

CASE REPORT

Dental follicular hamartomas in the opercula of teeth delayed in eruption: A case report and review of the literature

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KEYWORDS

Dental follicular hamartoma; Odontogenic; Delayed tooth eruption; Peripheral odontogenic fibroma **Summary** Odontogenic lesions of hamartomatous origin may occasionally result in delayed tooth eruption. However, the lack of a universally accepted terminology for such lesions may hinder their recognition. We present a case showing delay in the eruption of permanent mandibular second molars bilaterally; histopathologic examination of the excised opercula revealed features reminiscent of peripheral odontogenic fibroma with an abundance of stellate giant cells. After review and analysis of the pertinent literature, the lesions were diagnosed as dental follicular hamartomas to reflect their odontogenic (dental follicular) origin and hamartomatous nature.

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Introduction

Delay or failure of tooth eruption has been attributed to a number of systemic conditions or local factors.¹⁻⁵ Various pathologic entities of odontogenic origin may be responsible for delays or failure in eruption of teeth, including cystic lesions (e.g. dentigerous cyst), neoplasms (e.g. ameloblastic fibroma) and hamartomatous lesions (e.g. odontoma).¹⁻⁵ In addition to odontomas, other odontogenic hamartomatous lesions, designated with various different names in

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the past, have been implicated with disturbances in eruption.^{1,5–7} These hamartomatous lesions need to be discriminated from histologically-similar odontogenic tumors, in particular peripheral odontogenic fibroma (POdF), because of different pathogenesis and recurrence potential.^{8,9}

We review one case involving bilateral partially erupted second molars, the excised opercula of which were examined histopathologically, showing features consistent with odontogenic hamartomas. A discussion of the origin and histogenesis of these lesions, including a review of the epidemiologic, clinical and histopathologic features of previously reported odontogenic hamartomatous lesions that hamper tooth eruption, is provided.

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Case report

A healthy, 14-year-old Caucasian male presented for evaluation of tenderness in the area of the mandibular left and right second molars. His medical history was not contributory. On examination, the soft tissues overlying the distal portions of the partially erupted teeth #18 and 31 appeared enlarged. The opercula covering both teeth were excised and submitted for histopathologic evaluation. On gross examination, both specimens appeared as tan brown tissues measuring $0.6 \times 0.6 \times 0.5$ cm and $0.6 \times 0.4 \times 0.3$ cm, respectively. Histologically, the two lesions appeared identical; they were non-encapsulated and exhibited numerous islands of odontogenic epithelium in a dense, mildly inflamed fibrous connective tissue stroma, containing thick bundles of collagen fibers and few myxoid areas (Fig. 1A). Squamous metaplasia of the odontogenic epithelial islands was discerned (Fig. 1B and C). Round, cementum-like calcifications

were focally present (Fig. 1B and C). The stromal cells ran-

ged from spindle shaped to stellate-type giant cells with one or two nuclei to multinucleated giant cells (Fig. 1D). The overlying epithelium exhibited acanthosis and parakeratosis. In light of these histologic findings, inflammatory (e.g. pericoronitis) and reactive or hyperplastic (e.g. fibrous hyperplasia) lesions, which are unlikely causes of delayed tooth eruption, were excluded. In contrast, the observed lesions appeared histologically similar to POdF. Nonetheless, the localization within the opercula of partially erupted permanent molars and the presence of numerous stellate-type and multinucleated giant cells strongly supported a diagnosis of odontogenic hamartomatous lesions in a pericoronal location. A diagnosis of dental follicular hamartomas within the opercula was made. After excision of the opercula, both second mandibular molars erupted uneventfully (Fig. 2).

Discussion

In contrast to odontogenic cysts and neoplasms,^{1-5,10,11} odontogenic hamartomas that are present in the opercula of teeth and interfere with tooth eruption have received little attention in the literature.^{1,5,7} Previous studies have attempted to describe these lesions using terms such as



Figure 1 (A) Low power photomicrograph showing islands of odontogenic epithelium (double arrowhead) and focal round calcifications (arrowhead) in a dense connective tissue stroma (hematoxylin and eosin, original magnification $100\times$). (B) and (C) High power photomicrographs showing round cementum-like calcifications (arrowheads) and islands of odontogenic epithelium with squamous metaplasia (double arrowheads) in a dense fibrous connective tissue background (hematoxylin and eosin, original magnification $200\times$). (D) High power view demonstrating numerous stellate-type giant cells with one or two nuclei and occasional multinucleated giant cells (arrowheads) interspersed between thick bundles of collagen fibers (hematoxylin and eosin, original magnification $200\times$).

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