

Lingual Traumatic Ulceration (Riga-Fede Disease): Report of a Case and Review

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Abstract

The term Riga-Fede disease has been used to describe chronic traumatic ulceration that occurs on the ventral surface of the tongue by natal and neonatal teeth in infants. It is important to diagnose the lesion and treat it by eliminating the source of trauma. In this case report, the neonatal tooth was extracted and after 2 weeks the lesion was resolved and the baby was feeding well. In addition, a review of dental and medical literature is included.

Key words: Riga-Fede disease, natal teeth, neonatal teeth.

Introduction

Riga-Fede disease is a chronic, benign, ulcerative granulomatous process that occurs as a result of continuous trauma on the ventral surface of the tongue most commonly caused by neonatal or natal teeth in newborns (1,2). It may also be associated with repetitive tongue thrusting habits in older infants after the eruption of primary lower incisors (3). This condition has also been reported in children with familial dysautonomia (4).

The lesion was first described by Antonio Riga, an Italian physician in 1881. In 1890, Fede published the first histological studies. Subsequently, it was called "Riga-Fede disease" (5).

This condition may interfere with proper feeding which, in turn, may pose potential risks to infants due to nutritional deficiency. Consequently, proper dental management for these patients must be considered.

Case Report

A 38-day-old girl was referred to the Department of Pediatric Dentistry of Yazd University of Medical Sciences in June 2011, whose mother complaining of a lesion on the ventral surface of her tongue for 1 week. The mother stated that her baby had pain during suckling and could not be nursed well.

The medical history was not worthy of note. The infant's development was normal and she had received all her scheduled vaccine shots.

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Intraoral examination revealed one crown (tooth) in the anterior region of the mandibular arch. According to the mother, the tooth had erupted 10 days after birth. The neonatal tooth exhibited grade 2 mobility. The ventral surface of the tongue showed a $10\text{mm}\times 12\text{mm}$ ulceration that extended from the lingual frenum to the anterior border of the tongue (Fig. 1).

On palpation, the area elicited a pain response. There were no other lesions in any parts of the mouth. Based on the clinical findings, the diagnosis of Riga-Fede disease was made.

Because of the mobility of the neonatal tooth and the risk of aspiration, the extraction of neonatal tooth was considered as the treatment choice compared to more conservative alternatives.

There was no contraindication for extraction. The tooth was removed under topical anesthesia. In the 12-day recall, the lesion had healed completely. The mother was satisfied with the infant's improved feeding (Fig. 2).

Review

A literature search was performed for all reported cases of Riga-Fede disease in the English literature of PubMed, Cochrane, Science direct, Google Scholar and Scopus databases. A summary of these case reports is shown in Table 1.



Figure 1. Riga-Fede lesion on the ventral surface of the tongue caused by neonatal tooth



Figure 2. The healed lesion 12 days after tooth extraction

Table 1. Summary of all reported cases of patients with Riga-Fede disease

Authors	Year	Gender	Age (Month)	Site	(NEO) Natal Teeth	Treatment
Amberg (6)	1902	M	7	sublingual	no	excision
Bray (6)	1927	M	9	sublingual	no	excision
Bradley (6)	1932	F	8	sublingual	no	excision
Moncrieff (6)	1933	M	6	sublingual	no	weaning
Newman (6)	1935	M	6	sublingual	no	smoothing lower incisors
		M	8	dorsum of tongue	no	excision
Abramson (5)	1944	F	11	sublingual	no	excision
		F	9	sublingual	no	excision
Jacobs (7)	1956	unknown	0.3	sublingual	yes	excision
McDaniel et al (8)	1978	M	6	dorsum of tongue	no	excision
Rakocz(4)	1987	M	10	base and dorsum of tongue	no	composite coverage incisors
Tomizawa et al (9)	1989	M	1	sublingual	yes	composite coverage incisors
		M	5	sublingual	yes	composite coverage incisors
Eichenfield et al (10)	1990	F	6	sublingual	no	none
Goho (1)	1996	F	0.7	sublingual	yes	excision

