

BIGYAN RAJ GYAWALI

Ganesh Man Singh Memorial Academy of
ENT-Head and Neck Studies
Tribhuvan University Teaching Hospital
Institute of Medicine (IOM), Maharajgunj
Kathmandu, Nepal

Corresponding Author

Bigyan Raj Gyawali

Ganesh Man Singh Memorial Academy of
ENT-Head and Neck Studies,
Tribhuvan University Teaching Hospital,
Institute of Medicine (IOM), Maharajgunj,
Kathmandu, Nepal

Email: bigyan.gyawali@gmail.com

A RARE PRESENTATION OF RIGA FEDE DISEASE IN AN ADULT : CASE REPORT

ABSTRACT

Riga Fede disease usually presents as an oral mucosal ulcer commonly in paediatric population, often initiated by repeated dental trauma. Here, we discuss its rare presentation in a 32 year old adult lady who presented with an oral ulcer of 2 months duration.

Keywords: Adult, Riga Fede disease

INTRODUCTION

Riga-Fede disease (RFD) is one of the uncommon presentations of oral lesion as a mucosal ulcer. Though benign in nature, inability to diagnose it may lead to overzealous treatment such as excision as it mimics malignant ulcer of tongue. The proposed etiology is repeated trauma over the tongue caused by sharp teeth.¹

This disease was named after Italian physician Antonio Riga and Pediatrician Francesco Fede. It presents at several subsites of oral cavity mostly in the tongue but also in the lips, gingiva and palate.² Being mostly defined amongst the paediatric population, not much has been mentioned in literatures regarding its incidence in adult population. Here we present a case report of Riga-Fede disease presenting as a tongue ulcer in an adult female.

CASE REPORT

A 32 year old lady presented with painful ulcer in the tongue for 2 months duration. It was noticed as an erythema initially which gradually progressed to a painful ulcer over 2 months.

It was associated with odynophagia, however there was no history of bleeding from the ulcer, dysarthria, aspiration or any swellings in the neck. She didn't have any history of alcohol consumption, smoking although she was occasional consumer of betel nuts for past 7-8 years. No significant past medical and treatment history could be elicited.

Examination revealed approximately 1 x 2 x 0.5 cm oval ulcer at the right lateral border of the anterior two-third of the tongue (Fig.I). Ulcer had irregular margin covered with whitish slough.

The edge was sloping and the floor was covered with slough. Palpation revealed indurated margin with tenderness and base was firm in consistency. Oropharyngeal examination showed dental caries over lower right second molar with sharp



Fig I: Ulcer over right lateral border of tongue with caries of 2nd lower molar teeth

teeth edge (Fig.I). Mobility of the tongue was not restricted. There were no palpable lymph nodes or any obvious mass in the neck. Ear and nose examination showed no abnormalities.

Carcinoma tongue was taken as the first differential diagnosis considering the history of consuming betel nut, duration and the nature of ulcer. Biopsy was taken from margin of the ulcer and sent for histopathological examination which revealed hyperplastic stratified squamous epithelium with elongation of rete ridges and parakeratosis. Granulation tissue with vascular stroma and inflammatory infiltrates were seen. There was dense stromal infiltration of lymphocytes and predominance of eosinophils (Fig.II). All of these features were compatible with Riga-Fede disease.

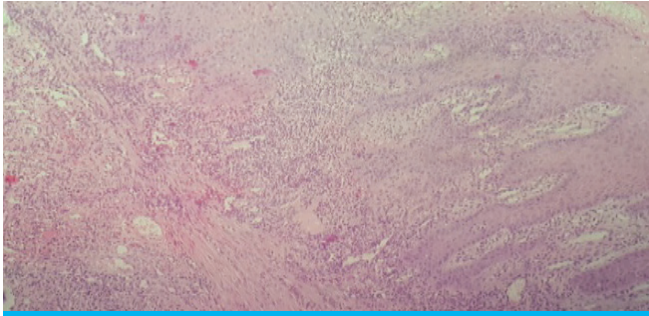


Fig II: Histological slide showing hyperplastic epithelium, parakeratosis and dense eosinophilic infiltration.

