

Gingival fibrous hamartoma associated with natal teeth

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A variety of gingival enlargements can occur in children, but they are rarely observed in the infant. The aim of this study is to present a case of a ten-month old male patient, with an anterior mandibular hamartoma associated to natal teeth, and to describe the clinical and histological characteristics of this anomaly and the treatment performed.

J Clin Pediatr Dent 29(3): 249-252, 2005

INTRODUCTION

The term hamartoma is used to describe a “tumor-like”, but primarily non-neoplastic, malformations or inborn errors of tissue development. Essentially, it is composed of an abnormal mixture of tissues native to the part with an excess of one or more of these tissues, and develops during the time when dental structures remain capable of additional development or maturation.¹ The treatment of hamartoma is

complete surgical excision. Recurrence is rarely observed and prognosis, even without a complete excision is often favorable.^{2,3}

The natal teeth are present at birth, while neonatal teeth erupt within the first month of life. Natal teeth are more frequent than neonatal teeth in a proportion of 3:1. The natal teeth incidence is 1 in 2000 to 2500 births, with females more affected in the mandibular anterior incisors.⁴ Treatment of natal and neonatal teeth should be studied in each case. When these teeth show excessive mobility, immediate extraction is the most common protocol. To eliminate the potential discomfort during breast-feeding and to prevent the development of ulceration in the floor of the mouth known as Riga-Fede disease, the incisal edges may be filed to decrease their sharpness.⁵

The purpose of this work is to report a case of a ten-month old patient, with a pedunculated mass on the anterior mandibular ridge associated to natal teeth.

CASE REPORT

A 10-month old male patient was brought to the Pediatric Dentistry Department at the University of São Paulo because of a “tissue” mass on the anterior mandibular ridge, which was present since birth. Parents reported that the child was born with two teeth nearby the lesion and both of them exfoliated in the first week of life due to great mobility. On the examination a firm, nodular, pedunculated and elliptical lesion, measuring 10 x 5 x 5 mm, was present on the anterior mandibular ridge with the same color of the mucosa and with a “mineralized tissue” in the center of the lesion (Figure 1).

An occlusal radiograph exhibited a soft tissue enlargement with focal opacity presenting the same density of dentin and cementum (Figure 2).

The initial diagnosis was congenital epulis of the newborn. After medical evaluation, and signed and

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Figure 1. Presence of pedunculated nodule with smooth surface and similar color to the mucosa in the anterior mandibular ridge.

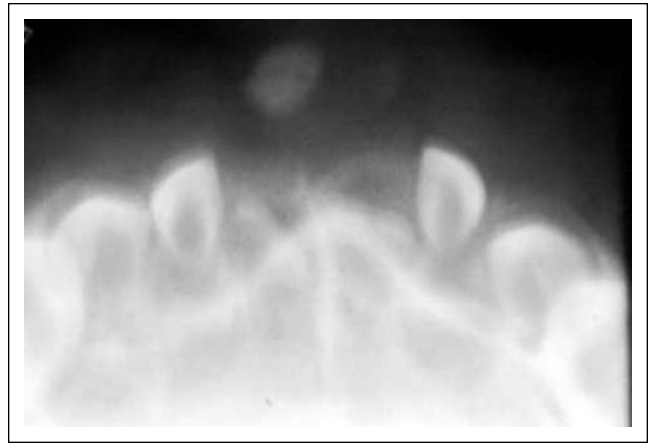


Figure 2. An occlusal radiograph that exhibits soft tissue enlargement with focal opacity that is the same density as dentin and cementum.



Figure 3. Surgical excision after local anesthetic technique.

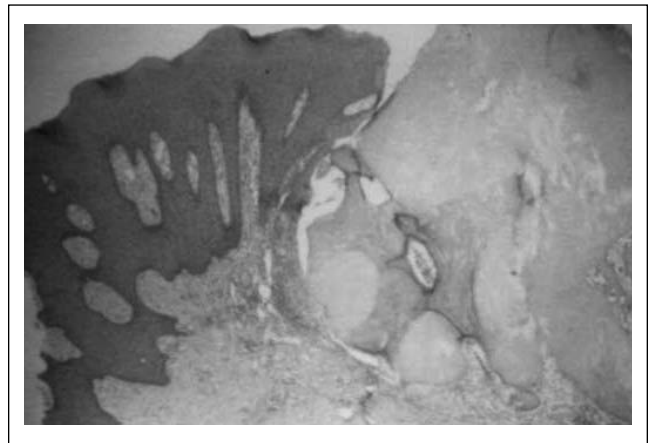


Figure 4. Panoramic view from tumor mass and anomalous dental tissue (HE, 40X).

informed consent from the parents, the lesion was excised under local anesthesia. The excision included the part of the lesion that was inserted at the alveolar bone (Figure 3). Finally, suture was done.

