an asymptomatic swelling, which is slowly growing and often associated with an unerupted tooth. However, the rare peripheral variant occurs primarily in the gingival tissue of tooth bearing area. It is most commonly diagnosed in second to third decade of life (mean age 13.2 years). Females are two times more commonly affected. Anterior maxilla is most commonly affected site (maxilla : mandible 2 : 1).[5]

Radiographically, follicular variant shows well-circumscribed unilocular radiolucency associated with the crown and often part of the root of an unerupted tooth. The radiolucency of the extra-follicular type is located between, above or superimposed upon the roots of erupted permanent teeth.[2] Displacement of neighbouring teeth due to tumour expansion is much more common than the root resorption. The peripheral lesion may show some erosions of the adjacent cortical bone.[6]

We present a rare occurrence of follicular type of AOT in a 14-year-old boy in anterior mandibular region. The lesion is usually more common in females and occurs two times more frequently in maxilla than in mandible.

CASE PRESENTATION

A 14-year-old boy reported with a painless swelling on the right side of the lower jaw, which had increased in size gradually over the previous of 2 months duration. Clinical examination revealed a bony hard swelling obliterating lower right labial vestibule in region of 42, 41, 31, 32, 34 and 35. 43 was clinically missing. No decayed or non-vital tooth was present. OPG revealed uniform, unilocular, well-circumscribed radiolucency in region of 33, 32, 31, 41, 42, 44 and 45 with a radio-opaque mass in it that looked like a tooth (43). 31, 41, 42, 44 and 45 were seen to be displaced (figures 1 and 2).On the basis of the clinical and radiographic findings along with the past records, provisional diagnosis of dentigerous cyst was made. However, absence of fluid during fine-needle aspiration suggested the lesion to be mural (figures 3 and 4).On the basis of all these findings, provisional diagnosis of 'central follicular AOT' was made and excisional biopsy was performed under general anaesthesia. Though no bone graft was put, patient showed excellent resolution of bony defect that was left after removal of the tumour. No signs of recurrence were seen clinically as well as radiographically during the follow-up (figures 5 and 6).

Excised tumour was approximately 5×3×2 cm in size. Longitudinal cutting of the excised specimen showed well-capsulated mural growth of soft tissue with a tooth (43) in it. H&E stained sections showed multinodular growth of polyhedral to spindle cells with a thick fibrous connective tissue wall. Tumour cells were arranged in varying patterns like solid islands and nests to thin anastomosing strands (plexiform pattern). Duct-like structures with lumina of varying size with some basophilic non-homogenous material were present. These duct-like structures were lined by a single layer of ameloblast-like cells with an eosinophilic rim inside. Eosinophilic material was seen in between the thin anastomosing strands of tumour cells as well as in the duct-like structures. Some foci of

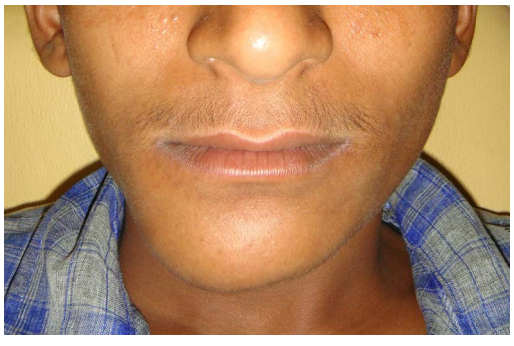

Figure 1 Preoperative photograph of patient.

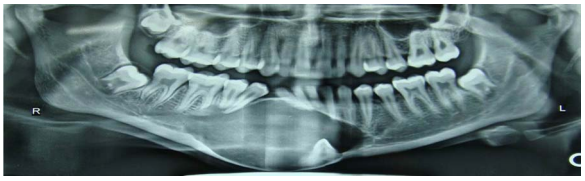

Figure 2 Preoperative OPG.

calcification were also present within the tumour parenchyma (figure 7).

On the basis of the clinical, radiographical and histopathological findings, the lesion was diagnosed as 'follicular AOT'

INVESTIGATIONS

Fine-needle aspiration expressed no fluid suggesting the lesion being mural.

