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Riga-Fede Disease or Neonatal Sublingual Traumatic Ulceration

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Short Communication

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Abstract

In this article, "Riga-Fede disease" or syndrome which is also called as "Neonatal sublingual traumatic ulceration" is briefly described. It is a traumatic ulcerative lesion caused by presence of teeth at birth or later in life along with protrusive and intrusive movement of the tongue. Knowledge about this condition is highly essential among all clinicians to provide appropriate treatment for the tender and tiny infants developed with this condition.

Keywords: Neonate; Natal Tooth; Neonatal Tooth; Riga-Fede Disease; Traumatic Ulceration

Introduction

Intraoral lesions occurring in neonates brings a panic condition among parents as it is associated with difficulty in breast feeding and insufficient sucking by the neonate. Riga-Fede disease or syndrome is one of that kind reported in maximum incidence in neonates. It is a reactive traumatic mucosal disease characterized by persistent ulceration of the oral mucosa mainly involving ventral surface of the tongue (60%) [1]. It is caused mainly by presence of natal or neonatal teeth which are present at the time of birth or within one month of new born baby causing repetitive trauma to the tongue during forward and backward movement of the tongue or during sucking [2]. Riga-Fede disease is referred by various synonyms like traumatic granuloma of the tongue, neonatal lingual traumatic ulceration, reparative lesion of the tongue, sublingual ulcer, sublingual fibro-granuloma and eosinophilic ulcer of the oral mucosa [3].

Discussion

The history behind the development of this term is quite interesting. This clinical intra oral condition was first described by Antonio Riga who is an Italian physician in the year 1881. Later Fede, the founder of the Italian paediatrics, published a series of histological studies and additional cases and finally gave its histology and benign nature of this condition in 1890 [4,5]. Thus, a name of Riga-Fede came into existence to this intraoral lesion. This condition is exclusively restricted to the tongue, develops soon after birth and associated with natal or neonatal teeth. In a retrospective review of 29 cases performed by van der Meij et al [3], all lesions of Riga-Fede disease occurred in association with natal or neonatal teeth. According to this study, the baby boy to baby girl ratio ranged from 1.8:1, with age of occurrence ranging from six to twenty-four months, with a mean age of ten months [3]. However, there is no gender predilection described in some reports with both male and female gender getting affected equally. Although most of the Riga-Fede conditions appeared to be asymptomatic but occasionally they seen associated with pain [6].

The management of Riga-Fede disease depends on many factors including tooth's mobility, risk of aspiration or swallowing, whether the tooth is a supernumerary or normal counterpart of primary teeth, whether it is causing

interference in breastfeeding, breast and oral soft tissue injuries and finally the general state of child's health. It is also reported that the ulceration is seen not only on the ventral surface of the tongue, but also on the lip, palate, gingiva, vestibular mucosa, floor of the mouth and the mother's breast [5].

Clinically, this lesion develops as an ulcerated area with prominent raised edges that occurs due to repeated trauma from the erupted neonatal or natal tooth and can transform into an enlarged fibrous mass with appearance of ulcerative granuloma and superficial necrosis. Therefore, it is difficult for the infant to suck and feed, putting the baby at risk of nutritional deficiencies and sometimes make them irritable. Sometimes, the raised edges of this lesion resemble a malignant lesion. Therefore, an awareness and knowledge about Riga-Fede disease is highly essential among all clinicians including dental and medical specialties to avoid unnecessary anxiety and biopsy. Research states that in one fourth of documented cases this disease entity was associated with underlying neuro-developmental disorder like cerebral palsy, familial dysautonomia, Down syndrome, Lesch Nyhan syndrome, encephalopathy and microcephaly. It is also reported in a patient with acquired immune-deficiency syndrome.

Being a benign, a rare clinicopathological entity and uncommon mucosal disorder, Riga-Fede disease should be differentiated by the traumatic ulcerative granuloma with stromal eosinophilia (TUGSE) [6]. The term TUGSE was first framed by Elzay in 1983 [7] refers to a chronic but self-limiting reactive ulcer of the oral mucosa. Histologically the excised specimen shows mainly an inflammatory infiltration consisting of numerous eosinophils with lymphocytes, macrophages, plasma cells and mast cells. There is a classification system given by Dominguez-Cruz, et al. [8] on Riga-Fede disease and they have classified into two types as shown in Table 1.

Type	Classification Name	Description
Type 1	Precocious Riga-Fede Disease	Associated with natal-neonatal teeth
		Appears in the first 6 months of the life
		Has no correlation with neurological disorders
Type 2	Late Riga-Fede Disease	Typically appears after 6-8 months of life
		Associated with the eruption of the first dentition
		May be related to neurological disorders

Table 1: Dominguez-Cruz et al classification on Riga-Fede disease.

Most of the time this rare entity occurs as unifocal, however, multi-focal lesions and recurrences have been shown in the literature. Early detection and diagnosis of this lesion is highly recommended to provide appropriate treatment which includes many factors. Failure to diagnose or treat these conditions inadequately may cause mutilation or deformity of the tongue, inadequate nutrients intake by the infant, dehydration and finally leading growth retardation and failure to thrive [6-9].

In patient with cleft lip and palate the occurrence of natal/neonatal teeth have been shown. As these neonates require preorthopedic devices in order to mold the soft tissue cartilages, a novel treatment approach such as Presurgical Naso-alveolar Molding (PSNAM) therapy prior to the definite primary or secondary surgical therapy is suggested. A recent bibliographic analysis about Indian contribution towards application of PSNAM in cleft lip/palate infants on global level was performed by Nagaveni et al and showed that there is an increasing demand and trend in the utilization of this revolutionary treatment modality not only in India but also in many parts of the world [10].

Moreover, this treatment approach should be carried out immediately following birth of the baby and within 4-5 months of the neonate's period. Because at this age, the levels of hyaluronic acid in circulating blood of the baby which is transferred from mother's womb is more and it is investigated that this chemical is highly essential for the easy molding of soft tissues and to obtain better post-operative results in cleft lip/palate infants following surgical therapy.

Therefore, in such cases if the persistent traumatic lingual ulceration is seen due to continuous movement of the ventral surface of the tongue against these natal or neonatal teeth is evident, this will interfere with the impression taking as well as insertion of the appliance inside the oral cavity of the neonate. It is been surveyed that an awareness about the application of presurgical naso-alveolar molding among clinicians, parents and nurses is totally lacking in many parts of the world [11,12]. This scenario highly indicates a greater number of research to be undertaken including awareness oriented epidemiological surveys among all health care professionals and other personnel who take care of cleft lip/palate infants.

Conclusion

Presence of Riga-Fede disease or Neonatal sublingual traumatic ulceration in neonates or infants brings anxiety and panic in their parents or guardians because of neonate refusing sucking. Therefore, creating awareness regarding this and gaining knowledge about diagnosis and treatment of this clinical entity is highly warranted in the arena of paediatrics.

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