

# Ankyloglossia Resulting in Riga-Fede Disease: A Case Report

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## PURPOSE

To discuss a unique case of posterior tongue-tie in an infant that resulted in ulceration of the ventral side of the tongue, also known as Riga-Fede disease, and how this may guide management strategies in cases of Riga-Fede disease secondary to different etiologies.

## BACKGROUND

- Ankyloglossia, or “tongue-tie”, is a congenital abnormality of the lingual frenulum, the soft tissue composed of the oral mucosa of the tongue and the floor of mouth fascia that resides below it.<sup>1</sup>
- Tongue-ties may interfere with the range of motion of a newborn’s tongue, which may impact their ability to latch and thus feed, and/or their speech.
- A frenotomy procedure involves cutting the abnormal frenulum to increase mobility of the tongue.<sup>1</sup>
- Some positive outcomes following frenotomies include reducing maternal nipple pain during feeds<sup>2</sup>, improved latching and feeding, and safety of the procedure.
- Other studies have noted concerns with frenotomies including misdiagnosis<sup>11</sup> and complications of the procedure.<sup>2</sup>
- There are currently no single validated tools used universally to characterize ankyloglossia and determine indication for frenotomy.
- Riga-Fede disease is a mucosal disorder that involves repetitive motions of the tongue over the mandibular anterior incisors, resulting in traumatically-induced ulceration of the ventral tongue and/or floor of the mouth.<sup>3</sup>
- A description of Riga-Fede disease in the setting of co-occurring ankyloglossia has not yet been identified in the existing pool of literature.
- This is the first case of a severe ankyloglossia posterior to the sublingual glands resulting in Riga-Fede disease and presents a definitive condition requiring treatment.

## CASE PRESENTATION

- A 6-month F presented with coughing that interrupted breastfeeding, resulting in a shortened duration of feeding periods. The child developed an ulceration of the ventral surface of her tongue.
- A pediatric dentist rounded the incisinal edges of the mandibular incisors. However, the interrupted breastfeeds and cough during breast feeds continued.
- On evaluation by a second pediatric dentist, the patient’s tongue had adequate protrusion but little elevation posteriorly to the sublingual glands on manipulation.
- When the tongue was elevated, a cord-type frenulum was firmly attached to the floor of the mouth. The ventral surface ulceration was covered by firm white fibrous tissue, and tender on palpation.
- A diagnosis of Riga-Fede secondary to ankyloglossia was made due to incisor ulcerative trauma from lingual protrusion exacerbated by insufficient tongue elevation on feeding.



Image 1: Patient with concurrent ankyloglossia and Riga-Fede disease

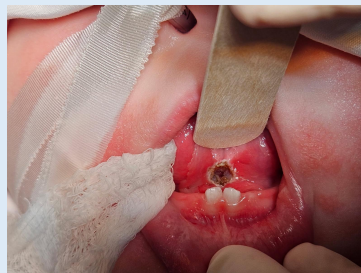


Image 2: Taken during ankyloglossia surgery following transection and dissection of the frenum and removal of fibrous ventral tongue tissue



Image 3: Suture approximating wound during ankyloglossia surgery

## CASE OUTCOME

- The tongue was elevated with a suture through the tongue tip. The sublingual glands were reflected and horizontal incision with bovie was made posterior to the sublingual glands in the midportion of the frenulum along its entire length. The submucosal fibrous connective tissues were transected and released with blunt dissection from genioglossis and mylohyoid muscles. The frenulum release allowed the tongue to reach the palate.
- On the 48-hour postoperative call to the patient’s mother, the child was able to breastfeed without interruption or coughing.
- On the 3-month postoperative visit, the mother stated the child began eating baby foods shortly after the 48-hour postoperative call and continued to thrive.
- On examination, at 3 months, the frenum area and lingual tongue mucosa had healed without complication.

## DISCUSSION

- In cases without concurrent ankyloglossia, studies have found that the most effective methods of treatment of Riga-Fede disease include extraction<sup>4</sup> and ameloplasty.
- This case is unique in that the condition was resolved without intervention on the responsible incisors, but rather the tight lingual frenulum that was exacerbating the Riga-Fede disease.
- Thus, in cases where Riga-Fede may be secondary to another cause such as ankyloglossia, it may be reasonable to consider first resolving the primary condition rather than taking immediate action to alter or extract natal teeth.

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