The specimen was submitted to histological analysis at the Oral Pathology Department of the University of

São Paulo. Decalcification with formic acid (20%) was performed first, and then tissue was sectioned and stained with hematoxylin-eosin. The specimen consisted of fibrous connective tissue with evenly distributed spindle cells, scattered cords and islands of odontogenic epithelium and chronic inflammatory cells. An

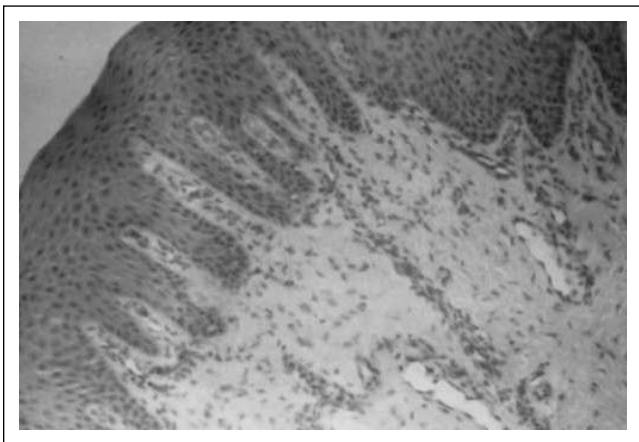


Figure 5. Photomicrography from hamartoma: acantosis in the epithelium and connective tissue with fibroblasts and collagen deposition (HE, 100X).



Figure 6. Aspect of surgical area after 6 months.

irregular aggregate of dentinoid and cementoid tissue was observed with entrapped pulpal tissue. The overlying keratinized stratified squamous epithelium exhibited acanthosis (Figures 4, 5). The histological analysis of the "mineralized tissue" observed in the center of the lesion is consistent with a poorly formed natal tooth.

According to the histopathological, clinical, and radiographic findings, the final diagnosis was gingival fibrous hamartoma and natal tooth. Six months after surgery, no abnormalities or recurrence were observed (Figure 6).

DISCUSSION

Developmental anomalies commonly diagnosed in newborns include Bohn's nodules, dental lamina cysts, Epstein pearls, natal teeth, congenital epulis and hamartomas. A hamartoma is classified as an odontogenic tumor; it is, however, only a developmental anomaly.¹ Hamartomas can present clinically as a congenital epulis especially due to the size and appearance, both lesions are usually excised, since they can affect breathing and nursing and most importantly to confirm a diagnosis by histological analysis.²

The congenital epulis of the newborn is predominantly seen in girls on the maxillary alveolar mucosa.^{2,3,6} However; hamartomas still do not have these epidemiological data well established. Treatment for both pathologies is surgical excision and recurrence is difficult to be observed.^{2,3} In some cases, it is possible to follow a spontaneous regression of the congenital epulis, but only when breathing and nursing are not affecting the infant.⁶

Sigal *et al.*⁷ reported a case of a newborn girl with two anterior natal teeth and two posterior bilateral mandibular hamartomas. In this case the lesions were considered hereditary. Differential diagnosis to hamartomas includes congenital epulis of the newborn and tumors such as teratomas, neuroectoderm tumor of infancy, and myoblastoma.² The congenital epulis of the newborn is clinically similar to hamartoma, usually appearing in the gingival mucosa. It is most commonly found in the maxillary alveolar ridge and ranging in size from millimeters to centimeters.^{3,8,9}

According to the studies of Kates *et al.*,¹⁰ the incidence of natal teeth was 1 in 716 births. All natal teeth found were mandibular central incisors in 61% of the cases. They came in pairs. In 95% of the cases, teeth were from the primary dentition. Our case presented only one tooth in the tumor mass and two others had exfoliated previously. Due to the absence of the primary central incisors on radiographic examination, we believe that the teeth that exfoliated were these, and the tooth in the lesion was a supernumerary.

