

Treatment alternatives for sublingual traumatic ulceration (Riga-Fede disease)

Rebecca L. Slayton, DDS, PhD

Dr. Slayton is an assistant professor, Department of Pediatric Dentistry, The University of Iowa, College of Dentistry. Correspond with Dr. Slayton at rebecca-slayton@uiowa.edu

Abstract

The term Riga-Fede disease has been used historically to describe a traumatic ulceration that occurs on the ventral surface of the tongue in neonates and infants. It is frequently associated with natal or neonatal teeth but may also occur in older infants after the eruption of primary lower incisors. Failure to diagnose and properly treat this lesion can result in dehydration and inadequate nutrient intake for the infant. Treatment should begin conservatively and should focus on eliminating the source of trauma. A case is presented in which modification of sharp tooth surfaces by the Pediatric Dentist and changes in feeding techniques by the parent were used successfully to resolve this lesion. By working together, the parent and the Pediatric Dentist can achieve positive results in a short period of time with minimal trauma to the infant. (Pediatr Dent 22:413-414, 2000)

raumatic ulceration of the ventral surface of the tongue (Riga-Fede disease) is most often associated with natal or neonatal teeth in newborns. 1,2 It is also seen in older infants, in children with repetitive tongue thrusting habits, and in children with familial dysautonomia (insensitivity to pain).³ The lesion was first described by Antonio Riga, an Italian physician, in 1881.4 Histologic studies and additional cases were subsequently published by F. Fede in 1890.⁵ Various terms have been applied to this lesion, including Riga's disease, Riga-Fede's disease, sublingual ulcer, sublingual granuloma and traumatic sublingual ulceration.6

This lesion was of particular concern in Italy in the late 1800s because it was frequently associated with malnourished infants and often resulted in death.^{4,7} In the United States, it was noted that the lesion was primarily seen in healthy infants shortly after the eruption of primary lower incisors. In some cases, it was also thought to be diagnostic of whooping cough.⁴ It was the general consensus of these early physicians that the lesion was of traumatic origin and that its treatment should involve elimination of the trauma. 4,5,7

The lesion begins as an ulcerated area on the ventral surface of the tongue. With repeated trauma, it may progress to an enlarged, fibrous mass with the appearance of an ulcerative granuloma. In an infant, the pain associated with an ulcerated oral lesion often results in dehydration, feeding difficulties and failure to thrive. In a child with other medical concerns, the potential for infection can add to the complications.

As is the case for most other traumatic lesions, the primary objective should be to eliminate the source of trauma so healing can take place. This may be accomplished in a number of ways. Unless the child is severely dehydrated or malnourished, it is preferable to begin treatment conservatively. Treatment

Table 1. List of Treatment Options

- 1. Smooth off incisal edges of lower incisors
- 2. Modify feeding behavior or feeding device
- 3. Treat symptoms—Kenalog in Orabase
- 4. Place increment of composite over the edges of O & P
- 5. Do nothing
- 6. Extract lower incisors
- 7. Excise the lesion

in previous reports has focused primarily on the teeth and virtually ignored feeding behaviors. To achieve the best success, all of the factors contributing to the trauma should be addressed. Teeth can be modified by smoothing the edges of the lower incisors with a finishing bur or by using a sandpaper disk. Alternatively, a small increment of composite may be bonded to the incisal edges of the teeth. Treatment with an ointment such as Kenalog in Orabase may be done to relieve symptoms enough to allow the child to nurse. The child should be assessed daily for dehydration. If conservative treatment options do not lead to rapid resolution of this lesion, it may be necessary to extract the lower incisors. It is usually not necessary to remove the lesion itself, as it will normally resolve after the trauma is eliminated. However, if the lesion persists after removal of teeth, an excisional biopsy should be performed.



Fig 1. Intraoral photograph at the time of the initial examination. A soft tissue enlargement with an ulcerated surface is evident on the ventral surface of the tongue.



Fig 2. Sippy cup with a modified lid. Holes were enlarged in the lid to increase flow of liquids.

Feeding behaviors also contribute to the trauma that causes this type of lesion. To address this, parents can modify the feeding methods so that trauma to the tongue is reduced. This may involve using a bottle with a larger hole in the nipple or a sippy cup that requires less vigorous sucking. Depending on the age of the infant, the parent can also attempt feeding by spoon to minimize trauma to the tongue. A summary of treatment options from most to least conservative is shown in Table 1.

Case description

A 10 month-old caucasian male with Down syndrome was referred to our clinic by his pediatrician because of a non-healing oral ulcer of approximately 2 months duration. In addition to his primary diagnosis, the infant was born with an atrial septal defect that was surgically repaired at age 2 months. On examination, there was a 2 cm mass with an ulcerated surface, located on the ventral surface of the tongue (Fig 1). The parents reported that they became aware of the initial lesion sometime after the eruption of the lower incisors (O and P). They had consulted their pediatrician and had been advised to apply Kenalog with Orabase to relieve the symptoms. The child had a moderate appetite but was not gaining weight.

Smoothing of the incisal edges was accomplished with a finishing bur in a high-speed handpiece. The child was positioned knee to knee with the mother restraining the hands. In addition, the parents were advised to modify their feeding methods to try to avoid the child positioning his tongue over the teeth and to minimize the amount of sucking needed to obtain fluids.

One-week follow-up was conducted by phone due to the distance the family would have to drive to come to the clinic. At that time, the parents reported that the ulceration had healed and the enlarged mass was almost completely resolved. They reported that they had switched to a different type of sippy cup that could be modified so that the holes in the spout were larger and required less effort to drink from (Fig 2). At a one month follow-up exam, the lesion had completely resolved and the child was able to feed comfortably (Fig 3).



Fig 3. Intraoral photograph at one-month follow-up visit. Lesion has resolved completely.

Discussion

This child continues to be at risk for recurrence of this lesion because of his habit of positioning his tongue back and forth across the lower incisors. However, early detection and intervention by the parents will help to minimize its progression.

In this case, conservative treatment resulted in the resolution of a difficult lesion. It is essential that any source of trauma be eliminated in order for this type of lesion to heal. In this case, modifying the teeth to reduce sharp edges and switching to a different type of feeding cup was sufficient to allow the lesion to resolve. Much of the credit for this success goes to the parents who were diligent in their efforts to minimize the re-traumatization of the lesion.

References

- 1. Goho C: Neonatal sublingual traumatic ulceration (Riga-Fede disease): reports of cases. ASDC J Dent Child 63:362-64, 1996.
- 2. Buchanan S, Jenkins CR: Riga-Fedes syndrome: natal or neonatal teeth associated with tongue ulceration. Case report. Aust Dent J 42:225-27, 1997.
- 3. Rakocz M, Frand M, Brand N: Familial dysatonomia with Riga-Fede's disease: report of case. ASDC J Dent Child 54:57-59, 1987.
- 4. Moncrieff A: Sublingual ulcer: with special reference to Riga's disease. Br J Child Dis 30:268-74, 1933.
- 5. Abramson M: Sublingual granuloma in infancy. J Pediatr 24:195-98, 1944.
- 6. Elzay RP: Traumatic ulcerative granuloma with stromal eosinophilia (Riga-Fede's disease and traumatic eosinophilic granuloma). Oral Surg Oral Med Oral Pathol 55:497-506, 1983.
- 7. Amberg S: Sublingual growth in infants. Am J Med Sci 126:257-69, 1903.