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Original Research Article

Prevalence of developmental anomalies among infants 0-12 months in Mehsana district, north Gujarat, India

Charmi Patel[1]*, Shoba Fernandes[1], Yash Bafna[1], Darshan Patel[2], Dimpal Parmar[1], Dharati Patel[1]

[1]Department of Pediatic and Preventive Dentistry, Narsinhbhai Patel Dental College and Hospital, Sankalchand Patel University, Visnagar, Gujarat, India
[2]Private Practitioner, Avani Children Hospital, Mehsana, Gujarat, India

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***Correspondence:**
Dr. Charmi Patel,
E-mail: charmipatel609@gmail.com

ABSTRACT

Background: Developmental anomalies like Epstein pearl, Bohn's nodule, gingival cyst, mucocele, natal teeth, tongue tie (TT), lip tie manifest in the neonate. A majority of them are benign and asymptomatic commonly resolving without any intervention. Some anomalies may require surgical intervention to avoid breast-feeding difficulties. Extensive clinical examination and knowledge of various lesions are essential for accurate diagnosis, management and parental advice. Aim was to determine the prevalence of developmental anomalies among infants (0-12months) and its effects on breastfeeding.

Methods: After approval of IRB (Institutional Review Board), multistage sampling was performed to include the population of Mehsana district by dividing the district into 3 zones (north, central, south). 480 Infants were included and examined in the study according to selection criteria. Infants with cleft lip palate were excluded from the study. Intraoral examination was performed and assessment of TT, lip tie was done using Hazelbacker (HB) criteria and Kotlow's classification. Informed written consent was obtained from parents. Mothers were asked about difficulties during breastfeeding via questionnaire. The option for surgical intervention was given to the parents when indicated.

Results: A total of 480 infants (male 297 and female 183) were examined. The prevalence of Epstein pearls was 39.4%, Bohn's nodule 6.5%, mucocele 1.5%, gingival cyst 0.4%, TT 5.2%, class III and class IV lip tie 94.1%, natal teeth 0.4%. The total, 14.8% infants who had lip tie, struggled to latch on to the breast while nursing which was statistically (0.02) significant.

Conclusions: The prevalence of class III and class IV lip tie (94.1%) and Epstein pearls (39.4%) was high as compared to other developmental anomalies among the infants of Mehsana district.

Keywords: Bohn's nodule, Breastfeeding difficulties, Epstein pearl, Natal teeth, TT-lip tie

INTRODUCTION

Infant general well-being is inextricably linked to their oral health. Preventive oral care in infancy is the basis of future oral health. A prominent concern for paediatric dentists is to ensure that newborns receive an oral health risk assessment from their primary oral health care provider or pediatrician before 6 months of age. Several intra oral findings of infants are unique and peculiar to the development period and often create apprehension and anxiety among parents. The lesions of the oral cavity comprise an important arena of the pediatric speciality, yet many are misdiagnosed or left untreated due to lack of resources and parental education. Their management requires thorough knowledge of the various lesions and accurate clinical assessment for diagnosis, prognosis, treatment and parental counselling. So, it is important to perform thorough examination, to rule out specific oral anomalies like tongue tie (TT), lip tie, natal teeth that may cause breastfeeding difficulties.[1]

The present study was performed as prevailing data regarding the prevalence of developmental anomalies among infants for Mehsana district, Gujarat, India, was limited.

