

Case Report

Congenital peripheral developing odontoma accompanied by congenital teratomatous fibroma in a 9-month-old boy: a case report

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Abstract: Peripheral odontoma is rare, and only two cases of congenital peripheral odontoma have been reported. Congenital oral fibroma is also rare. We describe a unique case of congenital peripheral developing odontoma accompanied by congenital teratomatous fibroma in an infant. Both tumors were difficult to detect on radiography. Two small masses were seen in the median anterior portion of the palatal mucosa of a 9-month-old boy. The masses had been present since birth and were surgically removed at age 28 months, when one of the masses had grown to a diameter of 8 mm. Histopathologic examination showed a fibrous lesion and a tooth germ-like rounded lesion composed of dental papilla, enamel organ, dentin, and cementum. Although congenital odontoma is rare, it should be considered when selecting appropriate treatment, as early radiographic detection is difficult. (*J Oral Sci* 55, 89-91, 2013)

Keywords: congenital odontoma; peripheral odontoma; congenital fibroma.

Introduction

Odontomas are the most common intraosseous odontogenic tumor of the maxilla and mandible and are regarded as hamartomas of both ectodermal and meso-

dermal origin. They are often associated with permanent teeth and rarely associated with deciduous teeth (1). In contrast to intraosseous odontoma, peripheral (extraosseous) odontoma is rare, and only two cases of congenital peripheral odontoma have been reported (2). We report here a unique case of congenital peripheral developing odontoma accompanied by teratomatous fibroma in an infant.

Case Report

A 9-month-old Japanese boy was referred to our university hospital with a chief complaint of swelling in the median anterior portion of the palatal mucosa. Two localized soft masses (both 3 mm in diameter) were seen at the lingual portion of the palate, behind the deciduous incisors and incisive papilla. The surface of the mucosa overlying the lesion was normal in color on inspection. The asymptomatic swelling had been present since birth. Radiography showed no apparent abnormalities in the lesion (Fig. 1). At age 12 months, the size of the masses was unchanged, and breast-feeding and food intake were normal. At 16 and 20 months of age, growth was noted in one of the masses, and swallowing with a straw had become difficult. At age 26 months, the diameters of the masses were 7 mm and 3 mm (Fig. 2). At age 28 months, one of the masses had grown to a diameter of 8 mm, and, as a result, both masses were surgically removed under the periosteum, with the patient under general anesthesia. Histopathologic examination after decalcification of the larger mass revealed a tooth germ-like rounded lesion in the mucosa and a fibrous lesion covered with intact oral epithelium. The larger mass was composed

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Fig. 1 Intraoral radiograph of patient at age 9 months.



Fig. 2 Photograph of clinical presentation at age 26 months shows two masses covered with intact mucosa on the palate and no malalignment of teeth.

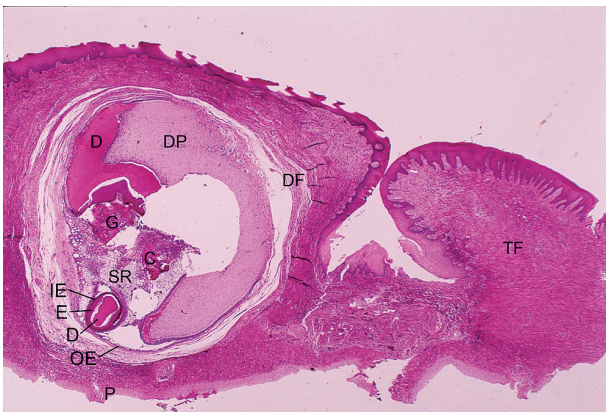


Fig. 3 (a)

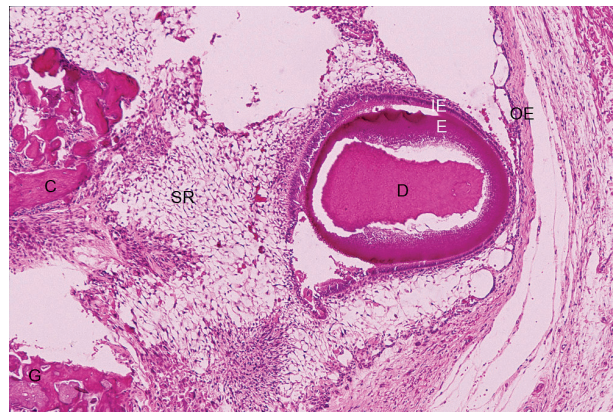


Fig. 3 (b)

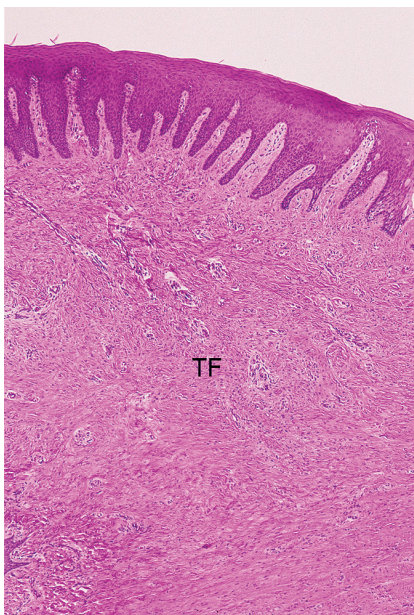


Fig. 3 (c)

Fig. 3 Histopathologic findings of the two masses (hematoxylin and eosin staining).

(a) Tooth germ-like rounded lesion in the mucosa and teratomatous fibrous lesion (original magnification, $\times 20$). (b) The larger mass is an odontoma composed of dental papilla, enamel organ, dentin, and cementum (original magnification, $\times 100$). (c) The smaller mass is a fibroma composed of dense fibrous connective tissue. Inflammatory changes were not seen (original magnification, $\times 100$).

D: dentin, DP: dental papilla, G: ghost cell, C: cementum, SR: stellate reticulum, DF: dental follicle, P: periosteum, TF: teratomatous fibroma, IE: inner enamel epithelium, E: enamel matrix, OE: outer enamel epithelium.

of dental papilla, enamel organ, dentin, and cementum (Figs. 3a-b). The smaller mass comprised dense fibrous connective tissue (Fig. 3c). The histopathologic diagnosis was peripheral developing odontoma accompanied by teratomatous fibroma. The patient's postoperative course was normal, and there have been no signs of local recurrence during the 10-year follow-up period.

Discussion

Definitive diagnosis of odontogenic lesions in small children can be challenging. Although congenital odontoma and peripheral odontoma are rare, they must be considered when selecting appropriate treatment. Ide et al. (3) reported that the mean age of patients with peripheral developing odontoma was 6.6 years (range, 1-14 years), which is 10 years younger than the mean age of patients with intraosseous odontoma. Depending on the degree of tissue differentiation, odontoma is classified as compound odontoma or complex odontoma; compound odontoma shows greater morphologic differentiation. With regard to etiology, odontoma is often associated with permanent teeth (1), supernumerary teeth or missing teeth (1), and history of trauma (4). Erupted odontoma (5) is also a variation. It is located on the coronal side of an erupting or impacted tooth and erupts into the oral cavity before the tooth. Thus, the pathogenesis of odontoma may vary. In the present case, the relation between the congenital peripheral odontoma and teratomatous fibroma was not clear. The extremely low frequency of congenital

odontoma is consistent with the fact that most cases of odontoma are associated with permanent teeth (1).

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