Squamous Cell Papilloma and Peripheral Adenomatoid Odontogenic Tumour: Case Report of an Unusual Association

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ABSTRACT

The squamous cell papilloma and the peripheral adenomatoid odontogenic tumour separately are well-recognized and well-established entities. However, to the best of our knowledge, the simultaneous occurrence of these lesions at the same site has not been previously described. A case of this unusual association that occurred in the anterior maxillary gingival tissue of a three-year-old Malay girl is reported here. It is probable that this is a coincidental finding and the two lesions represent a pathological 'collision' phenomenon.

INTRODUCTION

The squamous cell papilloma is a common benign tumour of the oral epithelium. It occurs most commonly in the palatal region, followed by the tongue, lip, gingiva and other intraoral sites.\(^1\) More than 50% of the cases occurred between the ages of 21 and 50 years, and the recurrence rate is 4.1%.\(^2\) However, the incidence in children has been reported as low as 7.5% and 8.0%.\(^3,4\) Squamous cell papilloma in the oral cavity shows clear association with human papillomavirus, although the aetiological role of human papillomavirus in this lesion is still unclear.

The adenomatoid odontogenic tumour (AOT) is a relatively uncommon lesion of putative odontogenic epithelial origin.\(^5-9\) It occurs predominantly in the second and third decades of life and shows a female preponderance.\(^7,9\) The usual clinical presentation is of a slow-growing swelling in the anterior maxillary region. Radiographically, it appears as a well-circumscribed, unilocular radiolucency, often associated with the crown of an unerupted anterior maxillary tooth (41.7% cases involve maxillary canines), simulating a dentigerous cyst. Two-thirds of cases reportedly contain detectable radiopacities.\(^7\) A rare extraskeletal variant has also been recognized. This peripheral lesion commonly presents as a nonspecific gingival growth, also in the anterior maxillary region.\(^10,11\) A case of an unusual association of a squamous cell papilloma and a peripheral AOT is reported here.

CASE REPORT

A three-year-old Malay girl was brought by her mother with a complaint of a painless growth involving the gingiva of her upper left deciduous lateral incisor and canine. The growth was first noticed when she was about one-year-old and since then the size had remained about the same. It occasionally bled during toothbrushing. According to the mother, because of this bleeding tendency, the child refrained from brushing the teeth in that area.

Intraoral examination revealed a pink papillary lesion arising from the gingiva surrounding the upper left deciduous lateral incisor and canine (Fig. 1). It measured approximately 2.0 x 1.5 cm, and was soft to firm with a tendency to bleed. The

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related teeth were not mobile. The upper left deciduous lateral incisor had drifted mesiopalatally whilst the upper left deciduous canine had shifted distopalatally. Oral hygiene was generally good except for the presence of mesial caries on the upper central incisors. All other soft tissues in the mouth appeared normal.

A panoramic radiograph revealed that all the permanent teeth were present and the texture of the jaw bone appeared normal. Periapical radiographs of the related teeth showed no evidence of any periapical pathology. However at the lesional site there was slight pressure resorption of the bone around the upper left deciduous lateral incisor and canine. Electric pulp testing of these teeth demonstrated that they were vital.

Based on the above clinical and radiographic findings, a provisional diagnosis of pyogenic granuloma was made. Complete excision of the lesion was done under general anaesthesia. The excised tissues were sent for histopathological examination. Review after one week showed good healing. Subsequent follow-up at 3 months after excision disclosed a recurrent papillary growth at the same site. This was again excised under general anaesthesia, and the tissue submitted for histopathological examination. In the most recent review about a year after initial treatment there was no further evidence of recurrence.

Fig. 1 Intraoral view showing gingival swelling involving the upper left lateral incisor and canine.

Fig. 2 Low power view showing the squamous cell papilloma (P) and the main adenomatoid odontogenic tumour mass (AOT). H&E. Original magnification x 7
Fig. 3 Another area of the papilloma. H&E. Original magnification x 7.

