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Peripheral odontogenic fibroma (WHO type) of the newborn: a case report

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Summary. *Aim.* The present paper reports a case of peripheral odontogenic fibroma (POF) (WHO type) in a newborn. The differential diagnosis and treatment were discussed.
Background. POF is well described in the literature, but this is the first report in a newborn.
Patient. A 4-month-old female newborn was referred to our department because of an exophytic, sessile, firm, and well-delimited lesion on the right upper alveolar ridge. The covering mucosa was apparently normal. The lesion measuring 10×3 mm was present since birth. The clinical diagnosis of congenital granular cell tumour (congenital epulis) or dental lamina cyst of the newborn was made. A conservative excisional biopsy was performed under local anaesthesia, and the specimen was submitted to histopathological examination.
Results. The microscopic examination revealed a pattern of POF (WHO type). Normal primary incisors teeth eruption, and no signs of recurrence were noted on 16 months follow-up.
Conclusion. Despite the rarity of POF in a newborn, this lesion should be included as a possible diagnosis to focal gingival growth.

Introduction

Odontogenic fibroma is a rare and controversial tumour because of the number of distinct types [1]. The World Health Organization (WHO) defined both variants of odontogenic fibroma, peripheral or intraosseous, as 'a fibroblastic neoplasm containing varying amounts of odontogenic epithelium'. It may contain dentin and/or material resembling cementum [2].

The peripheral odontogenic fibroma (POF) (WHO type) is the extra-osseous counterpart of the central odontogenic fibroma and exhibits a similar histological appearance. The POF is an elevated lesion that is not encapsulated and generally not ulcerated. It consists of relatively cellular fibrous connective tissue in which strands and remnants of odontogenic epithelium are scattered [3]. The epithelial component resembles the dental lamina formed during the early stages of odontogenesis. The lesion provides some evidence that it is originated from a recapitulation of dental lamina, because the epithelium is capable of producing inductive changes in the connective tissue that are similar to changes in the dental lamina during odontogenesis [4].

Clinically, POF affects a wide age group (range 2–80 years) with a low peak in the third decade, no racial predilection, and a slight female and mandibular preponderance, representing about 1·2% of all odontogenic cysts and tumours [5]. Recurrence rate is not precise because follow-ups were generally not available [5], but a case was reported by Michaelides [6]. It has a similar appearance to other focal growths of the gingiva, such as peripheral ossifying fibroma. Interdental lesions of POF often cause tooth separation [4]. The purpose of this article is to present an unusual case of a POF occurring on the upper alveolar ridge of the newborn.

Case report

A 4-month-old female Caucasian newborn was referred to the Stomatology Clinic at the School of Dentistry, Universidade Estadual de Montes Claros, Minas Gerais, Brazil because of an isolated, exophytic, sessile, firm, hard, and well-delimited lesion on the right upper alveolar ridge. The lesion,

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presenting since birth, was prominent with dimensions of 10×3 mm. No discharge or redness was observed and mucosa was normal in appearance (Fig. 1). The patient's physical status was good and investigation of her medical history was not relevant. She had not taken any medication. There were no significant radiographic findings on periapical view, performed with small standard film to verify possible calcification or osseous involvement. The clinical diagnosis of congenital granular cell tumour (congenital epulis) or dental lamina cyst of the newborn was made.

An informed consent was obtained from the parents, and a conservative excisional biopsy was performed under local anaesthesia as the treatment choice and to obtain the final diagnosis by means of specimen analysis under histophatological examination. The microscopic examination revealed a lesion consisting of cellular connective tissue with multiple small islands and strands of odontogenic epithelium resembling remnants of the dental lamina covered by stratified squamous epithelium (Figs 2 and 3). The histological diagnosis of POF was established. Normal primary incisors teeth eruption and no signs of recurrence were noted on 16 months follow-up (Fig. 4).

Discussion

The POF is an uncommon, benign, exophytic gingival mass of fibrous connective tissue covered with surface epithelium, containing odontogenic epithelium [6]. In 1982, Gardner described the nature of the POF, considering it as the extra-osseous counterpart of the central odontogenic fibroma [3]. The POFs are rare

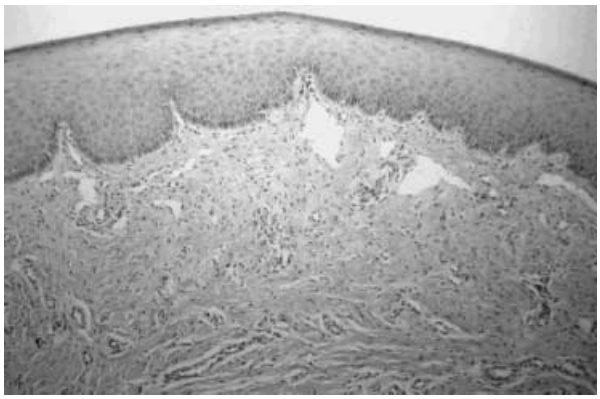

Fig. 2. Photomicrograph exhibiting the overlying epithelium of the lesion. The submucosa is composed of a dense fibrous connective tissue with zones of discrete loose connective tissue (haematoxylin & eosin, magnification 100×).