Excisional biopsy was performed and histopathological analysis gave the diagnosis of the lesion being 'AOT'. Along with clinical and radiological features, the lesion was finally diagnosed to be 'central follicular AOT'

DIFFERENTIAL DIAGNOSIS

- Dentigerous cyst
- Odontogenic keratocyst
- Calcifying odontogenic cyst
- Calcifying epithelial odontogenic tumour

TREATMENT

The lesion was surgically removed under general anaesthesia. No postsurgical complications were faced.

OUTCOME AND FOLLOW-UP

The patient was followed-up for 6 months, postsurgically every 2 months. He showed excellent resolution of the bony defect that was left behind after the surgery, and no signs of recurrence were seen.

DISCUSSION

AOT, according to the second edition of the WHO's 'Histological Typing of Odontogenic tumours', is defined as 'a tumour of odontogenic epithelium with duct-like structures and with varying degrees of inductive change in the connective tissue'.[7] It is a slow growing lesion that occurs more commonly in the anterior maxilla and is rarely reported in mandible. The lesion has a clear female predilection. Usually it does not exceed 1–3 cm in the greatest diameter, but can be larger also. The tumour described in the case report here sized unusually larger (5×3×2 cm), which indicates the described lesion to be rare. AOT is most frequently associated with a missing permanent tooth (maxillary canine to be most frequent). Adjacent teeth are displaced by the growing tumour. Both these features also are seen in the presented case. Root resorption is rare with AOT.[8 9]

The radiographic features of AOT many times resemble other odontogenic lesions such as dentigerous cysts, calcifying odontogenic cysts, calcifying odontogenic tumour, ameloblastoma, odontogenic keratocysts and periapical diseases.[10] Apart from classical unilocular well-circumscribed radiolucency, AOT also shows some small radiopaque foci (apart from the tooth embedded in the tumour mass) indicative of small calcifications in tumour mass.[11 12] If there are no flacks of radio-opacity, the lesion is more likely to be a dentigerous cyst. Case presented here shows presence of tooth embedded in the tumour mass in an OPG. However, the flacks of radio opacity were better appreciated with an intraoral periapical of the affected region.

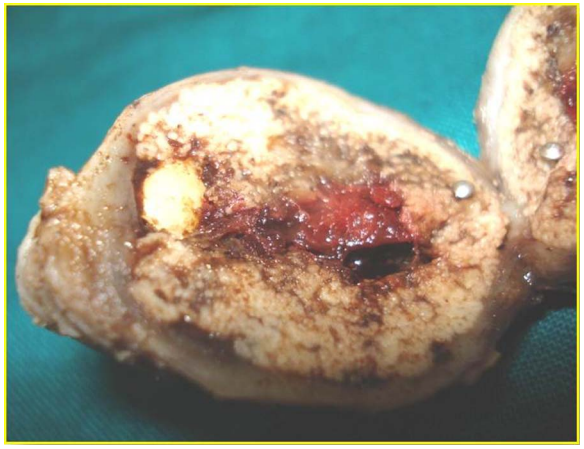

Figure 4 Tissue during grossing showing tooth in it.

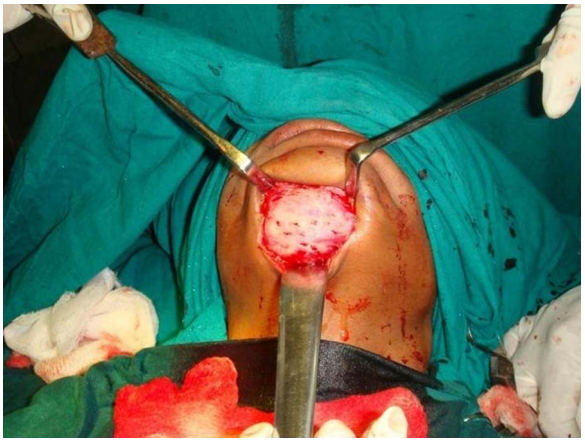

Figure 3 Exposed lesion after incision and reflection of overlying tissues.