METHODS

This descriptive cross-sectional study was conducted, upon procuring Institutional Review Board (IRB) approval in order to assess prevalence of oro-facial anomalies among infants (0-12 months) and its effects on breast-feeding on 480 subjects of general population in Mehsana district, North Gujarat, India between Feb 2022-Mar 2022. The study was registered under Clinical Trials Registry India (CTRI) with registration number CTRI/2022/02/040410. Sample size collection was done using the formula $n=4PQ/L^2$; Where p= prevalence: based on pilot study, prevalence calculated was 12%; Q = (1-p); L= margin of error = 3%. For the multistage sampling, Mehsana district was divided into three zones north, central, south zones. The lottery method was used to select one Taluka per zone. From each Taluka one government and one private children's hospitals were selected. Healthy infants (in age group of 0-12 months). Infant with any congenital anomalies like cleft lip and palate were excluded from the study. A total of 600 infants were assessed for enrolment in the study (STROBE chart). Out of the total, 3 infants were excluded as per the exclusion criteria (cleft lip palate), further 117 were excluded as they declined to participate, so finally 480 subjects were examined and mothers were interviewed. All the infants were examined by knee-to-knee position with mothers. Full head and neck examination with all soft/hard tissue examination of oral cavity was performed by trained and calibrated examiner. Intraoral examination was performed and assessment of TT, Lip tie was done using Hazelbacker (HB) criteria and Kotlow's classification.[2,3] The option for surgical intervention was offered to the parents after examination when required. The Chi-square test was applied to check the association between developmental anomalies and breastfeeding difficulties.

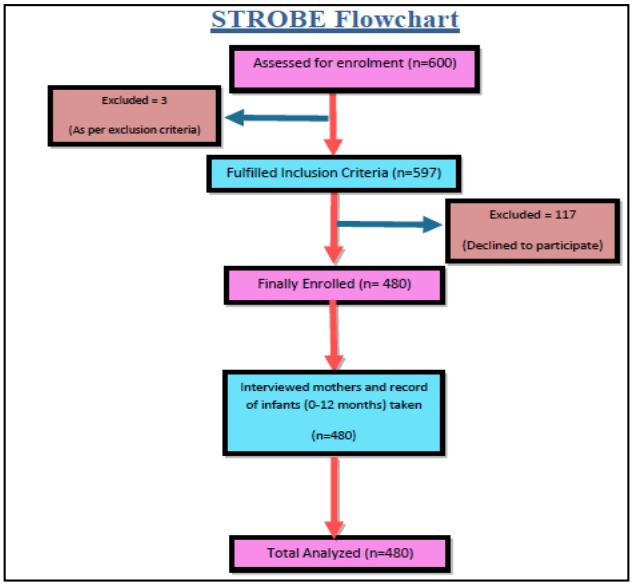

Figure 1: STROBE flowchart.

RESULTS

In the present epidemiological study, there were 183 females and 297 males among 480 infants of 0-12 months in Mehsana district. There were 381 infants of 0-3 months of age group, 55 were of 4-7 months of age group and 44 were 8-12 months of age group.

Table 1: Prevalence of developmental anomalies based on location.

	Location	Frequency	Percentage
Natal/neonatal teeth	Lower anterior	2	0.4
Epstein pearls	Mid-posterior palate	189	39.4
Bohn's nodules-maxilla	Buccal anterior	12	2.5
	Buccal posterior	8	1.7
Bohn's nodules- mandible	Buccal anterior	8	1.7
	Buccal posterior	1	0.2
	Lingual anterior	2	0.4
Mucocele	Lower lip	7	1.5
Dental lamina cysts/gingival cyst- mandible	Mandibular anterior alveolar ridge	2	0.4

In the present study prevalence of natal/neonatal teeth was 0.4%, dental lamina cysts/gingival cyst 0.4%, Epstein pearls (mid-posterior palate) 39.4%, Bohn's nodules 6.5%, mucocele 1.5% (Table 1). According to Kotlow classification prevalence of maxillary lip tie (class III and class IV) was 94.1% and class II was 5.8% (Table 2).

According To HB criteria there was 5.2% cases of TT. In the present study among 5.2% of TT, there were 0.6% in which frenectomy was indicated as their tongue on lift was heart shaped with a short lingual frenum (Table 3).

Table 2: Prevalence of Lip tie according to Kotlow Classification.[3]

Kotlow classification of maxillary frenum attachments	Frequency	%
Class II	28	5.8
Class III	292	60.8
Class IV	160	33.3
Total	480	100.0

Feeding difficulties faced by mothers of infants with TT, lip tie, natal teeth was minimal in the present study and statistically association between TT, lip tie, natal teeth with breastfeeding difficulties was insignificant (p value >0.05).

