

Case Report

Sublingual Tongue Deformity in Infants: Riga-Fede Disease - A Case Report

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ABSTRACT:

Traumatic ulceration that occurs on the ventral surface of the tongue in neonates and infants has been described as the Riga-fede disease. Most commonly associated with natal or neonatal teeth but can also occur due to primary lower incisors. Failure to diagnose at an early stage can affect the overall health of the infant. Conservative treatment approach is always recommended followed by surgical excision of the lesion in case the earlier did not yield positive results. Here we present a case of riga-fede disease in an 8 month old infant due to trauma from deciduous lower incisors.

Key words: Riga-fede disease, tongue deformity.

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INTRODUCTION

Traumatic ulceration occurring on the ventral surface of the tongue due to repetitive protrusive and retrusive movements over the lower incisors in neonates is termed as riga-fede disease. It is an uncommon benign mucosal disorder caused by natal or neonatal teeth, sometimes may be due to primary incisors.¹ This condition in infants has been given various other names like traumatic lingual ulceration, eosinophilic granuloma and traumatic eosinophilic ulceration of the tongue and oral mucosa, sublingual fibrogranuloma, sublingual growth in infants, traumatic atrophic glossitis.²

This disease was first identified, by an Italian physician, Antonio Riga in 1881 and was described by Fede in 1890 who performed the histological studies of the lesion thus being called Riga-Fede disease. The

expression “traumatic ulcerative granuloma with stromal eosinophilia” (TUGSE) was coined by Elzay in 1983, referring to a chronic but self-limiting reactive ulcer of the oral mucosa.²

Initially the lesion begins with ulceration on the ventral surface of the tongue, but with repeated trauma may lead to the formation of an enlarged fibrous mass resembling an ulcerative granuloma. Pain caused due to ulceration can lead to feeding difficulties in infants affecting nutrient intake leading to dehydration with other medical sequelae.³

Treatment focuses on eliminating or reducing the factors causing trauma. Treatment depends on the tooth's mobility and the risk of aspiration or swallowing; whether it is supernumerary or regular primary teeth; whether it is causing interference in

breastfeeding; breast and oral soft tissue injuries; and the general state of child's health.⁴

CASE REPORT

A 7 month-old male infant was referred to our department of dentistry by the department pediatricis (Rajendra Prasad government Medical College & Hospital, Tanda, Kangra, Himachal Pradesh) because of a non-healing oral ulcer on the anterior ventral surface of the tongue in the midline since last two months. The mother complained that the teeth were making breastfeeding difficult, irritating the infant who could not manage to suck and then cries continuously. Intraoral examination revealed two natal teeth in the mandibular anterior region and an ulcerated lesion on the ventral surface of the tongue. The lesion had a diameter of 12 mm and was located at the midline anterior portion of the ventral surface of the tongue (fig-1). On palpation lesion was well defined, firm in consistency and tender. The lesion had the impression of the teeth on its center due to repetitive trauma resulting from raking movements of the tongue against the anterior incisor teeth leading to the clinical diagnosis of Riga-Fede Disease. To prevent the repetition of the trauma on the tongue, reduce the discomfort to infant, and to allow wound healing, the sharp incisal edges were smoothened with a finishing bur. The mother was advised to bottle feed the infant for one week. On examination after a week the lesion was reduced in size and central ulceration was not present (Fig-2). The infant was in a better and more comfortable state with improved feeding and sleeping pattern. To rule out any medical condition associated with this, the infant was examined by the pediatrician and no underlying neurological disorder was identified. The mother refused for the surgical excision of the lesion thus the infant was kept under observation and follow up.

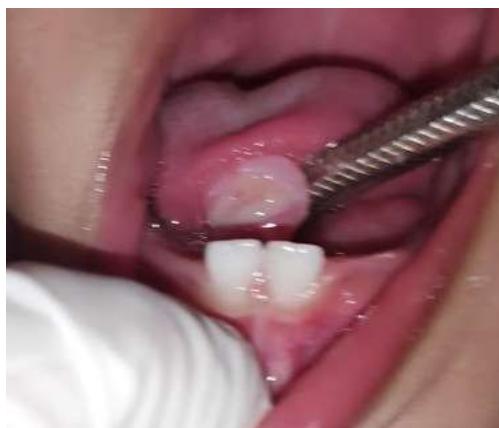


Figure 1: The lesion located at the midline anterior portion of the ventral surface of the tongue.