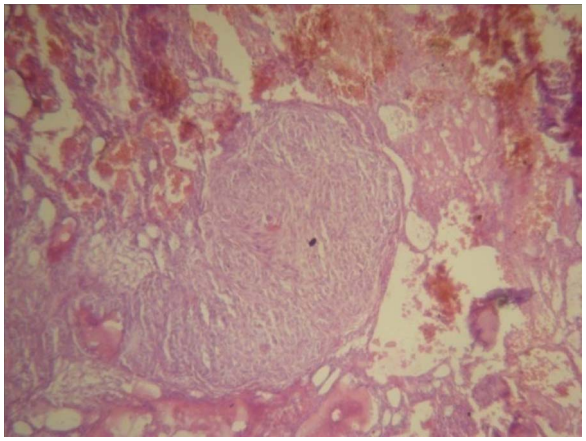

Figure 5 Six months postoperative photograph of the patient.

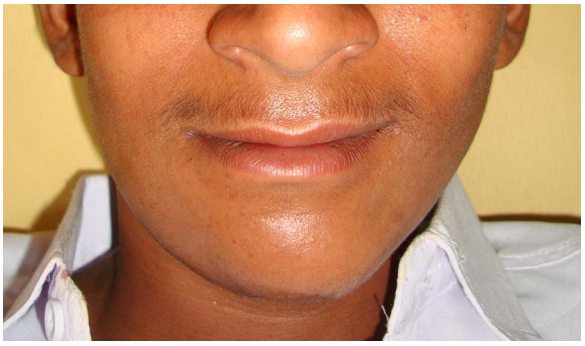

Figure 6 Six months postoperative OPG of the patient.

The origin of this tumour is controversial. The dental lamina remnants are likely to represent the progenitor cells as it not only arises from anterior maxilla but also, as seen in this case, in the body of mandible. According to a hypothesis, the lesion grows next to or on to a nearby dental follicle leading to envelopmental theory.[13] In this case, the lesion surrounds a fully formed permanent mandibular first premolar.

All the variants of AOT show identical histology. WHO defines AOT as a tumour of odontogenic epithelium with duct-like structures and with varying degrees of inductive changes 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 a large cyst.[7] Moreover, eosinophilic, uncalcified and amorphous material can be seen, and is called 'tumour droplets'. Some tumour droplets show a homogenous matrix whereas most tumour droplets reveal electron-dense plaques.[14] In this case, tumour droplets are not seen. The tumour does not show cystic lumen as well but few microcysts were seen here.

The lesion is well-capsulated and so once excised, the recurrence is very rare. Only three cases of recurrence have been reported in Japanese patients.[15] Out of three, one woman was reported with multiple recurrences. Treatment is usually conservative, but follow-up is necessary. The patient was treated with surgical excision under general anaesthesia and then was followed up for 1 year. During this phase, the defect left after the treatment resolved well and no signs of recurrence were seen.

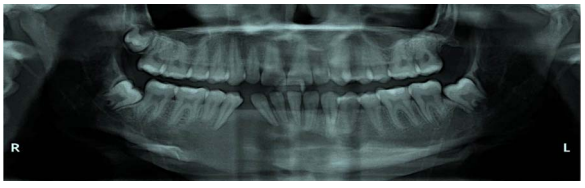

Figure 7 Low-power microscopic picture of lesion showing duct-like structures of varying sizes. Ameloblast-like cells arranged in different types of patterns (islands, nests, whirls, etc).

Learning points

- Adenomatoid odontogenic tumour (AOT) even though being more common with maxilla, may occur in the mandible too.
- It can sometimes grow to an aggressively large size as described in this case report.
- Clinical and radiographical features of an AOT may mimic other odontogenic lesions like periapical diseases, dentigerous cysts, calcifying odontogenic cysts, calcifying odontogenic tumours. Histopathological investigation is the key to the final diagnosis.
- All the forms of AOT show identical features.
- Conservative treatment is needed for AOT and recurrence is extremely rare.
- Even large bony defects can resolve very well without any bone grafts in young patients.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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Reminder of important clinical lesson