Case Report

Adenomatoid Odontogenic Tumour Mimicking a Periapical Cyst in Pregnant Woman

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Abstract

Adenomatoid odontogenic tumours (AOT) are uncommon odontogenic lesions characterized histologically by duct-like structures derived from the epithelial component of the lesion and can be distinctly classified into follicular, extrafollicular and extraosseous variants (Neville BW, Damm DD, Allen CM, et al. Adenomatoid Odontogenic Tumor. A Text Book for Oral and Maxillofacial Pathology, 2nd edition, 621–3). Most of these tumours develop in the second or third decade of life and have a distinct predilection for women. The follicular variant accounts for 75% of reported cases (Curran AE, Miller EJ, Murrah VA. Adenomatoid odontogenic tumor presenting as periapical disease. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 1997;84:557–60) and is associated with the crown of an impacted tooth, commonly the maxillary canine. We present a rare case of extrafollicular AOT mimicking a periapical cyst that originated in a woman in her first trimester of pregnancy and enlarged rapidly thereafter. The lesion was enucleated and sent for histopathology and immunohistochemistry, which revealed AOT with a cystic component with no dependence on oestrogen or progestrone for its growth. This case of AOT introduces us to the unique variation in its presentation and the difficulty in differentiation from periapical disease of inflammatory origin. [Singapore Dent J 2010;31(1):26–29]

Key Words: adenomatoid odontogenic tumour (AOT), periapical cyst, extrafollicular, gingiva, cortex, basaloid cells

A 25-year-old female patient in her second trimester of pregnancy, presented with a swelling of 3 months duration on the right side of the face. The asymptomatic swelling first originated when she was in her first trimester and gradually increased in size with occasional salty discharge intraorally in relation to the upper right lateral incisor. The patient did not give any history of prior trauma in the concerned region.

Examination revealed a firm swelling in the labial gingiva and alveolar mucosa extending from the right central incisor up to the right first premolar with normal overlying mucosa. On percussion upper right lateral incisor and canine were grade I mobile. Occlusal radiograph showed a well-defined oval radiolucency of about 3 cm in size, apical to and between 11, 12, 13 and 14 with loss of lamina dura around the apices of 12 and 13. Dental caries was not apparent in any tooth but 12 and 13 showed negative tooth vitality (Figure 1). The roots of lateral incisor and canine were displaced with root resorption visible in the lateral incisor (Figure 2). A clinical diagnosis of a radicular cyst was established. Access opening of 12 and 13 was executed, and a calcium hydroxide dressing was given. Due to persistent wet canal of lateral incisor it was decided to obturate the lateral incisor following enucleation of the lesion in the same appointment.

As the patient was in a stable term of pregnancy the lesion was surgically enucleated under

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microscopically some sections showed presence of a cystic lining component of reduced enamel epithelium continuous with a more basaloid appearing lining epithelium that became confluent with solid tumour mass. Areas of lining epithelium showed the presence of round calcified masses. The tumour proper was composed of spindle-shaped and basaloid cells arranged in solid nests in a whorl manner with some of these islands airing the presence of duct-like structures (Figures 3 and 4). The lumen of these ducts was lined by eosinophilic hyaline rings. Microcyst formation was noted in some areas (Figure 3). Pathological diagnosis was that of adenomatoid odontogenic tumour (AOT) displaying a cystic component. Immunoreactivity to oestrogen and progesterone receptor was not detected in the

Figure 1. Occlusal view showing a well-defined radiolucent lesion in relation to roots of 11, 12 and 13 with root resorption in 12.

Figure 2. Periapical radiograph showing the same lesion in relation to roots of 13 and 14 with root displacement of 13.

local anesthesia after consent from the patient and her gynaecologist. On surgical exposure it was observed that the lesion had perforated the labial cortex over the lateral incisor and canine and was attached to the apex of the canine with extension into the maxillary sinus. The displacement of root of canine, as visible on the radiograph was evident along with root resorption of the lateral incisor. The canine was extracted along with the lesion as there was inadequate bone support after excision of the lesion. A drain was left in situ for irrigating the sinus that was removed after a week. The recovery was uneventful and healing was satisfactory.

Figure 3. Low magnification showing duct-like structures along with microcyst formation; 10x.

Figure 4. Nuclei of tumour cells lining duct-like structures; 40x.

Histopathology and Immunohistochemistry

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Figure 5. No immunoreaction for oestrogen receptor in the tumour cells.

ameloblastoma-like cells and in the odontogenic squamous epithelium (Figure 5).

Discussion

This case illustrates an extrafollicular variant of AOT mimicking a periapical lesion in a pregnant lady. Few clinico-radiological indicators including age, sex and site of the lesion could have suggested the case was a possible extrafollicular type AOT, although this subvariant is not very common.

Typically AOT is a slow-enlarging swelling rarely accompanied by fluctuation, but in our case the tumour showed a considerably rapid growth in 3 months. A distinct radio-opaque border of the unilocular radiolucency is typical of the radiographical manifestation of AOT, but the lack of an intact periodontal ligament and lamina dura in the involved teeth along with negative vitality in the lateral incisor and canine makes a more likely diagnosis of radicular cyst. The radiolucency demonstrated root displacement in relation to canine with root resorption in the lateral incisor. Kim et al in their study have shown root resorption in 53.9% cases of radicular cysts. It is not clear whether the cystic component represents pooling of the mucoid stroma due to rupture of the thin lattice-work pattern or if the tumour developed within or adjacent to a pre-existing cyst; presumably, either could occur.

In the present case, the lesion mimicked a large radicular cyst, as this was found associated with two non-vital teeth without any carious exposure or history of trauma with loss of lamina dura and pus discharge from the lateral incisor. It is possible that the negative vitality could be the result of pulp necrosis, secondary to rapid tumour expansion resulting in the pulp being devoid of blood supply. The root canal infection might be secondary to the tumour expansion. It is also possible that both lesions were independent of each other and happened to occur simultaneously.

Nevertheless, the favourable treatment of AOT is enucleation, and recurrence if any, is rare. This patient gave birth to a healthy baby girl 3 months after delivery.

Figure 6. Postoperative radiograph taken 1 year later.
later and has been on regular follow-up for 1½ years (Figure 6).

Conclusion

Our case reveals a clear association of the roots of the teeth both clinically and radiographically with the AOT in a pregnant lady, which has not been reported previously. It is important for both the surgeon and the endodontist to be aware of this variation, so as to be in a position to identify future cases of this entity and to resolve issues concerning its association with preexisting periapical disease of inflammatory origin, if any.

References