



CASE SERIES: RIGA FEDE DISEASE IN PEDIATRIC OPD.

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KEYWORDS :

INTRODUCTION:

Riga fede disease is a benign sublingual ulceration in the floor of mouth caused usually due to repetitive backward and forward rubbing of the tongue over teeth. It is seen mostly in neonates with natal teeth, it is also seen in older age group infants and toddlers. Although this is a well-known entity among dentists but it still remains an unusual and less common condition presenting to the pediatricians. It was first described by Antonio Riga, an Italian physician in 1881 and was later published by Francesco Fede in 1890 along with the histologic description, hence known as Riga Fede disease. It commonly appears as ulcer on the ventral surface of the tongue (60%) however other areas of lip, palate, gingiva, vestibular mucosa and floor of the mouth can also be affected¹.

Case 1: An 11 months male child was brought to the pediatric opd with complaints of an ulcerated swelling at the base of tongue as noted by mother. The child was otherwise active, playful and well thriving. No history of any bleeding from the lesion. No history of any previous hospitalisations, bleeding diathesis or blood transfusions in the past. On examination child was stable, conscious and well oriented to mother. Anthropometry was normal. Systemic examination was also within normal limits. Complete hemogram and baseline investigations were also normal. The child was referred to department of dentistry for evaluation of the dentition and need of any tooth extraction if any. Topical steroids were prescribed for short duration and child was subsequently kept on regular follow-up. After 6 months the child was healed by itself.

Case 2: A 10 month child presented to the Pediatric OPD with history of not growing well for the last 3 months. There was history of feeding difficulty while breastfeeding and the child was not able to suckle well on breast. On Anthropometry, child was found to be having moderate acute malnutrition. General physical examination revealed an ulcerated lesion on the ventral aspect of tongue measuring 5 mm x 12 mm in size that extended from anterior border of the tongue to lingual frenulum (figure 1,2).



Figure 1,2: showing the sublingual ulceration on ventral surface of the tongue.

The lesion was tender on palpation. Examination of the rest of intraoral mucosa revealed no other lesions except two lower incisor teeth one of which was loosely fixed. Child had been repeatedly rubbing his tongue over these tooth which had subsequently led to this ulcer which was painful and now interfering with the feeding also. Being unaware of this, mother did not notice the lesion and this had subsequently led to feeding difficulty and malnutrition in the child. All other systemic examination and investigations were normal. Based on these clinical findings, the diagnosis of Riga fede disease was made and child was referred to dental surgeon where extraction of tooth was done under local anesthesia as it would have interfered with the healing process. Also the mother was counseled and an oral choline salicylate, glycerine and lignocaine composition gel was prescribed along with some multivitamins and zinc on discharge. On follow-up the lesions resolved gradually over 2 weeks and afterwards the oral intake improved to normal. Infant was subsequently kept under regular follow-up for the nutrition and growth monitoring which improved drastically over 1 year of follow-up.

Case 3: A 20 months male child was brought to the outdoor patient department by his parents for routine checkup as the child was suffering from mental retardation. Their chief complaint was that the child had decreased feeding for the last one month. The child was having frequent tongue movements and on oral examination a sublingual ulcer was found which was 10 x 15 mm size on the ventral aspect of tongue. On palpation, the lesion was painful to touch. There was no other lesions in the oropharyngeal region. Also the child was having moderate pallor. No other unusual findings were noted on further clinical examination. On investigations his hb was 8.9g/dl with microcytic hypochromic picture on the peripheral smear. Iron studies were suggestive of iron deficiency anemia. Rest of the laboratory reports were normal. Child was clinically diagnosed as sublingual ulceration (Riga fede disease) with iron deficiency anemia with mental retardation. He was started on iron, folic acid and vitamin B12 supplementation and was consulted with the dentist. Dental consultation was sought for the sublingual ulcer which revealed that the child had very sharp edged incisors. Smoothing of the edges was done and the child was prescribed some topical antibiotic and analgesic combination gel. The ulcer took 2 weeks to show some healing and eventually healed over next 4 weeks on follow up. After 6 weeks of follow-up the child was feeding well and improved.

DISCUSSION:

Riga Fede Disease is a reactive traumatic mucosal disease characterized by persistent ulcerations of the oral mucosa¹. It develops as a result of repetitive trauma of the tongue usually by the newly erupted mandibular incisor teeth during continuous protrusive and retrusive movements. Narang et al

described the constant traction due to tongue-tie as another cause in addition to the trauma caused by the teeth². Although most cases are known to occur at an average of 6 to 8 months but all three of our cases were either in late infancy or beyond infantile age.

With regard to RFD treatment, several management options have been reported. They may be used alone or in combination³:

- a. cellulose film or other protective dental appliance;
- b. oral disinfectant;
- c. corticosteroids;
- d. teething ring;
- e. smoothing the incisal edges;
- f. small increment of restorative material to the incisal edges;
- g. Dental extraction.

The first option for RFD treatment should always be conservative, avoiding natal and neonatal teeth extractions when possible⁴. Although the removal of the traumatic agent is almost always required. If conservative treatment options do not lead to a quick resolution of the injury, the tooth extraction may be needed³. Tooth extraction is also indicated in case of excessive tooth mobility. Spontaneous recovery is also known to occur.

Histological analysis should be done when there are no signs of improvement even after two weeks following initiation of treatment. Histology demonstrates a mixed inflammatory reaction under the ulcerated surface, including T cells, large mononuclear cells and numerous eosinophils⁵. RFD may be considered as a benign condition in healthy infants, but it may be associated with many other serious neurologic disorders especially cerebral palsy⁵.

Recommendations:

High suspicion and early detection of RFD is recommended to diagnose and treat these cases as, 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 as is evident in our case 2 and case 3 presenting with moderate acute malnutrition and anemia respectively. As the ulcer can be very painful, Riga-Fede disease may affect feeding and therefore dehydration, malnutrition and failure-to-thrive can result if unrecognised and untreated.

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