She was managed conservatively with topical steroids for two weeks following which the ulcer got completely healed. Palpation revealed no induration on follow up. However, considering the ulcer was due to repeated trauma caused by the sharp teeth, she was advised for the extraction of second lower right molar.

DISCUSSION

Riga-Fede Disease is a benign mucosal lesion presenting as an ulcer in the oral cavity usually caused by repetitive trauma with sharp teeth. It is usually prevalent in the pediatric population, commonly affecting the tip of the tongue.³ Less has been mentioned in the literatures regarding the prevalence of the disease in adult population. Wollina et al. in their case report described a 70 year old woman who had a similar presentation of tongue ulcer due to an ill fitting denture.⁴ It was later diagnosed as Riga Fede like lesion, which got completely healed after she was advised not to use the ill fitting denture for 2 weeks.

Common clinical presentation include oral mucosal ulcer which is usually tender and indurated on palpation. There is always an initiating factor such as a dental caries of right lower second molar in our case. Costacurta in their case report had two crowns of lower incisor neonatal teeth which induced the tongue ulcer in a 2 months old female infant.⁵ Similarly, Dundar et al. in their study showed denture related lesions in the oral cavity accounted for upto 36.4% of cases.⁶

Diagnosis mainly relies on clinical suspicion and confirmation is done by histopathological examination. Histologic findings show an inflammatory infiltrate consisting of eosinophils with lymphocytes, macrophages, plasma cells and mast cells.⁷ In our case, our first line of investigation was biopsy of the lesion and its

histopathological examination. It revealed features like hyperplastic stratified squamous epithelium, parakeratosis and inflammatory infiltrates, of which eosinophils were the most common.

Management of the disease is conservative in majority of the cases. However, in rare instances inability to diagnose the disease may lead to an aggressive approach such as excision of the lesion considering the possibility of malignancy. Various modalities of management options have been put forth such as cellulose film, topical steroids, smoothing of the sharp teeth edges, dental extraction.⁵ We advised the patient with topical steroids for 2 weeks, after which the lesion disappeared. To prevent the further recurrence we referred our case to dental OPD for the extraction of lower right molar tooth which was suspected of causing the lesion.

CONCLUSION

Riga Fede disease is a benign mucosal lesion of oral cavity presenting as ulcer. Though common in pediatric population, it should be kept as one of the differential diagnoses in adult cases who present with an oral ulcer. Presence of abnormally aligned tooth or tooth with sharp edges usually strengthens the diagnosis. Confirmation of diagnosis is by histological examination. Management is conservative with topical steroids and correction of the initiating factor.

REFERENCES

1. Baroni A, Capristo C, Rossiello L, Faccenda F, Satriano RA. Lingual traumatic ulceration (Riga-Fede disease). *Inter J Dermatol*. 2006;45(9):1096-7.
2. Joseph BK, Bairava Sundaram D. Oral traumatic granuloma: report of a case and review of literature. *Dental Traumatology*. 2010;26(1):94-7.
3. Ceyhan AM, Yildirim M, Basak PY, Akkaya VB, Ayata A. Traumatic lingual ulcer in a child: Riga-Fede disease. *Clin Exp Dermatol*. 2009;34(2):186-8.
4. Wollina U. Riga-Fede-Like disease in a 70 year old woman. *Indian J Dermatol*. 2010;55:92-4.
5. Costacurta M, Maturo P, Docimo R. Riga-Fede disease and neonatal teeth. *Oral Implantol (Rome)*. 2012;5(1):26-30.
6. Dundar N, Ilhan Kal B. Oral mucosal conditions and risk factors among elderly in a Turkish school of dentistry. *Gerontology*. 2007;53:165-72.
7. Baroni A, Capristo C, Rossiello L, Faccenda F, Satriano RA. Lingual traumatic ulceration (Riga-Fede disease). *Inter J Dermatol*. 2006;45(9):1096-7.