

DOUBLE TROUBLE - A RARE CASE OF RIGAFEDE DISEASE WITH LUDWIGS ANGINA

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INTRODUCTION:

Riga-Fede disease (RFD) is a benign lingual ulceration caused by repetitive trauma. It is also known as traumatic eosinophilic granuloma. It was described initially by Riga in 1881 and by Fede in 1890. It is usually caused by the sharp edges of newly erupted teeth by repeated forward and backward movements of the tongue over the lower incisors. Its most common site is on the tongue. But lip, palate, gingiva, vestibular mucosa and floor of the mouth may also get involved. It usually presents in early infancy in association with natal or neonatal teeth or eruption of lower incisors. The painful symptoms may be absent or acute, up to the point of preventing the baby from feeding.

Eruption of first primary tooth starts on an average at around six to seven months of age.¹ Presence of teeth at birth or within a month after birth is considered rare. These teeth are called natal teeth when they are present at birth and neonatal teeth when they erupted during the neonatal period (first 30 days of life).

CASE REPORT:

A 12-day-old baby girl came with complaints of swelling over the left side of face and bleeding from the floor of the mouth for past one week which was associated with difficulty in feeding and fever. The child also had the history of natal teeth present since birth, which was accidentally removed partially by the baby's relatives. On examination an ulcer was present over the ventral-surface of the tongue extending from middle third to the lower alveolar surface with remnant natal tooth and also hard indurated non-fluctuant swelling present over the left side of lower face with warmth,

tenderness and erythema involving submandibular region extending from angle of mandible to chin. Fortunately, the baby had no breathing difficulties. The diagnosis of RIGAFEDE DISEASE and LUDWIGS ANGINA was made based on the above clinical findings, following which immediate floor of the mouth debridement with removal of remnant natal teeth followed by incision and drainage of the abscess was done. During debridement, there was also a 0.5 × 0.5 cm. Sinus tract like ulcer noted in the lower lip at LABIO GINGIVAL SULCUS. Debrided tissue sent for HISTOPATHOLOGY examination and fungal smear and culture.



Fig A



Fig B

Ulcer at floor of mouth with remnant Tooth (Fig A and Fig B)



Fig C

LUDWIGS ANGINA



Fig D

Floor of the mouth debridement
With removal of remnant tooth



Fig E

Incision and Drainage

DISCUSSION:

RIGAFEDE'S DISEASE is a rare benign mucosal disease, defined as persistent ulceration of Oral mucosa as a result of repetitive trauma of antero ventral surface of the tongue by the sharp primary tooth due to frictional forward and backward movement of the tongue. This condition has been documented under various articles as a rare benign condition and not life threatening. But in our case, it leads to a potentially life-threatening condition LUDWIG'S ANGINA. LUDWIG'S ANGINA is life-threatening cellulitis of the soft tissue involving the floor of the mouth and neck. It involves three compartments of the floor of the mouth, the sublingual, submental, and submandibular. The infection is rapidly progressive, leading to potential airway obstruction. The most common etiology is a dental infection in the lower molars, mainly second and third, accounting for over 90% of cases. Any recent infection or injury to the area may predispose the patient to develop Ludwig's angina. Predisposing factors include diabetes, oral malignancy, dental caries, alcoholism, malnutrition, and immunocompromised status.

This coexisting condition is a rare event in newborn and any newborn with natal tooth to be followed up for RIGAFEDE DISEASE and its subsequent consequences. In our case infection of the sublingual and submandibular space is believed to be contracted from the site of RIGAFEDE'S DISEASE through lower lip labio gingival sulcus. This was identified intra operatively on exploration.



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RECOMMENDED READINGS:

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