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Case Report

Peripheral ossifying fibroma after natal tooth extraction: a case report

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Abstract: Peripheral ossifying fibroma (POF) is characterized by reactive gingival hyperplasia, and it is prevalent in adolescents, but rare in infants. Here, we present a case involving a 1-day-old boy with POF, diagnosed after the analysis of a soft tissue mass with a calcified structure that was found after the extraction of a natal tooth, and describe the clinical and histopathological features. This is an extremely rare case of POF associated with the extraction of a natal tooth. The findings suggest that POF can present as reactive gingival hyperplasia after natal tooth extraction. Clinicians should be cautious when dealing with a soft tissue mass with focal calcification and consider POF as a differential diagnosis. For an appropriate diagnosis, radiographic examination and excisional biopsy with histopathological examination will prove useful. Definitive early surgical intervention with thorough excision is recommended to prevent recurrence.

Keywords: Gingival hyperplasia, natal tooth, peripheral ossifying fibroma

Introduction

Peripheral ossifying fibroma (POF) is characterized by reactive gingival hyperplasia and is prevalent in adolescents [1]. Prepubertal patients are mostly not affected by POF, which is particularly rare in the first decade of life [2, 3]. The etiology of POF remains unclear, although various possible irritants, including calculus, plaque, microorganisms, dental appliances, and ill-fitting crowns, have been implicated [2]. Some reports state that POF originates from the periodontal ligament because of inflammatory hyperplasia [3, 4], while others have considered that it is related to the effects of hormones, because the conditions exhibit a high female predilection and a significantly decreased incidence after the third decade of life [5].

Natal teeth exist from birth, while neonatal teeth erupt within 30 days of birth. The incidence of natal and neonatal teeth varies from 1:2,000 to 1:3,500 [6]. Clinicians need to consider certain factors when deciding whether to extract or retain these teeth in the oral cavity; these include the degree of mobility, inconvenience and interference during breastfeeding, possibility of aspiration, and whether the tooth is a part of the normal dentition or a supernumerary tooth [6, 7]. The most common complication of natal teeth is the development of ulceration on the floor of mouth, a condition known as Riga-Fede disease. In order to eliminate potential discomfort during breastfeeding and prevent Riga-Fede disease, the incisal edges of natal teeth can be ground to decrease their roughness. If these teeth show excessive mobility, immediate extraction is the common approach [8].

Until recently, complications after the extraction of natal teeth have been reported in only a few cases. Some pathological gingival growths, including tooth-like structures, pulp polyps, hamartomas, and reactive fibrous hyperplasia have been associated with the extraction of natal and neonatal teeth [9-12]. Here, we present a rare case involving a 1-day-old boy who developed POF as a complication after the extraction of a natal tooth.

Case report

A 1-day-old boy was referred to the Department of Pediatric Dentistry at Wonkwang University

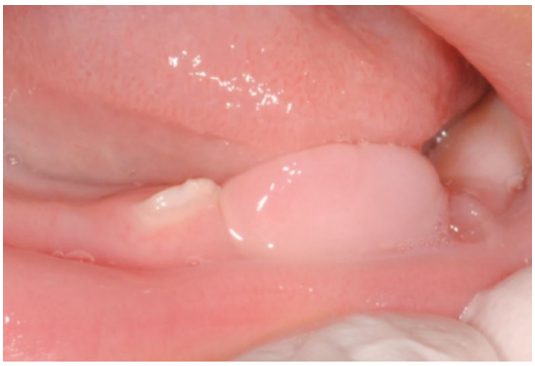

Figure 1. An intraoral photograph of a peripheral ossifying fibroma appearing as a pedunculated soft tissue mass 4 months after the extraction of a natal left mandibular central incisor in a 1-day-old boy.

Daejeon Dental Hospital for the evaluation of two primary natal teeth in the mandibular anterior region. The boy was born via normal vaginal delivery at 38 weeks of gestation, with no complications. He was healthy and weighed 2,640 g at birth.

The mandibular left primary central incisor was extremely mobile with minimal attachment to the surrounding gingiva, while the right primary central incisor was partially erupted and slightly mobile. Informed consent was obtained from the parents, and the left primary central incisor was extracted with forceps. Reassessment was recommended for the right primary central incisor.

Four months after extraction of the natal tooth, the infant returned for reassessment. The right primary central incisor had stabilized and did not appear to be mobile. However, clinical examination revealed a pedunculated soft tissue mass in the region of the extracted left primary incisor. The mass measured approximately 1.0 cm in diameter and was pink with a smooth, nonulcerated surface. The parent stated that the mass appeared a few weeks after the extraction and gradually increased in size (**Figure 1**). Radiographic examination revealed a soft tissue mass with a calcified radiopaque structure in the center. Three months later, the infant returned for follow-up assessment. The mass had increased in size and exhibited a stippled surface. Radiographic examination revealed obvious focal calcification in the mass (**Figure 2**), and we decided to perform an excision. An excisional biopsy was performed under local

