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Case Report

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Peripheral Odontogenic Fibroma in a Young Child: A Literature Review and a Report of a Rare Case

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Abstract

Peripheral odontogenic tumors (POTs) are uncommon, and even rarer in the pediatric population. A peripheral odontogenic fibroma (PODF) is the most numerous POT in the general and pediatric populations. The purpose of this paper was to report an unusual case of PODF in a very young child who was born at 29 weeks' gestation. The literature on PODFs in the pediatric population is reviewed.

Keywords: Peripheral odontogenic fibroma; Pediatric; Peripheral odontogenic tumor

Introduction

Peripheral odontogenic tumors (POTs) include the extraosseous counterparts of ameloblastomas, calcifying epithelial odontogenic tumors (CEOTs), squamous odontogenic tumors, calcifying cystic odontogenic tumors (CCOTs), adenomatoid odontogenic tumors (AOTs), ameloblastic fibromas, odontomas, odontogenic fibromas, and odontogenic myxomas, some of which are exceedingly rare. A comprehensive study carried out in an oral pathology biopsy service demonstrated that POTs represented only 0.05% of all submitted biopsy specimens [1]. Although POTs are uncommon among children, peripheral variants of odontogenic fibromas (e.g., CCOTs, AOTs, ameloblastic fibromas and odontomas) have been documented in the pediatric population. Peripheral odontogenic fibromas (PODFs) were found to be the most common of all POTs in general [1] and the most common POT in the pediatric population [2]. We report a case of PODF in a 3.9-year-old child whose only remarkable feature in her past history was being born at 29 weeks' gestation. The literature on PODFs in the pediatric population is reviewed.

Case Report

A 3.9 -year-old girl was referred by the community dentist to the Department of Oral Medicine in a University health center. The mother had noted an "odd-looking tooth" located at the anterior mandible. The child's past history was remarkable for premature induced birth (29th week of gestation) due to maternal uterine hematoma. The newborn was intubated and ventilated immediately after birth and remained so for a few days. She was also diagnosed as having Klebsiella pneumoniae-related sepsis for which she received antibiotic treatment. Other than infantile hemangiomas on the skin of her lower back, the child otherwise appeared to be healthy and fully developed, both cognitively and physically.

Clinical examination revealed the patient's face to be symmetrical with a normal skin tone. There were neither palpable submandibular or cervical lymph nodes nor any temporomandibular joint abnormalities.

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An anterior open bite was explainable by habitual sucking of pacifiers. The midline of the lower dental arch was approximately 1 mm to the left compared to the midline of the upper dental arch (Figure 1). The child was caries free, and her oral hygiene was highly satisfactory. The right maxillary central and lateral incisors demonstrated enamel hypoplasia. The left mandibular deciduous lateral tooth was approximately 3 mm wider compared to its right-sided counterpart, suggesting single tooth macrodontia. This tooth was also over-erupted, with obvious distal displacement and grade I mobility (Figure 2). The attached buccal and lingual gingivae surrounding it were symmetrically hyperplastic (Figure 2), with a smooth surface lining of normal-colored oral mucosa and firm consistency on palpation. There were no bloody or purulent discharges upon probing, and her periodontal status was otherwise sound. Occlusal trauma on the tooth was ruled out since it did not have occlusal contacts either in the resting position or in working/nonworking relations. Periapical radiographs revealed that the lower left lateral incisor had an open apex, while its right-sided counterpart had a closed apex (Figure 3). In



Figure 1: Right maxillary incisors demonstrating enamel hypoplasia. The lower left lateral is obviously wide compared to its right-sided counterpart, over-erupted, and has obvious displacement.

addition, the pulp chamber was disproportionately wide relative to tooth dimensions. There was obvious distal tooth displacement (Figure 3).

The clinical differential diagnosis included localized reactive hyperplastic lesions (LRHLs), such as peripheral ossifying fibromas (POFs), non-odontogenic benign tumors, such as myofibromas, and POTs, such as PODFs.

After the child was sedated with midazolam (7 mg) and nitrous oxide, the lower left lateral incisor was extracted and the hyperplastic gingiva was excised by scalpel and submitted to microscopic examination. The hematoxylin and eosin-stained slides revealed islands of inert odontogenic epithelium in a fibrous stroma. The remaining connective tissue contained mild chronic inflammatory infiltrate (Figure 4). The microscopic findings were consistent with the diagnosis of a PODF.

There was no evidence of recurrence, either clinically (Figure 5) or radiologically (Figure 6) at the one-week, 3-month, 8-month and 1-year follow-up visits. The extraction area demonstrated normal healing. There were no signs of inflammation, and no symptoms were reported. The radiographic periapical image taken at eight months following the procedure demonstrated normal appearance of the bone and of the deciduous teeth and adjacent tooth germs near the extraction area. Since recurrences have been reported in the literature [2], the child is scheduled for follow-up appointments every four months.



Figure 2: The attached buccal and lingual gingivae surrounding the tooth appear hyperplastic and covered by normal-looking oral mucosa.



Figure 3: Periapical X-ray. The lower left lateral presenting an open apex, a wide pulp chamber and tooth displacement.

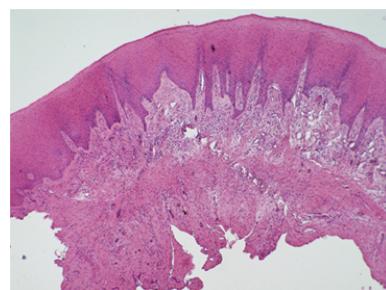


Figure 4: Excisional biopsy showing fibrous stroma lined by keratinizing stratified squamous epithelium with alternating areas of hypo- and hypercellularity. Small islands/rosettes of epithelium can be discerned, consistent with inert odontogenic epithelium. The rest of the connective tissue contained mild chronic inflammatory infiltrate. Peripheral odontogenic fibroma was diagnosed (Hematoxylin and eosin stain, original magnification x 40).



