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Research Article

TONGUE ULCER ASSOCIATED WITH NATAL TEETH IN AN INFANT- RIGA-FEDES DISEASE

Sumalatha, M.N*, Gadiputi Sreedhar., Pallav Singhal and Anu Garg

Department of Oral Medicine and Radiology Babu Banarasi Das College of Dental Sciences,
BBD University, BBD City, Sector-1, Dr. Akhilesh Das Nagar Faizabad Road, Chihat
Lucknow, Uttar Pradesh India

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ABSTRACT

Traumatic ulceration of the oral mucosa may result from physical damage via sharp teeth, food stuffs, accidental biting or talking. Ulceration of ventral surface of tongue is due to trauma (Riga-Fedes disease) is most often associated with natal or neonatal teeth. Presence of teeth at birth or within a month post-delivery is rare with varied occurrence rate from 1:6000 to 1:800 cases and cause ulceration which may remain for a long time, resulting in inadequate food intake, retardation of growth and sometimes result in dehydration, feeding difficulties, failure to thrive in an infant and ulceration may progress to large fibrous mass with repeated trauma. A Case of tongue ulceration in a ONE month old Muslim male baby has been presented with family history of natal and neonatal teeth.

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INTRODUCTION

Riga-Fedes disease is an oral condition found rarely in newborns that manifest as an ulceration on the lingual frenum, ventral surface of the tongue or on the inner surface of the lower lip due to trauma from natal or neonatal teeth¹. The term Riga fedes disease was named after its first description clinically by the Italian physician **Antonio Riga** in 1881 and histologically by **Francesco Fede** an Italian pediatrician in 1890². Synonymously this entity can be called as lingual traumatic ulceration, sublingual growth in infants, traumatic atrophic Glossitis, and sublingual fibrogranuloma³.

Riga-Fedes disease is a complication of natal or neonatal teeth in 6-10 % of cases⁴. The presence of natal or neonatal teeth in newborn is uncommon, varying from 1:6000 to 1:800 cases occurring in general with incidences of two or three teeth⁵. The self-mutilating behavior is quite common in children with lower intelligence, familial dysautonomia (Congenital insensitivity to pain), CIPA (congenital insensitivity to pain with anhidrosis), and in children with a habit of repetitive tongue thrusting are the main cause of riga-fedes disease with natal and natal teeth⁶.

The most common location for ulceration in riga-fedes disease is anterior tip of the tongue⁷ with erythema surrounding the centrally removable, yellow, fibrinopurulent membrane with

rolled white hyperkeratotic border immediately adjacent to the ulceration⁸. The healing process in riga-fedes disease takes longer duration leading to complications faced by mother and the child⁹.

Case-Report

A healthy **One-month-old** boy from **Muslim** community was brought by his parents to the dental OPD with complaint of ulcer on the ventral surface of tip of the tongue since two weeks (**Figure-1**).



Figure-1 Ulcer in the Ventral Surface of Tip of Tongue

The child was experiencing difficulty in feeding associated with pain, sleep disturbance and continuous crying with cranky nature. Patient's mother had noticed white projections in her baby's mouth after birth while breastfeeding and also gave history of similar kind of ulceration associated with teeth on the

*Corresponding author: **Sumalatha, M.N**

Department of Oral Medicine and Radiology Babu Banarasi Das College of Dental Sciences, BBD University, BBD City, Sector-1, Dr. Akhilesh Das Nagar Faizabad Road, Chihat Lucknow, Uttar Pradesh India

ventral surface of the tongue in patient elder siblings. On examination there was a close association of the ulcer with two teeth in the lower incisor region, they were white in color, with grade II mobility and tilted horizontally towards tongue from their position (**Figure-2**).



Figure-2 Position of Teeth In Relation To Tongue

Ulcer was measuring approximately 1x1.5cms with irregular Erythematous borders covered by pseudo membranous slough (**Figure-3**) and pain upon palpation. Otherwise the rest of the oral mucosa was healthy.



Figure-3 Ulcer With Erythematous Borders And Yellow Slough

The patient did not cooperated to take an intraoral radiograph. Based on clinical findings and family history the diagnosis of **traumatic ulcer associated with natal teeth -Riga-Fedes disease** was made. Since immediate extraction was the treatment of choice, with parent's co-operation and their consent vitamin-K (0.5=1.0mg) was administered intramuscularly as a part of immediate medical care to prevent hemorrhage and the teeth were extracted under topical anesthesia. The patient was reviewed after 15 days and the lesion was fully resolved with normal feeding and sleeping habits of the baby. The child was followed up further for 6 months to check for eruption of deciduous incisors (**Figure-4 & 5**).