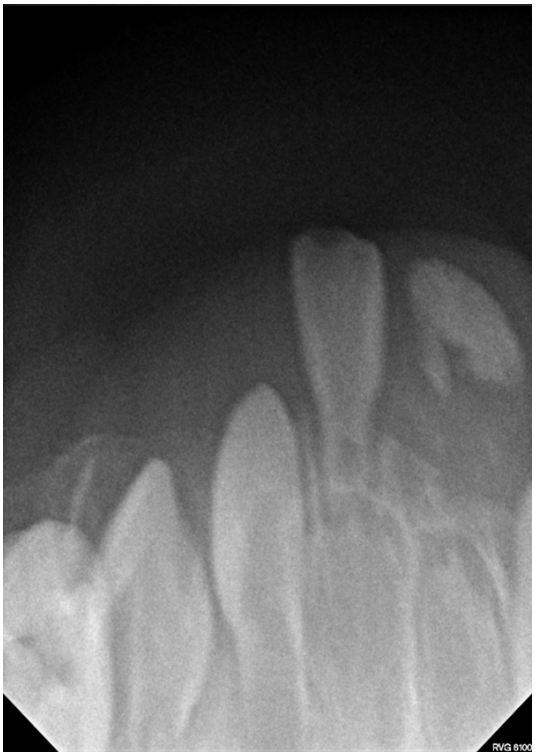

Figure 2. A periapical radiograph of a peripheral ossifying fibroma that developed after the extraction of a natal left mandibular central incisor in a 1-day-old boy. Focal calcification can be seen in the soft tissue mass.

anesthesia; the specimen obtained comprised whitish soft and hard tissue and measured 1.2 × 0.6 cm.

Histopathological examination revealed fibrous connective tissue with nonulcerated epithelial tissue and a focal calcified structure (**Figure 3**); there was fibrous connective tissue with irregularly placed fibroblasts below the epithelial tissue. The focal calcified mass exhibited osteocytes, osteoid, and some fibrous tissues. Osteoblasts covered the outer layer of the calcified mass (**Figure 4**). Because no dental papilla or dentinal tubules were observed, the calcified mass was considered to be a bone-like structure. The final diagnosis was POF.

The infant was followed up for 2 weeks after the excision. Uneventful postoperative healing was observed. No abnormality or recurrence was observed during a 3-year follow-up period, and the right primary central incisor was well maintained, despite its short root.

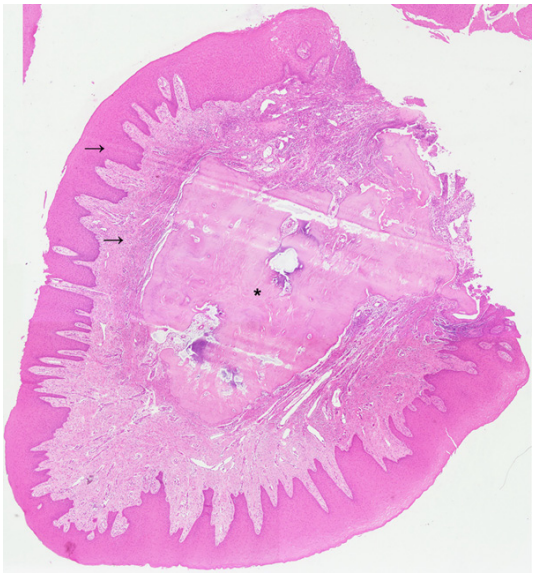

Figure 3. Histopthological analysis of an excised peripheral ossifying fibroma observed 4 months after the extraction of a natal left mandibular central incisor in a 1-day-old boy. Fibrous connective tissue with nonulcerated epithelial tissue (indicated by the arrows) and a focal calcified structure (indicated by *) can be observed.

Discussion

POF is generally described as localized, exophytic, gingival hyperplasia with a pedunculated or sessile base. The lesion can occur in both ulcerated and nonulcerated forms, and the color varies from red to pink [1]. A high female predilection and significantly decreased incidence after the third decade of life are observed, and there are very few reports of POF occurring in the first decade of life [3, 14].

POF is also referred to as reactive proliferation of the gingival mucosa, and the histogenetic origin of POF is considered to be the superficial periodontal ligament [2]. The etiology remains unclear, but trauma or local irritants, including calculus, plaque, microorganisms, dental appliances, and ill-fitting crowns, can stimulate the periodontal ligament to cause excessive proliferation of fibrous tissue and initiate osteogenesis with dystrophic calcification [4, 5, 13]. Eversole and Rovin [13] stated that the exfoliation of primary teeth and the eruption of permanent teeth can cause constant irritation, resulting in an increased incidence of reactive lesions presumed to arise from the superficial periodontal ligament.

Kohli et al. [14] previously reported a case involving a 6-month-old infant with POF that developed after the extraction of a neonatal tooth. The authors stated that extraction of a neonatal tooth stimulates active growth of the alveolar bone in a neonate, which may result in an exaggerated periosteal response and the formation of a reactive lesion with some potential for osteogenesis.

