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CASE REPORT

Adenomatoid odontogenic tumor arising from a dentigerous cyst—A case report

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Summary Adenomatoid odontogenic tumor (AOT) is a benign lesion derived from the complex system of dental lamina or its remnant. It is categorized into three variants (follicular, extrafollicular and peripheral). To our knowledge, there are only six existing cases of AOT associated with dentigerous cyst. We present an additional case from a dentigerous cyst around the crown of an unerupted canine in a 15-year-old boy. We provide histological evidence of an 'odontoma-like' area, as has been described in only one other previous case report. The clinical characteristics of our patient and the six previously reported cases are also briefly discussed. We believe that this case represents an odontogenic cyst with neoplastic development, containing both epithelial and mesenchymal components. As more cases accumulate, we will be able to study these rare lesions further whether the AOTs derived from an odontogenic cyst could represent a distinct 'hybrid' variant (separate to the three variants described thus far).

1. Introduction

Adenomatoid odontogenic tumor (AOT) is a benign odontogenic lesion. Philpsen et al. [1] subdivide this condition into three groups referred to as follicular, extrafollicular and peripheral. These variants have common histologic characteristics that indicate a common origin, as derived from the complex system of dental lamina or its remnant [1]. The follicular and extrafollicular variants account for 96% of all AOT (and 71% of these are follicular variants). The peripheral variant is the rarest, with only 18 cases reported so far [2]. The former two variants are intraosseous (central) and are more commonly found in the maxilla than in the mandible at a ratio of approximately 2:1.

The follicular variant is associated with the crown and often part of the root of an impacted (unerupted) tooth. This is most commonly the maxillary

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canine and rarely the permanent molars as well as the deciduous teeth. Therefore, the follicular variant is often initially regarded as a dentigerous cyst based on clinical and radiographic evaluation. Indeed, as many as 77% of follicular variants of AOT are clinically diagnosed as dentigerous cysts [3].

The extrafollicular variant is not associated with the crown of an unerupted tooth. Instead, this variant presents as a well-defined, unilocular radiolucency between, above or superimposed upon the roots of erupted, permanent teeth. Consequently, a clinical diagnosis of a residual, radicular, globulomaxillary or lateral periodontal cyst can be made according to the definite intraosseous location of the variant. The peripheral variant almost certainly always appears as a gingival fibroma or epulis in the anterior maxilla (88%) [1,2].

Here, we present an uncommon case of AOT arising from a dentigerous cyst around the crown of an unerupted canine in a 15-year-old boy.

2. Case report

A 15-year-old boy was referred by his family pediatric dentist for evaluation of the delayed eruption of his left upper canine and delayed exfoliation of his left upper deciduous canine. At this time, a firm, non-tender swelling was noted over the left upper buccal gingiva (Fig. 1A). The swelling was painless and not associated with either paresthesia or anesthesia. Clinical examination revealed a smooth nodule in the buccal gingiva, vestibule and palatal gingiva that extended from the left upper central incisor to the first premolar (Fig. 1A and B). Three milliliters of straw-colored fluid (and no cholesterol) were aspirated from the lesion (Fig. 1C). It was also noted that the anterior teeth were crowed and a retained left upper deciduous canine with grade III mobility were present. The left upper permanent canine could not been seen on clinical examination (Fig. 1A and B). Percussion pain was not elicited overlying any of the teeth involved and electric pulp vitality testing was unremarkable.

A panoramic radiograph revealed a well-defined, unilocular, round-shaped, and circumcoronal radiolucency over an unerupted canine. This was associated with a well-developed root formation, extending from the apex of the left upper central incisor to the distal aspect of the left upper second premolar and from the apex of the retained left upper deciduous canine to the cementoenamel junction of the unerupted left upper canine, and measured approximately 3 cm $\times$ 2.5 cm in diameter (Fig. 2A). A unilocular radiolucency with an unerupted left upper canine could also be seen on the upper left occlusal film (Fig. 2B). Based on these clinical findings, a diagnosis of dentigerous cyst was made. A small bony window of approximately 5 mm $\times$ 5 mm was made within the portion of labial plate that corresponded to the upper left central incisor. An incisional biopsy was performed and a histological diagnosis of dentigerous cyst with non-keratinized epithelial lining and fibrous connective tissue was made (Fig. 3).

Under general anesthesia, the mass was approached intraorally via a reflected mucoperiosteal flap. Bony perforation was noted over the apical

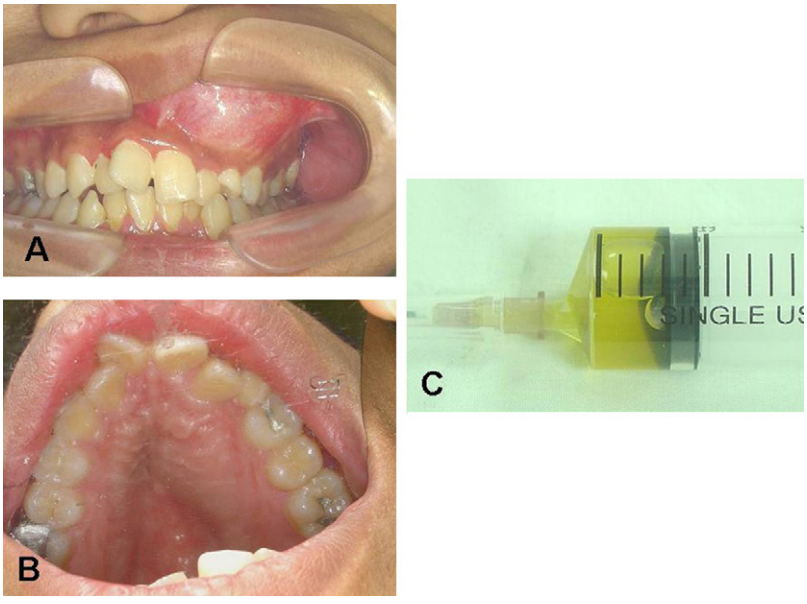

Fig. 1 Buccal (A) and palatal aspects (B) of the swelling. Aspiration of straw-colored fluid (C).