

Residual neonatal tooth in a pre-term infant: A case report and brief review

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Abstract

Sometimes aberrations may occur in the biological chronology, resulting in the presence of dentition at time of birth or soon after birth. Massler & Savara introduced a new nomenclature, where the teeth present at birth or soon after the birth were classified as natal or neonatal teeth respectively. The incidence of natal/neonatal teeth ranges from 1:2000 to 1:3500 live births, and the possibility of natal or neonatal teeth in preterm infants is extremely rare with only few case reports mentioned in literature. Most of the natal teeth are prematurely erupted primary teeth and should be maintained cautiously unless they are interfering with nursing or causing ulceration of the tongue. Frequently such cases do not encounter residual tooth formation subsequent to the extraction of natal and neonatal teeth but still there is an involvement of 9.1% of the risk and therefore the parents should be informed about the possibility and the need for regular follow up.

Keywords: Natal tooth, Neonatal tooth, Preterm infant, Low-Birth Weight, Residual tooth.

Introduction

Predominantly, the first tooth to appear in oral cavity is mandibular central incisor, that erupts at around six months of age.¹ However sometimes, aberrations may occur in the biological chronology, resulting in the presence of dentition at time of birth or soon after birth.² Massler & Savara introduced a new nomenclature for these teeth surfacing before the normal time^{1,3,4} where the teeth present at birth or soon after the birth were classified as natal or neonatal teeth respectively.^{2,3,5} Some authors have also used the terms like congenital teeth, fetal teeth, predeciduous dentition or dentitia praecox for such teeth in the dental literature.^{1,3,4,6,7} They routinely occur in the same position as that of the deciduous dentition and more commonly in the incisor and molar regions of the mandibular arch.^{7,8} The mandibular central incisors are the most frequently observed natal and neonatal teeth.^{3,4,9,10} The incidence of natal/neonatal teeth varies and ranges from 1:2000 to 1:3500 live births.¹¹ with a slight predilection for females being reported in several studies.^{1,9,11-14} Natal tooth has a higher prevalence,^{3,5,8,10,11} with a reported probability of occurrence thrice when compared to neonatal teeth.^{2,3,5,7}

The existence of the natal and neonatal teeth is surely a disruption of eruption pattern^{1,15} occurring in response to potential risk factors such as infection or malnutrition, febrile states, hormonal stimulation, heredity, hypovitaminosis, osteoblastic activity or trauma.^{1,9,12,16} The current concept of the etiology is based on the fact that superficial position of the developing tooth germ could have predisposed the tooth to early eruption resulting in this dental anomaly.^{17,18} Though the exact etiology is not clear but approximately 15% of cases reported with a history of natal or neonatal tooth in parents, siblings and other near relatives that followed an autosomal inheritance pattern.⁸

Natal and neonatal teeth are also seen in patients with cleft lip and palate and occur in 2% of cases with unilateral cleft lip and palate and in 10% of cases with bilateral cleft lip

and palate. Most of the times these teeth have no connection with any medical illnesses, however occasionally they are reportedly found to occur in certain syndromes such as Hallermann-Streiff, Ellis, Ellis Van Creveld, Craniofacial dystosis, Sotos syndrome.^{1,9,15,19,20}

Natal/neonatal tooth may appear like a normal primary tooth but in most of the cases the crown is small and conical in shape with poorly developed roots which are secured to the gum pads scarcely by a flap of soft tissue over the alveolar ridge.^{1,14} In addition they may be associated with hypoplasia of the enamel and dentin and appear yellow-brownish in colour.²¹

Most of the natal teeth are prematurely erupted primary teeth and should be maintained cautiously unless they are interfering with nursing or causing ulceration of the tongue leading to some serious complications.^{21,22} Many authors recommend their extraction while the newborn is in hospital itself as there are higher possibilities of either aspiration of the tooth by the infant when it exfoliates or imminent danger of abscess formation following the loss of attachment.^{4,13} The extraction should be performed with the appropriate considerations as the coagulation mechanism in infants is not fully developed.^{21,22} Infrequently, following the spontaneous loss or extraction of natal or neonatal teeth, the root still continues to grow in socket which may demand further treatment.⁴

The possibility of natal or neonatal teeth in preterm infants is extremely rare with only few case reports mentioned in the literature.^{23,24} Most of the studies have demonstrated conspicuous delayed eruption of primary and permanent teeth in low-birth weight children, which has been corroborated by different authors.²⁴ Based on the findings of certain cross sectional surveys and prospective and longitudinal studies, the correlation, between preterm infants and late eruption of teeth has been strongly substantiated with factors like gestational age, low birth weight and neonatal illnesses /nutritional deficiencies found to be significantly

responsible for the late timing of tooth emergence.²⁵⁻²⁸ Moreover few authors have also observed that the first primary tooth usually erupts at the normal chronologic age in some healthy preterm infants, but the complications related to the prematurity that require prolonged mechanical ventilation for neonatal illnesses results in inadequate nutrition which eventually leads to the delayed eruption of teeth.^{29,30} Despite this apparent association and interrelationship, natal or neonatal teeth have been reported in children born prematurely or having low birth weight but none of the reported case reports in literature have discussed a neonatal tooth in a preterm infant of 30 weeks gestation.