Table 3: Prevalence of TT according to Hazelbacker (HB) score.[32]

HB score	Frequency	%	Frenotomy
9	22	4.6	Not indicated
<8	3	0.6	Indicated
10	455	94.8	Normal
10	480	100.0	Total

Table 4: Association between age and different developmental anomalies.

Developmental anomalies	Age			P value
	0-3 months	4-7 months	8-12 months	
Tongue tie	21 (84%)	3 (12%)	1 (4%)	0.00
Lip tie (class III and class IV)	355 (79.4%)	54 (11.5%)	43 (9.2%)	0.19
Prescence of natal/neonatal teeth	2 (0.5%)	0.0%	0.0%	0.77
Epstein pearls	165 (43.3%)	15 (27.3%)	9 (20.5%)	0.002
Bohn's nodules	31 (8.1%)	0.0%	0.0%	0.01
Mucocele	6 (1.6%)	0.0%	1 (2.3%)	0.59
Dental lamina cysts/ gingival cyst	2 (0.5%)	0.0%	0.0%	0.77

Developmental anomalies were more significant in 0-3 months of age group in the present study (Table 4).

Table 5: Hazelbaker the assessment tool for lingual frenulum function (appearance items).[2]

Appearance items	Score
Appearance of tongue when lifted	2= round or square
	1= slight cleft in tip apparent
	0= heart-shaped
Elasticity of frenulum	2= very elastic
	1= moderately elastic
	0= little or no elasticity
Length of lingual frenulum when tongue lifted	2= >1cm or embedded in tongue
	1= 1 cm
	0= <1 cm
Attachment of lingual frenulum to tongue	2= posterior to tip
	1= at tip
	0= notched
Attachment of lingual frenulum to inferior alveolar ridge	2= attached to floor of mouth or well below ridge
	1= attached just below ridge
	0= attached at ridge

Scoring: Frenotomy is necessary if appearance score is <8

DISCUSSION

Infant oral health is one of the foundations upon which preventive education and dental care must be built to enhance the opportunity for a lifetime free from preventable oral disease. A variety of anomalies like Epstein pearl, Bohn's nodule, gingival cyst, mucocele, natal teeth, TT, lip tie can be found in the oral cavity of the neonates. Identification and differentiation of these developmental anomalies in the oral cavity remain crucial in order to assist doctors in prioritising the treatment of the issues and to inform concerned parents.[4] In the present study there were 183 females and 297 males among 480 infants of 0-12 months in Mehsana district.

Alois Epstein, a paediatrician from Prague, initially identified Epstein disease in 1880 as being characterised by tiny nodules in a newborn's oral cavity. Based on their clinical presentation, Fromm divided several kinds into categories in 1967. They can be seen in the oral cavity as growth ectodermal laminar surfaces (dental lamina cysts) or on the gingiva (Bohn's nodules) midline of the palate (Epstein's pearls).[5,6] In the present study the prevalence of Epstein pearl in the midline within the median raphe of the hard palate was 39.4%. This finding was similar ranging between 35.2-38.3% in other parts of India.[7-9] Although prevalence was lower in the present study when compared with study conducted by Haveri et al which shows 89.1% in Karnataka, India; Perez-agirre et al 66% in Korea, and in USA ranging between 65.1%-73%.[1,6,10,11] The prevalence was higher (27.4%) when compared with study done in Brazil.[12]

The age distribution for Epstien pearls showed 43.3% cases at 0-3 months infants, 27.3% at 4-7 months, 20.5% and 8-12 months babies which was statistically significant (p=0.00). This could be explained at least in part to the fact that in many cases, Epstein pearls disappear on their

own within a week or two of birth as a result of friction in the infants' mouth either from breastfeeding, bottle feeding, or use of a pacifier. When the cyst walls fuse with the oral epithelium, cystic keratin is discharged, and this is thought to be one of the mechanisms underlying the cysts' elimination in postnatal life. But it's possible that some of the cystic epithelium in the adult gingiva's midpalatal area will continue to be dormant.[8]

Bohn's nodules are smooth, keratin-filled nodules or papules that are whitish in colour that resolve within first 2 to 3 months of life.[13] The prevalence of Bohn's nodule in newborns, in current study was 6.5% which was lower as compared to studies conducted in India 47.4%, in Brazil 26.3%, in USA 23%, in Korea 70%.[1,12,14,15] Among study population, Bohn's nodule (2.5%) most commonly was found in the lower anterior region. In this study, infants of age 0-3 months exhibited 8.1% of Bohn's nodule when compared to 4-7 months and 8-12 months of age groups.

