Dental Traumatology

Dental Traumatology 2009; **25**: e48–e50; doi: 10.1111/j.1600-9657.2009.00773.x

Sublingual traumatic ulceration (a Riga-Fede disease): report of two cases

CASE REPORT

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Accepted 1 January, 2009

Abstract – Sublingual traumatic ulceration is rare and a benign, ulcerative and granulomatous process that occurs as a result of repetitive trauma of the oral mucosal surfaces by the teeth. Treatment should begin conservatively and should focus on eliminating the source of the trauma. In our cases, we chose the conservative treatment by grinding the teeth and placing composite resin over the offending teeth and applied orabase. At the follow-up, we confirmed that the lesion was resolved and infant was feeding normally.

Ulceration on the ventral surface of the tongue in newborn babies or infants is most commonly related to natal or neonatal teeth (1). Ventral lesions result from contact with mandibular anterior incisors. Rarer lesions on the dorsal surface are associated with the maxillary incisors (2). This particular type of ulceration has been called a Riga-Fede disease (3, 4).

Sublingual traumatic ulceration is rare and a benign, ulcerative, and granulomatous process that occurs as a result of repetitive trauma of the oral mucosal surfaces by the teeth (5). Treatment should begin conservatively and should focus on eliminating the source of the trauma. Failure to diagnose and properly treat this lesion can result in dehydration and inadequate nutrient intake with other medical sequelae (4).

The lesions begin in early infancy and can present signs of a developmental anomaly or underlying neurologic disorder, including both incidental and syndrome-associated natal teeth and inherited sensory neuropathies.

Case report

Case 1

An 8-month-old boy was referred to the department of pediatric dentistry in Kyunghee Dental Hospital with an extensive lingual ulceration. The parents reported that he had habitually bitten his tongue during 1 month and he was a poor feeder, because of the painful ulcer on the ventral surface of the tongue with intermittent bleeding in tongue (Fig. 1). There was no history of high fever, abnormal sweating, seizure, or natal teeth. There was no family history of developmental disorders and congenital syndromes.

On physical examination, the lesion was circular (3 cm × 2 cm) with erythematous, raised, and indurated border with overlying yellowish white slough on the ventral surface at the junction of attachment of lingual frenum (Figs 1 and 2). There are no clinical signs of tongue-tie. No biopsy was performed because of the age of the infant, the particular site of the lesion and the clinical evidence of diagnosis.

Based on these clinical findings, the lesion was diagnosed as Riga-Fede disease. We treated the oral ulceration by placing composite resin to smooth the incisal edges of his lower incisors and treated symptoms using corticosteroids in orabase. Within 2 months the ulcer healed completely. The child gradually regained weight without any relapse during the 6-month follow-up.

Case 2

A 2-month-old girl was referred to the department of pediatric dentistry in Kyunghee Dental Hospital with an extensive lingual ulceration from local dental clinic. The parents said that she had a natal tooth and habitually bitten her tongue from birth, but there was a little problem in feeding, because of ulcer on the ventral surface of the tongue (Fig. 3). There was no history of high fever, abnormal sweating and seizure. There was no family history of developmental disorders and congenital syndromes.

Fig. 1. Initial photograph of 8-month-old boy with tongue ulcer lesion caused by primary lower anterior teeth.

Fig. 2. Final photograph, 2 months after resin composite build-up. The lesion was resolved.

Fig. 3. Initial photograph of 2-month-old girl with tongue ulcer lesion caused by neonatal tooth.

On physical examination, she had a natal tooth on #81 with grade one mobility. The lesion was circular (2 cm × 2 cm) with indurate, fibrous ulceration on the ventral surface at the junction of attachment of lingual frenum (Fig. 3). There are no clinical signs of tongue-tie. No biopsy was performed because of the age of the infant, the particular site of the lesion and the clinical evidence of diagnosis.

Fig. 4. Final photograph, 1 month after grinding the sharp incisor edge. The lesion was resolved.

Based on these clinical findings, the lesion was diagnosed as Riga-Fede disease. We treated the oral ulceration by grinding of the incisal edges of her lower incisors and treated symptoms using corticosteroids in orabase. Within 1 month the ulcer healed completely. The child and parents was satisfied (Fig. 4).

Discussion

Ulceration of the ventral surface of the tongue in newborn babies or infants is most commonly related to neonatal or natal teeth. Another onset of the lesions usually coincides with the eruption of the primary teeth, principally the lower incisors, at 6–8 months of age (3). The lesion was first described by Antonio Riga in 1881. Histological studies and additional case reports were subsequently published by Fede in 1890 (5). Thereafter, this disease is known as Riga-Fede disease.

This benign ulceration occurs as a result of repetitive trauma of the oral mucosal surfaces by the teeth. Most commonly, the reactive ulceration arises on the ventral surface of tongue or the lingual frenum along the midline as the tongue is raked over the teeth. The presence of natal teeth and primary teeth combined with the instinctive infantile tongue thrusting reflex may also cause a traumatic ulceration to arise on the ventral surface of the tongue (6).

Riga-Fede disease often heralds an underlying developmental or neurologic problem, including familial insensitivities to pain. It has been well described in association with familial dysautonomia (6). Other neurological disorders related to self-mutilation, such as Lesch-Nyhan syndrome and Tourette's syndrome (7, 8). Therefore, it is important to note that this lesion may be the initial presentation of some serious underlying medical problems.

Several treatments for Riga-Fede disease have been described, all of which treatments aim to minimize the associated trauma (6, 9). Corticosteroids in orabase can be applied in order to relieve symptoms. It is reasonable to try conservative treatment at first; for example, grinding the sharp incisor edges, placing composite resin

in dome shape, or placing protective ring. If conservative treatment do not lead to healing of the wounds, it may be necessary to choose radical treatment, such as extract the teeth or treat the lesion by radiation or excise the lesion (10, 11). In our cases, we choose the conservative treatment by using grinding the teeth and placing composite resin over the offending teeth and applied orabase. At the follow-up, we confirmed that the lesion was resolved and infant was feeding normally. We think that these conservative treatments can resolve the ulcer.

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