This case report is unique and of significant clinical importance analyzed from the perspective of the early appearance of teeth in contrast to the delayed eruption observed in a preterm infant and the complication experienced due to the continued growth of dental remnants after the extraction of the neonatal tooth. This article also highlights the implications of the interdisciplinary proficiency of the neonatologist and pediatric dentist required for the dental management of a premature infant or low birth weight newborn, along with appropriate emphasis on the hematological limitations.

Case Report

A male subject was born 30 weeks with birth weight of 1600gms. The infant was kept in NICU for about 6 weeks for management of respiratory distress syndrome. (Fig. 1) Post birth, 6 weeks later the mother of the child reported to the concerned pediatrician with a chief complaint of the infant refusing the breast feed. The neonatologist observed abnormal swelling on the lower gum pads and referred the patient to the pediatric dentist for the opinion and management. The infant showed no evidence of any systemic illness, congenital anomalies or syndromic involvement. On intra oral examination a neonatal tooth was observed in the mandibular right central incisor region that was mobile on palpation. The radiographic examination of the child was not possible. The weight of the baby was 2.1kgs at this time. The mother had previous history of miscarriage and it was her second pregnancy and all the vitals were normal at the time of delivery. Parents could not recall any related family history. Since the tooth was mobile, it was extracted under LA infiltration with the dose of 0.3ml, 2% lignocaine. During the extraction precautionary gauze was placed in the oral cavity that served as a pharyngeal guard. The tooth was removed with help of periosteal elevator and extraction forceps in NICU itself. Complete hemostasis was achieved. The patient was kept under observation and showed no signs of any complications of cyanosis or hypoxia. The extracted tooth ie81 resembled a small rudimentary crown like structure with no root formation.

The mother of the infant contacted us telephonically around 40th week and reported about the occurrence of a similar tooth like structure on the left -front region of the lower jaw, that was mobile and associated with redness of the gums. The picture shared by her suggested the eruption of a neonatal tooth in 71 region. (Fig. 2) Since the tooth was

mobile and causing bleeding of the gingival tissue during feeding hence immediate extraction was advised. However, 2 days later when the mother and the patient reported to the clinic, it was found that the neonatal tooth had exfoliated naturally and mother had brought the tooth along. (Fig. 3) The baby was fine and had begun to feed normally so no intervention was done and a review checkup was suggested after 4 weeks.

During the patient review, around three months later, on intra oral examination, an elevated soft tissue growth was seen on the alveolar ridge in anterior mandibular region. The gingival mass was approximately 1 cm in diameter of normal pink colour without any sign of inflammation or ulceration. (Fig. 4) The IOPA radiograph of that area revealed, a diffuse mass of a calcified tissue in the socket corresponding to 71 without the evidence of any pathology in the neighbouring tooth germ. (Fig. 5) Since the neonatal tooth in relation to 71 had exfoliated naturally and no curettage of the socket could be performed, so the viable remnants of some ectomesenchymal tissues of dental papilla might have proliferated and continued to grow into a diffuse, irregular mass resulting into a residual neonatal tooth which is rare occurrence. Since it was asymptomatic and not causing any feeding issues, no treatment was rendered. The child was put under observation for any further developments. Two month later, the mother shared a picture via email which showed the lesion had reduced in size further and decided to return for follow up if it turned symptomatic (Fig. 6)