Until recently, only a few cases of complications after the extraction of natal teeth have been reported. Some cases exhibited tooth-like structures or masses with odontogenic components, including well-organized dentin, dentinal tubules, and cementum [9, 15]. Several reports have explained that a mass that develops after natal tooth extraction would originate from the remaining internal dental papilla, which continues to develop [9].

In the present case, a calcifying soft tissue mass developed after the extraction of a natal tooth. The case is unique in that POF, which is characterized by reactive gingival hyperplasia, developed from the periodontal ligament itself and did not result from the development of dental tissue associated with remnants of the natal tooth. We speculated that the development of this lesion was influenced by the stimulation provided by the extraction of a natal tooth; however, the presence of another natal tooth with slight mobility could also have stimulated the proliferation of POF. Stimulation of an exaggerated periodontal ligament response in the infant may have resulted in reactive gingival proliferation with ossification.

Radiographic examination is not commonly used for the diagnosis of soft tissue lesions. However, Kendrick and Waggoner [4] and Moon et al. [16] reported that focal calcification in POF can be detected on a plain radiograph, although computed tomography offers superior visualization. In the present case, we obtained a periapical radiograph, which clearly showed the focal calcification in the lesion. Thus, a radiographic examination may be useful for the detection of focal calcification in POF.

The diagnosis of POF is mostly based on excisional biopsy [17]. Histopathologically, POF is characterized by prominent areas of highly cellular fibrous tissue containing bony structures, osteoid, and other calcification foci [1-3]. Bu-

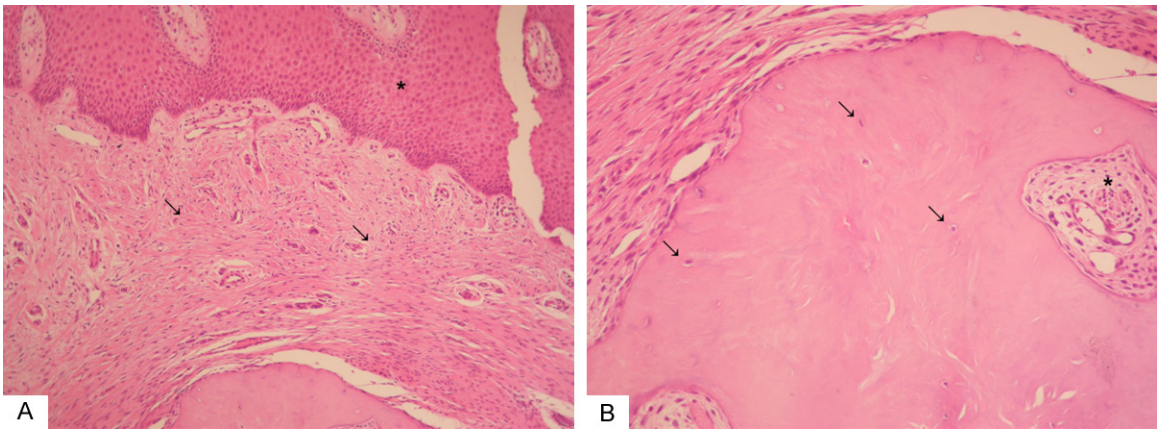

Figure 4. Histopthological analysis of an excised peripheral ossifying fibroma observed 4 months after the extraction of a natal left mandibular central incisor in a 1-day-old boy. (A) Nonulcerated epithelial tissue (indicated by * in A) and fibrous connective tissue with irregularly placed fibroblasts (indicated by arrows in A) can be observed. (B) The mass exhibits osteocytes, osteoid (indicated by arrows in B), and fibrous tissues (indicated by * in B).

chner and Hansen [1] reported 207 cases of POF and identified the histomorphological spectrum of the lesions. They reported that these lesions begin with a phase of epithelial ulceration, characterized by highly cellular fibroblastic connective tissue with dystrophic calcification and osteogenesis. Then, the ulceration heals and fibrous epulis forms with mineralized bone material [1, 5]. A few POFs contained scattered cementum-like material, referred to as cementicles, in addition to bone.

In children, reactive gingival lesions, including POF, can grow aggressively and significantly increase in size in a short period of time. In addition, POF can cause alveolar bone loss and displace or interfere with erupting teeth. Early recognition and definitive surgical intervention can lower the risk [3, 4]. Surgical excision or excisional biopsy is the most common treatment, and the recurrence rate reportedly varies from 7% to 45% [1, 5, 13]. Thorough deep excision of the lesion, including the periosteum, can prevent recurrence [3, 4].

In conclusion, we here reported a rare case of a 1-day-old infant who developed POF that presented as reactive gingival hyperplasia after the extraction of a natal tooth. Our findings suggest that clinicians should be cautious when dealing with a soft tissue mass with focal calcification and should consider POF as a differential diagnosis. To make an appropriate diagnosis, radiographic examination may prove useful, while excisional biopsy with histopathological examination should be performed. Definitive early surgical intervention with thorough excision is recommended to prevent recurrence.

Disclosure of conflict of interest

None.

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