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Co-Occurrence of Natal Teeth With Rapidly Involuting Congenital Hemangioma: A Case Report

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Abstract

We present in this report a case of an infant girl who was born with natal teeth along with an ulcerated rapidly involuting congenital hemangioma (RICH) on her thigh. RICH is a benign vascular tumor that is more common than other congenital hemangioma subtypes. Natal teeth are a birth anomaly that occurs in isolated form or in association with different syndromes like Ellis-van Creveld, Pierre Robin, Soto and Hallerman-Streiff. We have performed an intensive literature search to determine if there is a link between natal teeth and RICH, but no article has described any association thus far. It remains unclear whether this is a mere coincidence or whether there are common genetic factors that play a role in benign tumor growth and eruption of premature teeth. Further reports on the co-occurrence may shed light on possible common risk factors contributing to natal teeth and congenital hemangioma.

Keywords: Natal teeth; Rapidly involuting congenital hemangioma; Ulceration

Introduction

Congenital hemangiomas are benign vascular tumors with low incidence rate. They grow *in utero* and reach full growth at birth. On the other hand, infantile hemangiomas are more common and appear after birth [1]. Congenital hemangiomas appear in the head or limbs more often than in other locations in affected infants. They can be classified into two types based on the course of progression: rapidly involuting congenital hemangioma (RICH) and non-involuting congenital hemangioma (NICH). Their sizes vary from a few centimeters to more than 10 cm, and they are typically solitary although multifocal lesions have been reported [2, 3]. RICH involutes completely by the age of 14 months whereas NICH never regresses, but grows in proportion with the child size and may eventually require excision [2].

Another uncommon congenital anomaly is natal teeth. Depending on the time of appearance in the oral cavity, teeth that are present at birth are called natal teeth, while teeth that erupt within the first 30 days from birth are called neonatal teeth [4]. The lower central incisor in the mandibular region is the most frequent location for both natal and neonatal teeth. Natal teeth are multifactorial disorder with some defined genetic components. They have been reported in association with different disorders such as Ellis-van Creveld, Soto, Pierre Robin and Hallerman-Streiff syndromes [5]. In our intensive literature search, we did not find any record of natal teeth in association with RICH, and our personal communications with some pediatricians did not confirm the association between these two entities together due to the sporadic occurrence of RICH. Therefore, we think that this association is rare but worth reporting because it remains unclear whether this is in fact an association or a mere coincidence.

Case Report

This is a case report of a Hispanic newborn girl that was diagnosed with RICH associated with two incisors natal teeth (Fig. 1A). Birth was spontaneous vaginal without complications. Amniotic fluid was stained with meconium. Apgars were 9/9 at 1/5 min. Exam after delivery revealed a 7.5 × 5.5 cm grayish telangiectatic tumor on the outer upper right thigh. There was a small, roughly circular erosion centrally with surrounding pink scarring; a large feeder artery at 12 o'clock was noticed. Two firmly fixed lower incisor teeth were present, compatible with natal teeth. The scalp, face, neck, chest, back, axillae, abdomen, groin, external genitalia, buttocks, bilateral upper and lower extremities including the hands and feet were normal. The dif-

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Figure 1. An infant girl born with two natal incisors (A), and with rapidly involuting congenital hemangioma on her thigh (B).

ferential diagnosis of the hemangioma was RICH, NICH or arteriovenous malformation. A dermatology referral was made. The patient was prescribed mupirocin ointment on a non-stick gauze and dressing twice daily (Fig. 1B). The newborn screen and CBC were normal. Her teeth were not loose and as they did not interfere with feeding, no intervention was carried out. In the following weeks, close observation showed the lesion to be involuting centrally and peripherally (Fig. 2A). The central erosion was fading (Fig. 2B), and thus the hemangioma was categorized as RICH. Mupirocin was stopped after 6 weeks.