Fig. 1: Subject being treated for acute respiratory distress syndrome



Fig. 2: Neonatal tooth in 71 region



Fig. 3: Exfoliated rudimentary tooth



Fig. 4: Soft tissue growth in 71 region

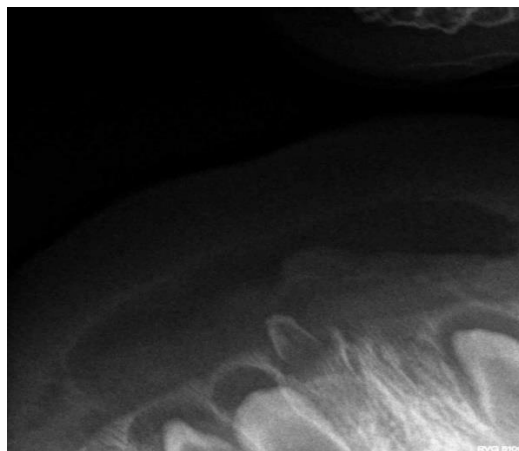


Fig. 5: Radiograph showing residual neonatal tooth 71 region



Fig. 6: Reduced size of soft tissue growth

Discussion

As specified by WHO criterion, a preterm or premature birth is either when the birth occurs earlier than 37th weeks of gestation or if the birth weight of the infant is less than 2500gms and is estimated to occur in 6% of the live births globally. The circumstances responsible for preterm child births are complex and largely unknown and primarily related to the mother and fetus.^{25,31} Most commonly responsible factors include very young age of the mother, severe anemia, multiple pregnancies or multiple fetuses, maternal smoking, stress, inadequate fetal care among others.³⁰ In cases of newborn infants with a very low birth weight (less than 1500 gm), or with gestational age below 30 weeks, significant delay in the eruption of first deciduous tooth has been demonstrated when compared with those of low birth weight (LBW<2500gms) and normal birth weight(NBW 2500 gms) or full term infants.^{25,26,31} Since the prematurely born infants exhibits various health issues and serious complications that are handled during the neonatal period with a constant support of mechanical ventilation which may also tend to cause delay in dental eruption.³⁰ Traumatic oral manipulations during prolonged mechanical ventilation for neonatal illness also result in postnatal nutritional deficiencies resulting in disruption of the process of dental development^{29,32} Nutrition is found to be an important factor contributing to the normal development and eruption of the primary dentition as well as being a consequential neonatal indicator in the recovery from the neonatal respiratory

distress.^{29,33,34} Notable correlation has also been observed in postnatal weight gain and the age of tooth eruption in preterm infants.²⁹

Despite the general observation of delay in timing of eruption of first tooth, natal/neonatal teeth have been reported in preterm infants though it is extremely rare.⁸ El Khatib K³⁵ reported the presence of natal teeth in two preterm infants, in a retrospective study of 17000 infants who were examined in the Neonatal Department of Children Hospital between 1984 and 2001.³⁵ In the present case the neonatal tooth was diagnosed in mandibular right central incisor region, in newborn male at a gestational age of 35 weeks. The neonate was born through cesarean section. The baby was anemic and born with respiratory distress syndrome and was admitted in NICU for the management. Many clinicians have highlighted the importance of correct diagnosis of natal and neonatal teeth, as these teeth are usually primary teeth of normal dentition and should not be wrongly identified as supernumerary teeth.¹ The majority of natal /neonatal teeth belong to the normal series and only a few percentage are supernumerary.^{13,35} In this case it was strenuous in achieving a radiographic evaluation due to the infant's age and fragile condition that prevented an immediate confirmation of whether the tooth was of normal series or a supernumerary tooth. However, during the follow up reviews and radiographic appraisal it was diagnosed to be a primary tooth.

The decision to maintain or to extract the neonatal tooth should be evaluated in each case with due considerations made in relation to clinical management, parental opinion and complications involved with this rare occurrence. On occasion they may exfoliate spontaneously or require extraction. Extraction of natal /neonatal teeth is indicated in certain situations where the tooth is supernumerary, or if extremely mobile and a risk of dislocation and eventual aspiration is anticipated.^{9,13,22,36} Although several authors have suggested the possibility of aspiration of these teeth, however in fact, it is unrealistic to occur and there are no confirmed reports in the literature of the aspiration but incidents of spontaneous tooth exfoliation have been reported¹⁶. Spouge and Feasby⁵ have pointed out that inhalation of these teeth has never been reported in literature and the risk is probably more imaginary than real. Another observable clinical complications related to natal /neonatal teeth are ulceration of tongue, lingual frenum or mother's breast, during feeding.^{3,9,13,23} Sometimes these teeth result in the ulceration on the ventral surface of the infant's tongue or mother's breast sufficient enough of causing hindrance in feeding which may put the neonate at risk of nutritional deficit.^{36,37} In such cases the preferred treatment includes either the grinding of teeth to smoothen the edges or the placement of round smooth composite resin over the incisal edges or extraction of the offending tooth.^{1,9,13} Some authors suggest that early extraction of these teeth might lead to overcrowding in the permanent dentition as the spaces created by their loss are occupied by the pathological migration of the adjacent teeth.^{9,23}