Fig. 4 Details of the adenomatoid odontogenic tumour. H&E. Original magnification x 100.
HISTOPATHOLOGICAL FINDINGS

Microscopic examination of the specimen from the primary excision showed a papillary lesion composed of parakeratinized stratified squamous epithelium thrown into folds; each of these folds was supported by a connective core (Figs. 2 and 3). Deep to this papillary lesion, a circumscribed mass of odontogenic epithelium arranged in whorls, ducts and cords, and containing interspersed calcified material was identified (Figs. 3 and 4). Eosinophilic droplets that stained positive with Periodic acid Schiff were seen within the tumour epithelium. In one area odontogenic epithelial strands containing mineralized deposits and in continuity with the overlying oral epithelium (Fig. 5) were also encountered. Away from the main adenomatoid odontogenic tumour mass, occasional odontogenic epithelial rests (Fig.6), one showing evidence of early ductal differentiation, (Fig. 7) were observed. The overall histological findings were interpreted to represent those of squamous cell papilloma and peripheral adenomatoid odontogenic tumour.

Histological examination of the recurrent papillary growth showed features of recurrent squamous cell papilloma and peripheral AOT tissues.

Fig. 5 Odontogenic strands containing mineralized deposits (arrows) and in continuity with overlying oral epithelium. H&E. Original magnification x 100.

Fig. 6 An area of odontogenic epithelial rests. H&E. Original magnification x 200.
DISCUSSION

A review of the English-language literature disclosed that synchronous occurrence of a squamous cell papilloma and a peripheral AOT at the same site has not been previously reported. This case is therefore unique in this respect. This finding is just coincidental and that the two lesions most likely represent a pathological "collision" phenomenon. The other notable finding is that this case also most probably represents the youngest patient with a peripheral AOT report to date. The youngest documented case of AOT was a four-year-old white girl who presented with a two-year history of a gingival growth in the maxillary central incisal region. For squamous cell papilloma, the youngest reported case was two years of age.

Histogenetically, the intraosseous AOT is believed to take origin from both the preameloblasts and cells of the stratum intermedium of the enamel organ epithelium. The peripheral AOT, on the other hand, is purportedly thought to originate from the basal layer of the oral epithelium. Demonstration of odontogenic epithelial proliferations in the form of islands and nests extending from the overlying oral epithelium in previous and the present case supports this view. In this respect, the peripheral AOT and squamous cell papilloma reported here share a common origin, i.e., the oral gingival epithelium but follow different and distinct morphodifferentiation pathways. While HPV has been implicated in the aetiology of the papilloma, the cause of AOT remains unknown.

Squamous cell papilloma is a slow growing benign lesion. Thirty percent of the lesions reported has existed between 1 to 10 years before removal. The current lesion had been present at that site for about two years. The related teeth were carious probably because of lack of brushing in that area as informed by the patient’s mother. In addition, the drifting of the left lateral incisor and canine mesiopalatally and distopalatally, respectively, as observed in this case, may be the result of the long standing pressure exerted by the slow growing lesion. The resorption of bone around these teeth may be attributed to this same pressure effect. Reportedly, AOT may also cause delayed eruption of the permanent tooth and regional swelling of the jaw. This possibility was explained to the patient’s parent in the subsequent review visit following histological confirmation of the nature of the gingival lesion excised.
The AOT is a benign, hamartomatous lesion that seldom recurs after complete excision. In view of this, it is likely that the small area of AOT tissue found in the excised 'recurrent growth' of this case represented residual tumour left behind after the primary excision. Whether this little AOT tissue observed is another separate or 'satellite' focus away from the main AOT mass (as was noted in the primary lesion) also needs to be considered. Odontogenic tumours with a diffuse presentation, for example, the peripheral odontogenic fibroma, has been reported. However a similar variant in the AOT has not been described. The squamous cell papilloma too if incompletely removed may recur as was observed in this case. However, about 9 months after the second excision, there was no evidence of further recurrence of both entities.

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REFERENCES