Figure 5: Follow-up at one year following the excisional biopsy revealing satisfying tissue recovery and no recurrence of the lesion (occlusal view).

Discussion

PODF is the only POT that is more frequent than its central counterpart. The mean age at diagnosis for the population at large is 32.3 years, and there is a slight female preponderance. The mandibular incisor/canine and premolar areas are the most prevalent sites, affecting both the buccal and lingual aspects of the gingival [1]. PODFs present as exophytic gingival masses that histopathologically show unencapsulated fibrous connective tissue with inert odontogenic epithelium [3]. Table 1 lists a summary of age, gender and location of POFs in the pediatric populations reported in the literature.

We defined “pediatric population” in accordance with Buchner et al. [1] as being 0- to 19-year-olds. We reviewed the clinical information of all the documented cases of PODFs and POTs among pediatric patients that appeared in the English-language literature. Medline’s PubMed interface was searched using the keywords “peripheral” combined with “odontogenic tumor”, “odontogenic fibroma”, “ameloblastoma”, “calcifying epithelial odontogenic tumor”, “squamous odontogenic tumor”, “calcifying cystic odontogenic tumor”, “adenomatoid odontogenic tumor”, “ameloblastic fibroma”, “odontoma”, “odontogenic fibroma”, and “odontogenic myxoma”.

One review of the English language literature specific to PODFs in children aged 0-19 years [2] revealed that 35 of cases of all ages ($n=151$, 23.2%) involved children, the youngest of which was five years old. Another case series described 6 of all 23 patients (26%) being between 12-18 years of age [1]. Daley and Wysocki’s survey [4] revealed that 6 of 36 new cases of all ages were within 0-19 years of age, the youngest being 12 years old. Reports on pediatric populations under the age of 9 years are uncommon, although a few cases have been documented, including two that involved infants [5,6] and one each of a 2-year-old [3], a 3-year-old

Case No	Study	Age (years)	Pediatric cases	Gender	Location
1	Buchner et al.[1]	2	1	M	Maxilla
2		18	1	F	Maxilla
3	Weber et al. [7]	8	1	F	Diffused
4		3	1	M	Diffused
5	Daley et al. [4]	19	1	M	Mandible
6		13	1	M	Mandible
7		18	1	F	Mandible
8		12	1	F	Maxilla
9		14	1	M	Maxilla
10		16	1	F	Unknown
11	Siar & Ng [6]*	05-Dec	1	Unknown	Unknown
12		4	1	Unknown	Unknown
13		9	1	Unknown	Unknown
14	Martelli-Júnior et al. [5]	11	1	Unknown	Unknown
15	Buchner et al. [1]	03-Dec	1	F	Maxilla
16		18	1	M	Mandible
17		16	1	F	Mandible
18		12	1	F	Maxilla
19		15	1	M	Mandible
20		12	1	F	Maxilla
21	Alaeddini et al. [8]	17	1	M	Mandible
22		18	1	M	Mandible
23		12	1	M	Mandible
24		8	1	M	Mandible
25	Ritwik& Brannon [2] **Current case	9	1	M	Mandible
26		Unknown	35	Unknown	Unknown
27		4	1	F	Mandible

Table 1: Age, gender and location of peripheral odontogenic fibromas in pediatric populations described in the literature.

*Only age ranges are described, indicating that pediatric cases were included, without further specifications.

**One patient is 5 years old.

[7], a 4-year-old [6], and a 5-year-old [2], and two others, each describing an 8-year-old [7,8]. We believe our case to be the fifth to document a PODF in a child younger than four years of age.

Overall, POTs comprise only 0.05% of all submitted oral biopsy specimens, with a relative frequency of 51% among the various histopathologic types of PODFs, 29% of peripheral ameloblastomas and 13% of peripheral calcifying cystic odontogenic tumors. Other histopathological types of POT are sporadic [1]. POTs are uncommon among children patients. One pediatric case of PCCOT was documented in a series of 6 cases (16.6%), [1] and another was reported in a single case report [9]. The age range of peripheral adenomatoid odontogenic tumors in 15 out of 15 pediatric cases was 3 to 19 years (mean, 11.9 years) [10-13]. The age range of peripheral ameloblastic fibromas in 7 out of 7 pediatric cases was 1 to 8 years (mean, 4 years) [1,10,14-16]. The age range of peripheral developing odontoma in 8 out of 8 pediatric cases was 1 to 14 years (mean, 6.6 years) [10]. There is no documentation of the peripheral counterparts of ameloblastomas in pediatric cases [1], keratocystic odontogenic tumors [10], squamous odontogenic tumors [17] or odontogenic myxomas [18].



Figure 6: Periapical X-ray at one year following the excisional biopsy demonstrating normal appearance of bone in the extraction area of the lower left lateral.

Like other tissues of the body, the oral structures can be affected by prematurity birth. The effects of prematurity in the mouth are associated with enamel hypoplasia, crown dilacerations, palatal distortions and distortions of dental arches [19-30]. There is no published correlation between preterm birth and neoplastic lesions, with the exception of infantile hemangiomas [31].

This case report and literature review highlights the rarity of PODFs among young children. Any associations between odontogenic lesions and premature birth are still unclear.

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