Figure 2: The lesion was reduced in size and central ulceration was not present after one week.

DISCUSSION

Riga-Fede disease should be considered in infants presenting with oral ulceration due to presence on natal or neonatal teeth. Teeth present at the time birth, are the natal teeth, and the teeth that erupted in the first 30 days of the child's life are neonatal teeth. Exact etiology of natal and neonatal teeth is not known. Some hypotheses are dominant autosomal inheritance; endocrine disturbance resulting from pituitary, thyroid, and gonads; excessive or increased resorption of overlying bone resulting in early teeth eruption; poor maternal health, endocrine disturbances, febrile episodes during pregnancy, and congenital syphilis.⁵ Similar lesions can be seen occurring in older children and adults known as 'oral traumatic granuloma' but the term Riga-Fede is reserved for those presenting before two years of age. Simple chronic traumatic ulcerations are most commonly seen on the tongue, lips, and buccal mucosa (sites usually injured by the dentition). Most reported Riga-Fedes have been on the anterior ventral surface of the tongue but lesions have also been seen involving the tongue's dorsal surface. Riga-fede disease has been seen to have a significant male predominance.⁶ As in this case, the site affected is anterior region on the ventral surface of the tongue in a male infant of 8 months.

A number of medical disorders have been seen associated with riga-fede disease. Riga-Fede persists in infants who have severe cerebral palsy (CP) and who cannot control spasticity of the tongue and therefore suffer repeated ulceration. Besides the possibility of CP, Lesch– Nyhan syndrome (disorder of purine metabolism), familial dysautonomia (disorder in autonomic and peripheral sensory functions), or congenital indifference to pain may be an underlying problem and tongue biting may be the initial presentation.¹

Treatment is focused on the conservative management of the oral lesions. The prime concern is always to

remove the etiology causing trauma. Early diagnosis is very important in this condition to avoid infant's discomfort. A failure to diagnose or treat such lesions properly may induce deformity or mutilation of tongue, dehydration, inadequate nutrients intake by the infant and growth retardation.² Various treatment options include smoothing off the incisor edges, covering the rough incisor edges with composite resin, changing feeding habits by using a bottle with a larger hole in the nipple, placing a nasogastric tube, or relieving symptoms by application of a local corticosteroid. In case when the conservative treatment options do not lead to rapid resolution of this lesion, it may be necessary to extract the lower incisors. If the lesion persists after removal of teeth, an excisional biopsy should be performed.⁷ Recurrence is usually uncommon in this condition. In this particular case we opted for the conservative approach by smoothing the incisal edges and changing the feeding pattern and habits of the infants. The lesion regressed within a week eliminating pain and discomfort. As the mother didn't give consent for the surgical excision of the lesion, the patient was kept under observation and follow up.

CONCLUSION

Early detection of Riga-Fede disease is recommended to diagnose and treat these severe diseases, but also for

otherwise healthy infants. A failure to diagnose or treat such lesions properly may induce deformity or mutilation of tongue, dehydration, inadequate nutrients intake by the infant and growth retardation.

REFERENCES

1. Baghdadi ZD. Riga-Fede disease: association with microcephaly. *Int J of Paediatr Dent* 2002;12:442-5.
2. M. Costacurta, P. Maturo, R. Docimo. Riga-Fede Disease And Neonatal Teeth. *Oral & Implantology - Anno V - N. 1/2012*.
3. Slayton RL. Treatment alternatives for sublingual traumatic ulceration (Riga-Fede disease). *Pediatr Dent* 2000; **22**: 413-414.
4. Volpato LER, Simões CAD, Simões F, Nespolo PA, Borges AH. Riga-Fede Disease Associated with Natal Teeth: Two Different Approaches in the Same Case. Case reports in dentistry. Vol. 2015, Article ID 234961, 4 pages.
5. S. Mhaske, M. B. Yuwanati, A. Mhaske, R. Ragavendra, K. Kamath, and S. Saawarn. Natal and neonatal teeth: an overview of the literature, *ISRN Pediatrics*, vol. 2013, Article ID 956269, 11 pages, 2013.
6. Taghi A, Motamed M HK. Riga-Fede disease: A histological study and case report. *Indian J Dent Res* 2009;20:227-9.
7. Meij EH, Tjalling W, Eggink HF, Visscher J. Traumatic lingual ulceration in a newborn: Riga-Fede disease. *Italian Journal of Pediatrics* 2012, 38:20