Discussion

The precise incidence of congenital hemangiomas is unknown. In a prospective study of 594 infants born in Mary Birch Hospital for Women in San Diego, 29 infants out of 594 were identified with hemangiomas. Two out of the 29 had congenital hemangiomas subtype (0.3%) and 27 infants had infantile hemangiomas (4.5%) [6]. Most cutaneous hemangiomas follow a benign course without any complications and they often require no intervention. Some lesions may ulcerate, bleed or leave a scar. Other complications include pyogenic granuloma-like growth [7], a life threatening hemorrhage and thrombocytopenia [1, 8]. Although most hemangiomas are isolated anomalies, several studies have reported that hemangiomas may run in families as an autosomal dominant trait suggesting the involvement of causative genetic factors [9]. Genetic mutations have been identified in the inherited forms of vascular anomalies but they have not been replicated in other independent studies [10].

In contrast, the incidence of natal and neonatal teeth is 1:2,000 - 1:3,500 in European and Asian population, respectively [11, 12]. There is controversy concerning the sex distribution; some studies reported more females affected than males [13]. An example is the study by Barfiwala et al which found 35 out of 50 cases were in females [14]. Occasionally, natal teeth are associated with complications such as Riga-Fede disease in which an oral ulceration manifests itself on the ventral surface of the tongue or on the inner surface of the lower lip [15]. It is caused by trauma to the soft tissue from erupted teeth. Another morbidity associated with natal teeth is ingestion or aspiration of the teeth if they are loose. Feeding difficulties can occur with injury to the mother's breast.

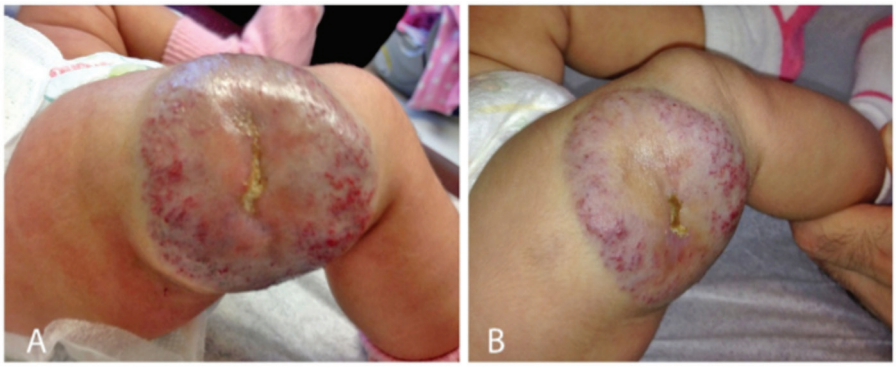

Figure 2. A depiction of rapidly involuting congenital hemangioma 4 weeks postpartum (A). Regression of the lesion and healing of the ulceration 9 weeks postpartum (B).

A retrospective study by Moura et al on 23 children with natal or neonatal teeth showed that most cases were natal teeth (83%) and presented a mild degree of mobility (64%). Only two patients had breast-feeding difficulties (9%). Tooth maintenance was the most common treatment of choice (64%), and only two patients with Riga-Fede disease presented with oral ulceration (9%) [16]. The treatment of the natal and neonatal teeth is usually guided towards avoiding the possible complications. Observation and extraction are the main options. Extraction should be done only if the teeth are loose or if they cause feeding difficulties for the mother or the infant.

In conclusion, this report is the first to document the co-occurrence of natal teeth with a congenital hemangioma. While it appears to be unique, further reports could shed light on frequency and on the contribution of possible genetic factors.

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Conflict of Interest

The authors declare that there is no conflict of interest regarding the publication of this report.

Author Contributions

Dr. Sakkalaek drafted the initial manuscript. Dr. Klein and Dr. Fakhouri reviewed the manuscript, modified it and approved the final manuscript as submitted. All authors have made substantive contribution to this manuscript, and all have reviewed the final paper prior to its submission.

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