The remnants of dental lamina that form gingival cysts or dental lamina cysts in infants are frequently seen. These are asymptomatic, 1-3 mm in diameter, nodular, and creamy white in colour. They are most frequently found bilaterally on the ridge of the alveolar bone.[16] The observations in this study of gingival cyst of neonates located at mandibular anterior alveolar ridge was 0.4%, which was lower as compared to study conducted by George et al with 13.8% of prevalence of gingival/dental lamina cyst in India, in Brazil 11.1%, in Korea 45%, in USA 53.5% and 25% of children from different parts of USA.[1,8,12,16,17] Predominantly all the authors observed location of dental lamina cyst to be in mandibular anterior ridge. However, Cataldo et al 36.5% found the prevalence of cysts on the maxillary alveolar ridge in USA.[11] In the present study, age 0-3 months of infants exhibited 0.5% of gingival cyst when compared to 4-7 months and 8-12 months of age groups. It is more difficult to elucidate the method of their disappearance. Paula et al suggested that as the cysts developed, the epithelium differentiated; fused with the oral epithelium and the contents are discharged.[18] Some cysts may produce keratin, enlarge, extend to the surface and rupture during the first few months after birth.

A well-defined, transparent, and fluctuant swelling is called a mucocele. Its most common occurrence site is the lower lip lateral to the midline. The peak frequency seen in the second and third decades.[19] Prevalence of mucocele in the present study was 1.5%, located at buccal mucosa of the lower lip which was lesser when compared with Tantray et al 2.4% in India, Shapira et al 11.6-24.5% in Israel.[14,20,21] In this present study, there were 1.6% of Mucocele at 0-3 months infants, none at 4-7 months and 2.3% at 8-12 months.

Worldwide the prevalence of natal and neonatal teeth varies from 0.003% to 0.1%.[14] In the Mehsana populace, the prevalence of natal teeth was 0.4% located in the lower anterior region. In other parts of India prevalence ranges from 0.1-0.6%.[7,8] Globally various countries presented the prevalence of natal/neonatal teeth which was 0.4% in Korea which is similar to present study.[1] A lower prevalence was found by in various parts of USA ranging between 0.07-0.2%, in Finland 0.1%[22], in UK 0.03%, in Canada 0.03%-0.05% and in Hong Kong 0.07%.[15,21-25] Higher prevalence was also observed in USA ranging 0.7-0.9%.[17] The mothers of infants with natal teeth reported no difficulties while nursing. Probably the well-rounded, relatively firm placement of the teeth avoided major discomfort for mother and infant. Jamani et al reported breastfeeding difficulties and Bafna et al reported presence of an ulcer on the ventral surface of the tongue (Riga Fede) in infants with natal teeth.[26,27]

Ankyloglossia also known as 'tongue-tie (TT)', is a congenital anomaly observed in newborns and children, and is characterized by an abnormally short lingual frenulum. The prevalence of TT in present study was 5.2% which is higher as compared to the study conducted in India was 0.52%, in Sweden 2.5%, in Korea 1.5%, in Peru 4.93%.[1,28-30] Also lesser when compared to study reported in India 17.7%, in UK 10.7%, in US 9%, in Spain 46.3%.[15,31-33]