Figure 4 & 5 Erupted Primary Teeth After Six Months Recall Visit.

DISCUSSION

Medical and dental literature from past two hundred years reported Infants born with teeth or teeth erupting immediately after birth and referred them as natal teeth, congenital teeth, and precocious dentition¹. In modern literature **MASSLER** and **SAVARA** satisfactorily defined them as natal and neonatal teeth the former present in the oral cavity at birth and later erupt from birth to 13th day of life with hypoplastic enamel, underdeveloped roots, and mobility⁷.

The occurrence of natal and neonatal teeth varied with prevalence rate ranging from 1:700 to 1:30,000 with other reports around 1 in 2000-3500 live births and the incidence rate from 1 in 11, 100, 2000, 6000, 10,000 live births from different studies¹⁰.

With conflicting findings of their occurrence, females in generally affected more as compared to males and Muslims children's exhibited more natal/neonatal teeth as compared to Hindu children. Natal teeth are more frequent, approximately three times more common than neonatal teeth, with most common location being the Mandibular incisor region (85%)¹⁰. The findings from our case were correlating with above findings except for sex the present finding was in male as supported by **Neville B.W (2002)**¹¹.

The debatable etiology of natal and neonatal teeth put forward with various hypotheses like hereditary transmission of a dominant autosomal gene, endocrine disturbances, infections, nutritional deficiency, febrile status, superficial position of tooth germ, environmental factors. The most acceptable theory is based upon the result of a superficial localization of the dental follicles, related to hereditary factors¹⁰. According to **Bodenhoff** and **Gorlin** 15% children with natal and neonatal teeth had parents, siblings or close relatives with a history of having similar condition¹². 90-99% teeth are precociously erupted from normal complement of primary teeth only 1-10% is supernumerary¹². A positive family history for natal teeth in this case, lends support to the suggestion that hereditary influences are involved in the etiology of natal teeth and supernumerary origin because the patient sibling also had the same problem and the primary teeth was erupted later 6 months after extraction.

Clinically, these teeth are small, conical or of normal size, shape with immature appearance, enamel hypoplasia, and small root. They may exhibit brownish yellow/ whitish opaque color attached to the pad of soft tissue above the alveolar ridge, occasionally covered by mucosa and as a result have an exaggerated mobility, the risk of being swallowed or

aspirated¹⁰. In the present case teeth were small, hypoplastic brownish yellow in appearance.

Trauma to the tip of the ventral surface is a complication of natal or neonatal teeth as a sequence of the fact that the tongue in infants lies between the alveolar ridges and it usually develops when the tongue is rubbed repeatedly over the teeth, usually lower front teeth as the newborn baby pushes the tongue forward when feeding (infantile tongue-thrusting reflex) it can also develop in older babies when their lower front teeth erupt at the normal time. The usual age range is 1 week to 1 year, with peaks in early infancy and 6-8 months of age^{7, 8, 13}.

The diagnosis of these teeth is done based on history, physical examination, and by clinical and radiographic findings of the infant to rule out them being part of normal dentition or supernumerary so that indiscriminate extractions would be performed. According to many reviews most of these teeth are primary dentition and not supernumerary but the diagnosis is important to plan treatment, keeping the view in maintaining normal dental occlusion¹⁴. Extraction was carried out without radiograph but in recall visit patient had erupted primary central incisors, so we confirmed the teeth were of supernumerary origin.

Treatment of Riga-Fedes disease has varied over the years focusing on eliminating the source of trauma with early treatment of excising the lesion and smoothening of incisal edges. If extraction is carried out, it is necessary to ensure that the underlying dental papilla and Hertwig's epithelial root sheath are removed by gentle curettage as root development can continue if these structures are left in situ¹⁰. These teeth with hypomineralization and limited surface area for resin bonding leads failure of restoration and chances of swallowing the resin present the practical problem considering the age of the children¹⁵. Rapid healing after biopsy is typical in large traumatic ulcers with rare recurrence rate⁸. In the present case ulcer healed within fifteen days which was in contrast to findings of Terzioglu Ahmet⁹.

CONCLUSION

To conclude infants with prematurely erupted teeth must be carefully examined for further treatment planning, and parent counseling to bring about awareness.

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