In the present case the decision of the extraction of 81 was made because of the mobility associated with neonatal

tooth which was interfering with the feeding. The extraction was done under local anesthesia infiltration with the dose of 0.3ml, 2% lignocaine. Since the infant was preterm prophylactic administration of Vit K was done by the neonatologist ahead of the planned dental procedure, as a part of first hand medical care for the prevention of hemorrhage, because blood coagulation mechanism may not have established fully.^{37,38} Another rare complication observed in newborns is methemoglobinemia, related to the use of local anesthetics, that can be acquired or congenital and is more common in infants younger than 3 months of age. Clinically, the infant reveals signs of cyanosis and hypoxia along with appearance of dark blue coloured skin following the application of the anesthetic agent.³⁹ Caution must be used while selecting local anesthetic agents for infants. Lidocaine, mepivacaine, articaine have moderate risk while Prilocaine and Benzocaine have high risk of inducing clinically symptomatic methemoglobinemia. The combination of prilocaine and lidocaine is considered to have a low risk if used for dental analgesia. Even a single spray of topical anesthetic agent can result in episode of methemoglobinemia.³⁹ It's a life threatening clinical condition and the dentist should be aware of this medical condition and be able to provide prompt diagnosis and effective management. It is better to take an opinion of a specialist like hematologist before proceeding with the extraction of a natal or neonatal tooth which should be ideally performed in a hospital set-up under the supervision of a cardiologist and pulmonologist in susceptible patients.^{39,40}

Complications following extraction are very rare but some authors have reported the recurrence of natal or neonatal tooth or continued growth of the radicular mass. Sook Hee Kim et al²¹ (2016) reported the development of tooth like structures with no soft tissue masses, following the extraction of neonatal teeth in 3 of the 8 cases they investigated from 1962 to 2009 and the excised mass in one of the cases showed intact dental papilla.²¹ Similar cases have been reported in literature where there was continued formation of the tooth material following the spontaneous exfoliation or extraction of the natal and neonatal tooth.⁴ The explanation for the resulting residual tooth or the masses containing dentin or other tissues of odontogenic ectomesenchymal origin could be related to the retained dental papilla. It has been suggested by few authors that during the extraction of natal tooth.²¹ only the external hard tissue is removed while the internal part of the dental papilla remains within the socket that continues to proliferate following extraction or exfoliation, and this risk is enhanced if the tooth naturally exfoliates or enamel shell is removed by patient's fingers.^{21,40,41} Based on the cases reported by Southam JC it has also been hypothesized that subsequent to the extraction of the coronal portion of the natal and neonatal tooth, the exposed part of the dental papilla along with odontoblasts and remnants of Hertwig epithelial root sheath are presumed to become extinct but seldom, enough portion of the tooth forming components withstand necrosis and remain vital and result in formation dental structures.⁴¹ The mesenchymal stem cells present in the dental papilla morpho-

differentiate into the several types of the cells like osteoblasts, chondrocytes, adipocytes, endothelial cells and odontoblasts.⁴³ It is assumed that progenitor cells destined to become odontoblasts convert into osteoblasts which have a similar lineage. The presence of dentin, osteodentin and osteoid in the excised masses could be the result of such differentiation, although more scientific evidence is needed to prove this.²¹ Till date there is no consistent nomenclature to define the formation of these hard tissues following the loss of the natal and neonatal teeth. It has been referred as “tooth like structures”^{10,41} “tumour like masses”⁴² or “Odontogenic Remnants” or “irregular masses of Dentin”.⁴¹ The term “Residual Natal Tooth” was introduced by Tsubone H et al (2002)⁴³ for the calcified hard tissues that were removed following the extraction of natal tooth,⁴³ but no terminology has been formulated so far for the natal or neonatal teeth occurring in preterm infants. The frequency of residual tooth is not well established yet and there is only one published report by King and Lee (1989) which revealed only 9.1% of infants with natal and neonatal teeth developed residual teeth following the exfoliation or extraction of these teeth¹⁰. This suggests that that most of such cases do not encounter residual tooth formation subsequent to the extraction, but still there is an involvement of 9.1% of the risk and therefore the parents should be informed about the possibility and the need for regular follow up.⁴

To prevent the formation of residual tooth it has been suggested by some authors to follow curettage of the underlying tissues of the dental papilla after the extraction of the natal or the neonatal tooth.^{4,6,10,23} Since routine curettage would necessitate the need of the use of injective local anesthesia which has a risk of inducing trauma and would be an aggressive approach. If it is possible to remove natal and neonatal tooth with only topical anesthesia then no curettage is recommended and the infant can be monitored for the residual tooth. In our case 71 exfoliated naturally and no curettage was possible which resulted in the residual tooth formation. Moreover, further studies are needed to mandate curettage after extraction of natal or neonatal teeth.⁴

Conclusion

The dentist and the neonatologist handling the infant should be aware of the possibility of residual natal/neonatal teeth and should be careful while extracting the natal or the neonatal tooth. If the teeth are not causing any abnormal clinical manifestations, they should be retained as long as possible. Parents should be counselled and educated about the sequelae of such extractions in preterm and full-term infants and reinforce maintaining good oral hygiene and follow regular dental reviews.

Conflict of Interest: None.

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