CASE REPORT

Adenomatoid odontogenic tumour

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SUMMARY

Adenomatoid odontogenic tumour (AOT) is a benign non-invasive odontogenic tumour, having mostly a slow and sustained growth pattern. AOT is an uncommon lesion of odontogenic origin, which affects young individuals, with a female predilection and mostly occurring in the second decade. In the literature, it has been considered as a hamartoma rather than a true neoplasm because of its limited size, minimal growth potential and the lack of recurrence. We present an extrafollicular central variant of AOT with a occurrence rate of 30%, adjacent to the incisors.

BACKGROUND

Adenomatoid odontogenic tumour (AOT) is an uncommon tumour of odontogenic origin, which is usually associated with unerupted teeth or dentigerous cysts, which is usually located in the anterior region of the maxilla without pain, and represents 3% of all odontogenic tumours.¹ Although AOT has been reported in patients from 3 to 82 years of age, its predilection for young patients is well established. It is unique among odontogenic tumours and unexplainable that more than two-thirds (69%) are diagnosed between the ages of 10 and 19 years; more than half (53%) are diagnosed in teenagers; 21% are diagnosed between ages 20 and 29; and altogether, 88% are diagnosed in the second and third decades. The tumour has been diagnosed approximately twice as frequently in women than men.²⁻³ The tumour is usually associated with unerupted teeth, frequently canines or lateral incisors.⁴

Regarding its pathogenesis, the lesion originates from odontogenic epithelium (enamel organ or dental lamina remnants) with inductive influence on odontogenic ectomesenchyme and consequent production of dentinoid material.⁵ It often causes expansion of surrounding bone and displacement of adjacent teeth. However, the slow growing nature of the lesion may cause the patients tolerate the swelling for years until it produces an obvious deformity.⁶

Radiographically, the tumour usually appears as a well-circumscribed, unilocular radiolucency, which may be associated with an unerupted tooth, most often a canine.⁷

Conservative surgical enucleation has been considered as the treatment modality of choice. For periodontal intrabony defects caused by AOT guided tissue regeneration with membrane technique is suggested after complete removal of the tumour.⁸

Recurrence of AOT is exceptionally rare. Only three cases in Japanese patients are reported in which the recurrence of this tumour occurred.⁹ Therefore, the prognosis is excellent.

CASE PRESENTATION

A 19-year-old female patient came with swelling and displacement of teeth of her upper left lateral incisor and canine, since 6–7 months.

An examination revealed solitary swelling about 2×2.5 cm (figure 1), oval in shape was seen over the anterior maxillary attached gingiva region extending from free marginal gingiva of 11 and 12 superiorly till 2.5 cm and mesiodistally from upper anterior labial frenum till distal of lateral incisor, oval in shape with diffused margins. Overlying mucosa was normal in colour with firm to hard in consistency on palpation, without any tenderness. Mesial tilting of crown 22 and distal tipping of 21 was noted.

INVESTIGATIONS

Blood investigations performed prior to planning of treatment, revealed no significant finding. Bleeding and clotting time were within normal limit, also the blood sugar levels were in the normal range. HIV and hepatitis B tests gave negative finding.

Radiographic examination revealed a well-defined intrabony radiolucency with sclerotic margins, causing displacement of associated lateral and central incisor was seen in intraoral periapical radiograph (figure 2). Occlusal radiograph revealed expansion of labial cortex associated with the lesion (figure 3).

DIFFERENTIAL DIAGNOSIS

- Globulomaxillary cyst
- Lateral periodontal cyst
- Calcifying odontogenic cyst
- Calcifying epithelial odontogenic tumour

Figure 1 Clinical picture.

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Figure 2 Intraoral periapical radiograph showing radiolucent lesion in lateral approximation of central and lateral incisor.

TREATMENT

Subsequently, surgical enucleation was planned for the pathology in accordance with the small size of the lesion. Endodontic treatment of central and lateral incisor was performed prior enucleation, and composite splinting of the involved teeth and adjacent teeth was performed to protect teeth from inadvertent mobilisation during enucleation. Enucleation was performed (figure 4) and the encapsulated specimen (figure 5) was sent for histopathological examination.