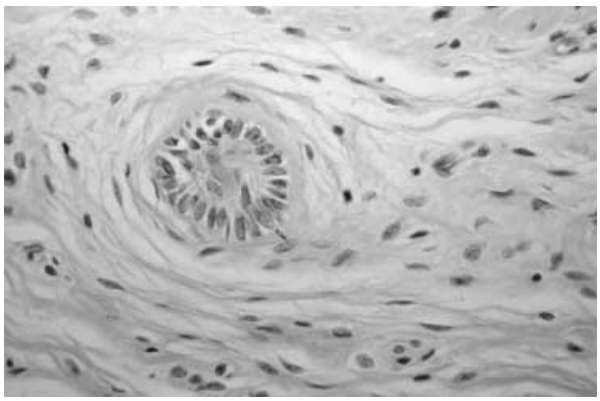

Fig. 3. Photomicrograph exhibiting cellular fibrous connective tissue with small islands of odontogenic epithelium. Nearby the islands, a myxomatous connective tissue is noted (haematoxylin & eosin, magnification 400×).

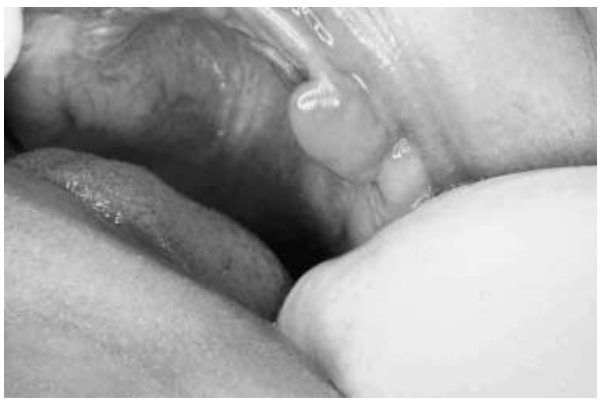

Fig. 1. Clinical view of an isolated, sessile, firm, hard, and well-delimited lesion located on the upper alveolar ridge of the maxilla of a newborn.

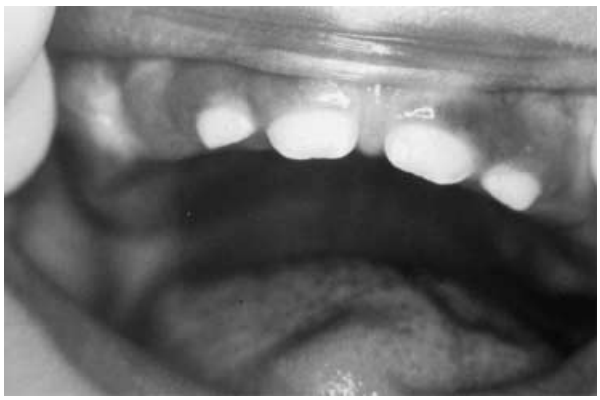

Fig. 4. Clinical view of 6-month follow-up showing irruption of the primary incisors and no signs of recurrence of the lesion.

lesions [7]. Siar and Ng (2000) showed results of their clinicopathological study in which POF comprises approximately 1.2% of all odontogenic cysts and tumours (according to WHO) [5].

Although POF usually presents as a localized gingival swelling, the clinical features were not sufficiently distinctive to separate them from the other gingival masses such as peripheral ossifying fibroma [1]. Despite clinical similarities, the peripheral ossifying fibroma is a reactive fibrous lesion of the gingiva, unlike the POF [8]. As a result of marked epithelial proliferation, the POF may be confused histologically with peripheral types of ameloblastoma or of calcifying epithelial odontogenic tumour. The epithelial islands of the POF are smaller than those of the ameloblastoma and do not exhibit such features of the basal cell layer, as hyperchromatism, intracytoplasmic vacuoles, and polarization of the nucleus away from the basement membrane, as found in various degrees in that tumour. Additionally, hard tissues, such as dentin or cementum-like are not found in ameloblastoma, although they are not present in all examples of POF [3,5].

There is another lesion that should be considered in the differential diagnosis of the POF. Occasionally, focal fibrous gingival hyperplasias contain remnants of odontogenic epithelium [3], but this hypothesis was displayed because of the age of the patient and the slow prevalence of gingival hyperplasias (0·08%) in children and youths [9]. Dental lamina cyst of the newborn, as named, is derived from the remnants of the dental lamina that remain in the soft tissues of the jaws. These cysts are generally seen on the alveolar ridges of newborn infants as small, often multiple swellings. The microscopic appearance consists of a superficially located thin-walled cystic lesion lined by a thin, stratified, squamous epithelium, and containing compacted desquamated keratin. Because these cysts usually resolve spontaneously in response to normal function, they require no treatment [10]. This clinical hypothesis established was a remote possibility, as the size of the unique lesion did not correspond to the literature.

Congenital granular cell tumour (congenital epulis) is a rare benign tumour that occurs on the gingival of the anterior alveolar ridge of the jaws, affecting females 8–10 times more often than males. This lesion may be sessile or pedunculated with red or normal colour, with size varying from millimetres to few centimetres in diameter. Histologically, it is similar to the granular cell tumour. The treatment of congenital epulis is generally surgical excision because spontaneous regression is rare [11], and incomplete excision has been no obvious tendency to recur [12]. This clinical diagnosis, representing the major possibility, led us to perform the surgical treatment by means of a conservative excisional biopsy as recommended in the literature. The histopathological examination revealed patterns of POF, and the treatment choice was coincident with the treatment employed [2,5]. This conservative local excision represents the definitive treatment and no complications were expected with it, because of the simplicity of the procedure and low morbidity, confirmed by follow-up.

In summary, we showed the clinical and histopathological findings of POF in a female newborn, emphasizing the need to consider this entity as a possible diagnosis by the clinician, when focal gingival growth was identified. The POF was well described in the literature, but this is a first report in a newborn.

What this paper adds

- Since this is the first report of POF in a newborn, it may contribute to improve our knowledge about this lesion.

Why this paper is important to paediatric dentists

- This paper may lead the paediatric dentists to consider this entity as a possible diagnosis to focal gingival growths of newborns.

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