		F	0.3	sublingual	yes	composite coverage incisors
Uzamiş (11)	1999	M	2	sublingual	yes	extraction
Slayton (3)	2000	M	10	sublingual	no	smoothing lower incisors
Toy (12)	2001	M	20	sublingual lower lip	no	unknown
Baghdadi (13)	2001	M	10	sublingual	no	smoothing lower incisors topical corticosteroid
Baghdadi (14)	2002	F	12	sublingual	no	smoothing lower incisors topical corticosteroid
Terzioğlu et al (15)	2002	M	7	sublingual	no	none
Zaenglein et al (2)	2002	M	10	tongue lower lip	no	unknown
Ahmet et al (16)	2003	F	9	sublingual	no	none
Hegde (17)	2005	F	1	sublingual	yes	extraction
Campos-Muñoz et al (18)	2006	M	11	sublingual	no	nasogastric feeding tube
Baroni et al (19)	2006	M	11	sublingual	no	topical odontologic cream teething ring
Domingues- Cruz (20)	2007	M	24	lower lip	no	extraction
Narang et al (21)	2008	M	9	sublingual	no	teething ring release of tongue tie
Jariwala et al (22)	2008	F	1.5	sublingual	yes	extraction
Ceyhan et al (23)	2009	M	15	sublingual	no	topical corticosteroid
Taghi et al (24)	2009	M	8	sublingual	no	composite coverage incisors
Choi et al (25)	2009	M	8	sublingual	no	composite coverage
		F	2	sublingual	no	incisors smoothing incisal edges
Eley et al (26)	2010	F	11	sublingual	no	excision
Dubois et al (27)	2010	unknown	6	tongue lower lip	no	Unknown
Nagaveni et al (28)	2011	M	1	Unknown	yes	extraction
		F	1	unknown	yes	extraction
		M	1	unknown	yes	extraction
		F	1	unknown	yes	extraction
		M	1	sublingual	yes	vitamin K administration +
		F	1	unknown	yes	extraction
		F	1	unknown	yes	extraction
		M	1	unknown	yes	extraction
		M	1	sublingual	yes	extraction
		F	1	unknown	yes	extraction
		M	1	unknown	yes	extraction
		F	1	unknown	yes	extraction
		M	1	unknown	yes	extraction

		M	1	sublingual	yes	vitamin K administration +
		F	1	unknown	yes	extraction
						extraction
						extraction
Van der Meij et al (6)	2012	M	6	sublingual	no	excision
Marie et al (29)	2012	F	10	lower lingual apex	no	unknown
Costacurta et al (30)	2012	F	2	ventral surface of the tongue	yes	extraction
Rachel Dunlop (31)	2013	F	2	anterior ventral tongue	yes	extraction

Discussion

The teeth observed at birth or during the first 30 days of life are called natal and neonatal teeth. In the past, this eruption anomaly was superstitiously associated with good or bad omens in the folklore. This explains the abundance of reports about this condition since 59 B.C., as observed in cuneiform inscriptions discovered in the 19th century (25).

Riga-Fede disease is initiated by the erosion produced by the sawing of the lingual frenum on the sharp, cutting edges of lower central incisors. With repeated trauma and secondary infection, granulation tissue forms and produces a tumor like mass, which is usually disc-shaped and about 1mm in diameter. In the center, there is usually an ulcerated area covered with fibrinous pseudomembrane. The lesion sometimes causes difficulty in nursing but there are no other symptoms. There is no associated lymphadenopathy or adjacent inflammation.

In this case report, Riga-Fede disease is associated with neonatal teeth and is similar to 29 cases which have been reported since 1902.

According to all case reports of Riga-Fede disease since 1902 including ours, the most common site where the ulcer occurs, is the sublingual region of the mouth.

Treatment of the disease should begin conservatively and should focus on eliminating the source of trauma. Failure to diagnose and late treatment can result in dehydration and malnutrition. For traumatic lesions which have a clear irritating factor, the factor should be removed first. If healing does not occur after 2 weeks, biopsy is indicated (32).

There are several treatment options for Riga-Fede disease but conservative method was chosen as treatment option over extraction of the teeth. In the case of mild to moderate irritation to the tongue, a conservative approach such as smoothing the incisal edge with an abrasive instrument is advocated (17). Alternatively, a small increment of composite resin may be bonded to the incisal edges of the teeth (25). Among several reported cases since 1902, in 24 cases the teeth

were extracted. Composite restoration and incisal edge smoothing were only performed in 6 and 5 cases, respectively. Because of the risk of aspiration and the difficulty of placing a composite restoration, it seems that many clinicians prefer to extract the irritating teeth. In this case because of mobility of the tooth and the risk of aspiration, the tooth was extracted. In such a case; this procedure should not pose any difficulties since the teeth can be removed with the forceps or even with fingers. Defer the extraction until the 10th day of life to prevent hemorrhage and assess the need for administering vitamin K before the operation. This waiting period is necessary in order for the commensal flora of the intestine to establish and produce vitamin K as well as for prothrombin production in the liver (33). For our 38-day patient, however, this waiting period was not necessary and tooth extraction was performed safely without the need for vitamin K administration. Two weeks after tooth extraction, the lesion was fully recovered and the infant was feeding normally.

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