Clinically, both natal and neonatal teeth present a conic shape and immature structure. Features like color variation from brown to yellow, hypoplastic enamel,

insufficient root development and excessive mobility predisposing to spontaneous loss or exfoliation may be present.¹¹

Hayes related a similar case from a newborn black girl with natal teeth and abnormal growth of tissues in 2000.¹²

Sigal *et al.*¹⁷ demonstrated a case of a newborn girl with two mandibular incisor natal teeth and bilateral mandibular odontogenic hamartomas is presented. The hamartomas were evident as pedunculated masses on the posterior mandibular gum pads. The natal teeth were extracted at birth, and the hamartomas were excised at 5 months of age. Microscopic investigation of the hamartoma demonstrated the presence of all odontogenic tissues with the exception of an enamel organ. In addition, there was a strong family history of natal teeth, which may suggest a hereditary basis for the development of the odontogenic hamartoma.

Even though hamartomas are benign lesions,¹²⁻¹⁷ they may bring great concern to parents since they affect the newborn child. Therefore, the correct diagnosis is important as is informing the parents about the favorable prognosis and adequate treatment of these lesions.

REFERENCES

- Gardner DG. The concept of hamartomas: its relevance to the pathogenesis of odontogenic lesions. *Oral Surg Oral Med Oral Pathol* 45: 884-6, 1978.
- Hayes PA. Hamartomas, eruption cyst, natal tooth and Epstein pearls in a newborn. *J Dent Child* 67: 365-8, 2000.
- Volpe F, Verrioli M. Congenital granular-cell epulis. A histochemical case study. *Minerva Stomatol* 46: 267-71, 1997.
- Zhu J, King D. Natal and neonatal teeth. *J Dent Child* 62: 123-8, 1995.
- Buchanan S, Jenkins CR. Riga-Fedes syndrome: natal or neonatal teeth associated with tongue ulceration. Case report. *Aust Dent J* 42: 225-7, 1997.
- Marakoglu I, Gursoy UK, Marakoglu K. Congenital epulis: report of case. *J Dent Child* 69: 191-2, 2002.
- Sigal MJ, Mock D, Weinberg S. Bilateral mandibular hamartomas and familial natal teeth. *Oral Surg Med Oral Pathol* 65: 731-5, 1988.
- Henefer EP, Abaza NA, Anderson SP. Congenital granular cell epulis-report of a case. *Oral Surg Med Oral Pathol* 47: 515-8, 1979.
- Takahashi H, Fuyita S, Satoh H. et al. Immunohistochemical study of congenital gingival granular cell tumor (congenital epulis). *J Oral Pathol Med* 19: 492-6, 1990.
- Kates GA, Needleman HL, Holmes LB. Natal and neonatal teeth: a clinical study. *J A D A* 109: 441-3, 1984.
- Rusmah M. Natal and neonatal teeth: a clinical and histological study. *J Clin Pediatr Dent* 15: 251-3, 1991.
- Onishi T, Sakashita S, Ogawa T, Ooshima T. Histopathological characteristics of eruption mesenchymal calcified hamartoma: two case reports. *J Oral Pathol Med* 32: 246-9, 2003.
- Yonemochi H, Noda T, Saku T. Pericoronal hamartomatous lesions in the opercula of teeth delayed in eruption: an immunohistochemical study of the extracellular matrix. *J Oral Pathol Med* 27: 441-52, 1998.
- Philipsen HP, Thosaporn W, Reichart P, Grundt G. Odontogenic lesions in opercula of permanent molars delayed in eruption. *J Oral Pathol Med* 21: 38-41, 1992.

15. Sciubba JJ, Zola MB. Odontogenic epithelial hamartoma. *Oral Surg Oral Med Oral Pathol* 45: 261-5, 1978.
16. Baden E, Splaver T. Odontogenic gingival epithelial hamartoma: report of case. *J Oral Surg* 31: 932-5, 1973
17. Sigal MJ, Mock D, Weinberg S. Bilateral mandibular hamartomas and familial natal teeth. *Oral Surg Oral Med Oral Pathol* 65: 731-5, 1988.