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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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.

KEYWORDS
Dental follicular hamartoma; Odontogenic; Delayed tooth eruption; Peripheral odontogenic fibroma

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 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.
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 x 0.6 x 0.5 cm and 0.6 x 0.4 x 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 ranged 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](image-url)
"odontogenic giant cell fibromatosis (OGCF)" and "pericoronal hamartomatous lesions". Nonetheless, these terms have not been widely accepted resulting in under-recognition and potential misdiagnosis. Moreover, the distinction of these hamartomatous lesions from histopathologically-similar odontogenic neoplasms, most notably POdF, is not always clearly defined.8

Philipsen et al.5 used the term OGCF to describe an odontogenic hamartomatous lesion present within the opercula of permanent first and second molars delayed in eruption; this finding was observed in 29.7% of examined specimens.5 Histologically, OGCF presented as a non-encapsulated lesion harboring fibrous connective tissue with thick bundles of collagen fibers, containing a large number of spindle shaped or stellate cells and large multinucleated cells, as well as proliferating strands and islands of odontogenic epithelium with occasional squamous cell metaplasia; no calcifications were seen. In a similar study of Yonemochi et al.,1 50.8% of the examined opercula of teeth delayed in eruption were diagnosed as pericoronal myxofibrous hyperplasia (PMH), while 13.1% of cases were classified as infantile ameloblastic fibromatosis (IAF). Histopathologically, PMHs exhibited hyperplasia of odontogenic mesenchymal tissues with a myxoid appearance in which odontogenic epithelial islands and mesenchymal multinucleated giant cells were present in 58.1% and 51.6% of cases, respectively; the presence of calcified material ranging from enamaloid through dentinoid to bone was demonstrated in 6.5–9.7% of cases.1 IAF was usually located adjacent to PMH and showed an ameloblastic fibroma-like histology with atrophic ameloblastic components and poor encapsulation. These authors proposed that PMH and IAF be categorized into a new disease entity termed "Pericoronal Hamartomas of Odontogenic Origin", which may also include odontomas.1

Both studies by Philipsen et al.5 and Yonemochi et al.1 concluded that the presence of hamartomatous lesions in the pericoronal areas of teeth delayed in eruption interferes in the pathway of an erupting tooth and may cause sufficient tissue derangement and remodeling to impede tooth eruption. The lesions described in the present case were also located in the opercula of teeth delayed in eruption, showing histopathologic characteristics virtually identical to those of OGCF with the additional finding of cementum-like calcifications;3 they also fell into the spectrum of histologic features of PMH.1 Indeed, Yonemochi et al.1 suggested that the nosologic spectrum of PMH includes OGCF as well as additional odontogenic hamartomatous lesions that are devoid of giant cells but exhibit a myxofibrous connective tissue with or without calcifications and odontogenic epithelial islands.

Because of the odontogenic fibroma-like features of pericoronal odontogenic hamartomatous lesions, distinction from POdF may be difficult. Interestingly, the term odontogenic epithelial hamartoma was used in the past to describe lesions that were clinically and histopathologically identical to POdF and considered to represent a transitional stage between a developmental anomaly and a true odontogenic tumor.14,15 Although a hamartomatous or reactive origin of POdF has been debated, POdF is widely accepted as a benign odontogenic neoplasm that represents the soft-tissue counterpart of the central odontogenic fibroma (CODF).8,9,12,13

In this regard, recent studies have documented a significant recurrence rate of POdF, which further supports its neoplastic behavior.9 Because of the histologic similarities but disparate clinical behavior of neoplastic POdF and hamartomatous pericoronal lesions, clinicopathologic correlation is necessary for their distinction. Identification of stellate-type and multinucleated giant cells, which are not described in POdF,8,9,12,13 also favors a hamartomatous lesion.

Noteworthy is that odontogenic fibroma-like changes may also be seen in the dental follicles of impacted teeth.6,16–18 According to Gardner,19 lesions surrounding the crown of an impacted tooth and consisting of fibrous connective tissue with variable amounts of odontogenic epithelium and calcifications should be classified as hyperplastic dental follicles, despite their superficial resemblance to CODF. Interestingly, there are a few reported cases of enlarged dental follicles with odontogenic fibroma-like changes that were associated with multiple impacted teeth and considered to be hyperplastic or hamartomatous in origin.5,16–18 Although the exact relationship of these central lesions to peripheral odontogenic hamartomas is unclear, it is possible that there is a spectrum of histologically-similar pericoronal hamartomatous lesions of dental follicular origin which interfere with tooth eruption and may be distinguished into central or peripheral variants based on their anatomic location.

In conclusion, odontogenic hamartomatous lesions present within the operculum may represent a relative common cause of delayed eruption of permanent teeth. These lesions exhibit an indolent, non-neoplastic clinical behavior and should be discriminated from neoplastic, potentially recurrent POdF. Adoption of a term that describes the dental follicular location and hamartomatous nature of these lesions, such as "dental follicular hamartomas", may facilitate their recognition by clinicians and pathologists alike.

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References