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CASE REPORT

Myxoid calcified hamartoma and natal teeth: A case report

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Summary We report the case of a 4-month-old Caucasian male baby who presented an uncommon mass in the anterior mandibular ridge. The patient was born with two natal mandibular incisors, which exfoliated some weeks after birth, followed by a growth of nodular lesion in the same region. Based on the clinical and histopathological features, the diagnosis was of myxoid calcified hamartoma. Immunohistochemical and scanning electron microscopic analysis of the lesion were performed. Hamartomas in the mandibular ridges associated with natal teeth are rare, but they must be considered in the differential diagnosis of common lesions, such as congenital granular cell epulis.

1. Introduction

Hamartoma is an abnormal growth of tissues in their normal site, which arises during the development of the affected organ or structure. Typically, the tissues in hamartomas appear disorganized and ill-defined, merging with the normal surrounding structures. Treatment of hamartoma is complete surgical excision, with excellent prognosis and rare recurrence [1,2]. Natal teeth usually occur in pairs, most commonly corresponding to the lower primary central incisors. Their incidence is approximately 1 to 2.000/3.000 live births, representing early eruption of normal primary deciduous dentition, and less than 10% of them are supernumerary. Morphologically, natal teeth are conical and yellowish, with poor or absent root formation, nevertheless, few cases might resemble normal primary dentition. Complications include discomfort during suckling and laceration of the mother's breasts, besides sublingual ulceration or risk of teeth's aspiration. Surgical extraction usually is indicated [3,4].

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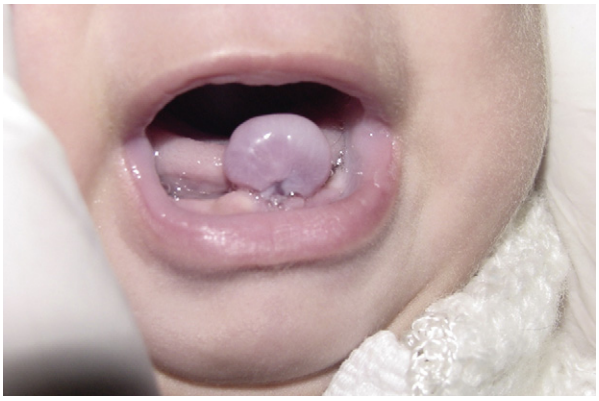

Fig. 1 Clinical view of a nodular pedunculated lesion in a 4-month-old baby, diagnosed as myxoid calcified hamartoma.

Association of natal teeth with a hamartomatous lesion in the alveolar ridge in babies is extremely rare. We report a case of an uncommon hamartoma affecting a 4-month-old Caucasian male patient, who displayed previously two natal teeth on the same location. To the best of our knowledge, this is the first case of a unique lesion with unusual clinical and histopathological features reported in the English-language literature.

2. Case report

A 4-month-old Caucasian male patient was referred to a private dental office, in Lages, Santa Catarina, Brazil, because of a mass on the anterior mandibular ridge, which was present for 1 month (Fig. 1). Parents reported that the child was born with two teeth on this location, which exfoliated after some weeks due to mobility, followed by a progressive tissue growth in the same place. Intraoral examination showed a firm, pedunculated, and nodular lesion, covered by normal mucosa, measuring 16 mm $\times$ 12 mm $\times$ 9 mm, located in the anterior mandibular ridge. It caused discomfort during breast-feeding. The clinical diagnosis was of a reactive or developmental lesion, and under local anaesthesia, an excisional biopsy was performed. In the histopathological examination the lesion showed an acantotic keratinized stratified squamous epithelium, with discrete rete ridges covering a dental papilla-like myxoid tissue, numerous blood vessels (Fig. 2A) and a single calcified mass located in the center compatible with osteodentin-like tissue (Fig. 2B). Considering the clinical and histopathological findings, a diagnosis of hamartoma was established, including the descriptive terms myxoid and calcified (myxoid calcified hamartoma associated with natal teeth). After 1 year of follow-up, no recurrence was observed.

Immunohistochemical (IHC) and scanning electron microscopic studies were carried out to analyze the myxoid and calcified components of this lesion. Vimentin (Vim 3B4, dilution 1:400; Dako A/S, Glostrup, Denmark), pan-cytokeratin (AE1/AE3, dilution 1:500; Dako A/S), CK19 (RCK 108D, dilution 1:400, Dako A/S), alpha-smooth muscle actin (1A4, dilution 1:400; Dako A/S), CD34 (QBEnd 10, dilution 1:100, Dako A/S), D2-40 (dilution 1:100, Dako A/S) and S-100 (polyclonal, dilution 1:12,000; Dako A/S) were used. The surface epithelium was positive for AE1/AE3, while the mesenchymal myxoid tissue was positive for vimentin (Figs. 3A and B). Positivity for CK19 was observed only in the basal and suprabasal layers of the surface epithelium. Isolated and small nervous fibers were S-100 immunoreactive (Fig. 3C). The smooth muscle cells of the blood vessels were positive for alpha-smooth muscle actin (Fig. 3D). The vascular endothelium was highlighted with

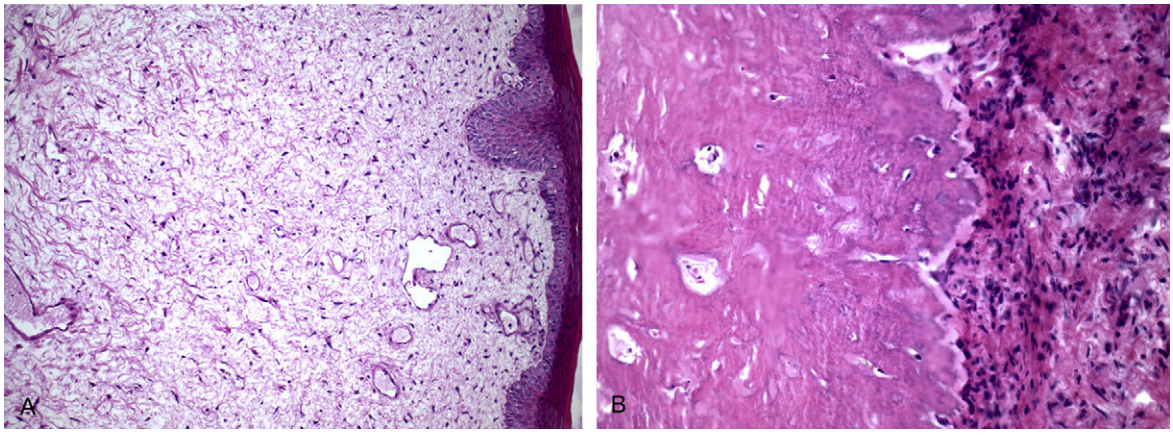

Fig. 2 (A) Myxoid tissue with scattered spindle cells resembling dental papilla (H&E, OM 20$\times$). (B) Calcified tissue displaying focal cellular areas, separated of the adjacent connective tissue by eosinophilic transitional cell rich zone (H&E, OM 20$\times$).