Peripheral Tumor with Osteodentin and Cementum-like Material in an Infant: Odontogenic Hamartoma or Odontoma?

Aikaterini Sfakianou, DDS, MSc¹
Dimitris E. Emmanouil, DDS, MSc, PhD²
Konstantinos I. Tosios, DDS, PhD³
Alexandra Sklavounou, DDS, MSc, PhD⁴

ABSTRACT

The purpose of this report is to describe a peripheral tumor on the mandibular alveolar ridge of a seven-month-old Caucasian boy, consisting of ectomesenchymal odontogenic tissues, in particular osteodentin and cementum-like material, in a cellular or loose vascular connective tissue stroma. This case may be considered either a peripheral odontogenic hamartoma or a peripheral odontoma. (J Dent Child 2016;83(1):XX)

Keywords: infant, gingival mass, hamartoma, osteodentin, oral pathology

Peripheral (extraosseous) tumors with dental hard tissues developing on the soft tissues of the tooth-bearing areas of the jaws are rare; the most well-known examples are peripheral odontoma¹ and peripheral odontogenic fibroma.²

The purposes of this report are to describe a peripheral tumor on the mandibular alveolar ridge of a seven-month-old Caucasian boy, consisting of ectomesenchymal odontogenic tissues, in particular osteodentin and cementum-like material, in a cellular or loose vascular connective tissue stroma, and to discuss its classification.

CASE REPORT

A seven-month-old Caucasian boy was referred by his pediatrician to the Department of Pediatric Dentistry, School of Dentistry, University of Athens, Athens, Greece, for diagnosis and management of a nodule on the gingiva, first noticed by his parents a few days before presentation. No discomfort during breast-feeding was noticed by the mother. The baby was born full term to non-consanguine parents, and his medical history was non-contributory.

Clinical examination revealed a pedunculated mass on the anterior mandibular alveolar ridge, lateral to the midline, measuring approximately 1.1 cm x 0.9 cm x 0.9 cm (Figure 1). It was covered by normal-appearing mucosa and felt firm on palpation. The rest of the oral
mucosa was within normal limits. The parents denied the presence of natal or neonatal teeth, and no primary tooth had erupted.

With a provisional clinical diagnosis of congenital epulis of the newborn, 0.5 ml of 4% articaine hydrochloride with 1:100,000 adrenaline hydrochloride were infiltrated at the base of the lesion for an excisional biopsy. The lesion was easily removed, and the underlying bone was found to be intact. The post-operative course was uneventful, and no recurrence has been recorded during a five-year follow-up. The central primary mandibular incisors erupted normally a few weeks following excision of the lesion (Figure 2).

The specimen was fixed in 10 percent neutral-buffered formaldehyde and submitted for microscopic examination at the Department of Oral Pathology and Medicine, School of Dentistry, University of Athens. Grossly, it was resistant to sectioning, and a dental radiograph showed the presence of a central, circumscribed mass of calcified tissue (Figure 3). Following decalcification in an ethylenediaminetetraacetic acid- (EDTA) based solution, a five-μm-thick tissue section was cut. The tumor was composed of connective tissue, centrally occupied by a mass of woven bone (Figure 4a) with osteoid rimming and osteocytes (Figure 4b) but no osteoblasts or osteoclasts.

Focally, discrete dentin tubule-like structures were seen crossing the bone; adjacent to those areas, a flattened rim of cells consistent with odontoblastic layer was identified (Figure 4c). The connective tissue consisted of long cellular strands (Figure 4b), focally showing basophilic globules consistent with cementum-like material.
odontogenic hamartomas that presented bilaterally on the posterior maxillary ridges and unilaterally on the anterior mandible. Meanwhile, dentinoid in a dental papilla-like stroma was present in the case of a newborn boy with odontogenic hamartoma on the anterior mandibular gingiva of a four-month-old Caucasian boy, a month after exfoliation of natal incisors. 

As the presence of osteodentin and cementum-like material (i.e., ectomesenchymal odontogenic hard tissues) on a fibrous or loose vascular connective tissue stroma. 

DISCUSSION
The tumor presented herein was peripheral (extraosseous) because during surgical excision it was not connected to the underlying bone, which was intact, and was not associated with a natal tooth, primary tooth or tooth germ. A post-excisional radiograph showing complete circumscriptum of the hard tissue is also supportive of the presumption that the hard tissue was not connected to the mandibular cortical bone. Its unique microscopic feature was the presence of osteodentin and cementum-like material (i.e., ectomesenchymal odontogenic hard tissues) on a fibrous or loose vascular connective tissue stroma.

There are rare reports in infants of peripheral tumors containing osteodentin, dentin, or dentinoid material. Osteodentin surrounded by dental papilla-like myxoid tissue was described in a case of myxoid calcified hamartoma, on the anterior mandibular gingiva of a four-month-old Caucasian boy, a month after exfoliation of natal incisors. The authors did not characterize this tumor as odontogenic, due to the absence of odontogenic epithelium. Dentin associated with tooth germs was found in the case of a newborn girl with bilateral hamartomas on the posterior mandibular ridge. Dentinoid and cementoid associated with natal teeth were reported in a case of gingival fibrous hamartomas located on the anterior mandibular ridge of a 10-month-old boy. Meanwhile, dentinoid in a dental papilla-like stroma was present in the case of a newborn boy with odontogenic hamartomas that presented bilaterally on the posterior maxillary ridges and unilaterally on the anterior mandible.

In the last two cases, odontogenic epithelium was identified, in contrast to our case and some other cases of odontogenic hamartomas. For dentin to be formed, inductive interactions between odontogenic epithelium and ectomesenchyme are expected; therefore, odontogenic epithelium should be present either elsewhere in the lesion or earlier in its formation. The combination of dental hard tissues and odontogenic tissues of epithelial differentiation is seen in odontomas, and there are case reports of peripheral developing odontomas in newborns. Therefore, cases of hamartomas with dental hard tissues, regardless of the presence or absence of odontogenic epithelium, may represent a stage in the development of an odontoma that is currently considered to be a hamartoma.

Osteodentin-like material was also seen in two cases of eruption mesenchymal calcified hamartomas that were associated with erupting teeth. In the present case, a connection of the lesion with an erupting tooth was not evident macroscopically.

Differential diagnosis in the present case included peripheral odontogenic fibroma (POdF) and peripheral ossifying fibroma (POF). POdF may contain bone, cementoid, or dentinoid in a fibrous, fibromyxoid, or myxoid connective tissue background, but the presence of odontogenic epithelium is required for its diagnosis. There is a single case report of a POdF (World Health Organization type) in a four-month-old girl in which no hard tissues were present. POF is a common reactive lesion of the gingiva consisting of cellular fibroblastic tissue and woven bone, trabecular bone, cementum-like material, dystrophic calcifications, or a combination of those components.

Four cases of POF were reported in one-week- to seven-month-old infants, two of which were on the anterior mandibular ridge and one on the molar maxillary alveolar ridge. One case was associated with a primary tooth, and another developed one week after extraction of a neonatal tooth. The hard tissue is typically described as osteoid, but it should be noted that, in the present case, dentin tubule-like structures were evident in a limited area of the hard tissues; therefore, its presence in previous reported cases of POF in infants cannot be fully excluded.

In conclusion, we suggest that peripheral tumors with dental hard tissues in the form of dentin, dentinoid, or osteodentin are hamartomas of odontogenic origin and can be better classified as peripheral odontomas.

REFERENCES