Primary closure was performed and postoperative antibiotic and analgesic was prescribed. Subsequent histopathological report revealed it to be a adenomatoid odontogenic tumour. Histological picture showed a cystic lining of non-keratinised stratified squamous epithelium with proliferation into the underlying connective tissue. The lesional tissue contained islands of odontogenic epithelium with formation of rosette like structures (figure 6). The tumour islands predominantly consisted of

Figure 3 Occlusal radiograph showing displacement of associated teeth and labial cortex expansion.

Figure 4 Bony defect after enucleation of pathology.

stellate reticulum like spindle-shaped cells and tall columnar cells at places (figure 7). Some of the rosette like structures centrally showed clear cell changes. Connective tissue was fibrocellular with areas of hyalinisation, moderate chronic inflammatory cell infiltrate and marked vascularity. Spicules of dystrophic calcification were evident within the epithelial islands as well as the connective tissue.

OUTCOME AND FOLLOW-UP

Regular follow-up of the patient was performed and 12 months postoperative radiograph revealed satisfactory bone healing of the defect with no signs recurrence (figure 8). Alignment of the teeth is being accomplished with orthodontic fixed appliance.

DISCUSSION

AOT is a relatively uncommon distinct odontogenic neoplasm that was first described by Steensland¹⁰ in 1905. However, a variety of terms have been used to describe this tumour. Unal *et al*¹¹ produced a list containing all nomenclatures for AOT reported in the literatures. Many different names like adenoameloblastoma, ameloblastic adenomatoid tumour, epithelioma

Figure 5 Encapsulated excised specimen.

Figure 6 Histopathological slide showing odontogenic islands in the form of rosette like structures.

adamantinoma or teratomatous odontoma have been used before to define the lesion currently called AOT. The origin of the AOT is controversial, because of its predilection for tooth-bearing bone, it is thought to arise from odontogenic epithelium.^{2,3} AOT is a slow growing lesion, constituting only 3% of all odontogenic tumours with a predilection for the anterior maxilla (ratio 2:1) relative to mandible usually associated with impacted canine, of young females in the second decade of life.¹² The lesions are typically asymptomatic, but may cause cortical expansion and displacement of the adjacent teeth, as in the case reported here. There are three clinicopathological variants of AOT, namely intraosseous follicular, intraosseous extrafollicular and peripheral. The follicular type shows a well-defined unilocular radiolucency associated with the crown and often part of a root of an unerupted tooth, thus mimicking a dentigerous or follicular cyst. The extrafollicular type is not associated with an unerupted tooth and the well-defined, unilocular radiolucency is found between, above or superimposed upon the roots of erupted, permanent teeth, just as depicted in the case above. In approximately two-thirds of the intrabony variants of AOT, discrete calcific foci are noticeable in radiographs, unlike our case which did not show any evident calcific deposits in radiograph.² Remarkably, all variants of AOT showed identical histology, but the histological typing of the WHO defined the AOT as a tumour of odontogenic epithelium with duct-like structures and with varying degrees of inductive change in the connective tissue.¹³ Conservative surgical enucleation is the treatment modality of choice. As AOT being encapsulated tumour enucleation produce excellent results, with almost null recurrence rate noticed. The case reported

Figure 7 Photomicrograph (x400) showing rosettes predominantly composed of spindle-shaped cells and stellate reticulum-like cells.

Figure 8 One-year follow-up showing healing of the bony defect.

responded well to the conservative surgical technique with an excellent outcome. The case presented of AOT matches with the existing texts and researches that had been employed but exists as rarity in diversity.

Learning points

- ▶ When there is any displacement of the teeth, as reported in our case, radiographic investigation is an absolute necessity to find the underlying pathology.
- ▶ Our case was typical of adenomatoid odontogenic tumour (AOT) as far as age, sex, size, gross appearance and histopathology were concerned, but we found extrafollicular variety of AOT which was not associated with any impacted or supernumerary tooth.
- ▶ Any tumour of the dental origin has to be excised as it can lead to destruction of the underlying bone.
- ▶ Histopathological investigation is mandatory to arrive at the final diagnosis of the lesion.
- ▶ Platelet-rich plasma could be used to fill the bony defect for the faster bone formation.

Contributors All the authors have made significant contributions towards the article write up, diagnosis, treatment and follow-up.

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