In the present study, TT prevalence was high among 0-3 months age group subjects (84%) as compared to age group 4-7 months and 8-12 months which was statistically highly significant. In the present study 4.6% cases had HB score 9 and 0.6% had less than 8 (Table 3 and 5).[2] Among all TT cases 16% mothers of infants with TT faced difficulties while nursing. The milder form of TT may be the reason for minimal feeding difficulties. A study performed by Messner et al stated that difficulties reported by mothers of affected infants was 25% and concluded that TT can adversely affect breastfeeding in a minority of cases.[34] Rech et al demonstrated 4.93% case of TT, among them only 26.7% presented difficulties with breastfeeding in the first hours of life and only 13.3% persisted having difficulties in breastfeeding throughout the months, and were referred for frenotomy.[30]

Kotlow defined the "lip-tie" as the problematic frenulum (the "lip-tie") that prevented the newborn from successfully latching by preventing the top lip from flanging around the nipple. His grading system for lip-tie severity has been employed as a clinical consideration in the decision to release.[35] As regards the prevalence of upper lip tie in newborns, according to Kotlow's classification the prevalence of class II Lip tie in our present study was 5.8%, class III Lip tie was 60.8%, class IV lip tie was 33.3%. There were no cases of class I lip tie in this study population showing similar findings with the study conducted by Ghaheri et al who also revealed 46% of class II and class III lip tie, 54% of class IV lip tie cases.[36] Class II lip tie in our present study was 5.8% which was lower in comparison to the study conducted by Flink et al which was 6.7%, George et al 68.7%.[8,29]

Niranjan et al 79.1% and Flink et al 76.7% who found maxillary frenum attached on the alveolar crest (class III) which is higher as compared to our study (60.8%) and its lower when compared to the study conducted by George et al which was 31.3%.[7,8,29]

Class IV Lip tie (maxillary frenum attached palatal to the alveolar ridge extending to incisive papilla) in our present study was 33.3% which was higher to the study conducted by Flink et al (16.7%).[29] However George et al did not find any cases of maxillary frenum attached palatal to the alveolar ridge (class IV) which is dissimilar to our study.[8] The probable reasons for the difference are effects of racial differences and sample variations. A study conducted by Pransky et al reported the prevalence of lip tie was 6% in Canada.[37] The probable reason for this may be differing inspection criteria for assessment which were flanging of upper-lip, or curled upper-lip during breastfeeding.

In present study there were 78.5% cases of lip tie (class III and class IV) at 0-3 months infants, 11.9% at 4-7 months, 9.5% and 8-12 months infants respectively. Lip tie (class II and class III and class IV) prevalence was high among 0-3 months age group subjects (79.4%) as compared to 4-7 months age group and 8-12 months which was 11.5% and 9.2% respectively which was statistically highly significant. In the present study there were 14.8% infants who had lip tie and struggling to latch on to the breast while nursing. A study by Ghaheri et al demonstrated that maxillary labial frenulum restriction can also affect latch quality on to the breast showing 81% infants with lip tie had poor latch on to the breast.[36] Wiessinger et al presented a case report of an infant with lip tie having poor latch on to the breast with positive familial history of lip tie.[38] Cordaro et al also reported that 88% of infants with lip tie and TT had difficulty in achieving a good latch onto the breast.[39]

Based upon the findings of the study, Epstein pearls was more prevalent than Bohn's nodules in the populace of Mehsana district. Mucocele, dental lamina cyst/gingival cyst, natal teeth were less prevalent than Epstein pearls and Bohn's nodules. Prevalence of developmental anomalies like TT, lip tie was similar to the population in other parts of country. In the current study mothers of infants with TT, upper lip tie and natal teeth faced minimal discomfort while nursing hence surgical intervention was not a requisite.

CONCLUSION

This epidemiological descriptive cross-sectional study showed all the developmental anomalies most commonly were observed in the age group of 0-3 months with the male gender predilection. The prevalence of gingival cyst and natal teeth 0.4% which was present on the mandibular anterior region. In the present study, the prevalence of TT was 5.2%. As regards to the prevalence of lip tie 94.1% (class III and class IV).

The present study plays a vital role to highlight a hidden need for early intervention of the commonly encountered developmental anomalies seen in infants in Mehsana district of Gujarat. More studies should be conducted to Improve awareness among clinicians and parents that would lead to greater degree of diagnosis and remedial care for the same.

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