

Intraoral Findings and Other Developmental Conditions in Mexican Neonates

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ABSTRACT

Purpose: The objective of this study was to investigate the prevalence of intraoral findings and other minor developmental malformations in newborns from San Luis Potosi, Mexico.

Methods: Study subjects were neonates born in San Luis Potosi Morones Prieto Hospital between September 1989 and February 1990. All subjects are examined at this hospital within 20 hours of birth. Premature babies or those requiring intensive care were excluded. Examinations are performed by a team consisting of a geneticist, an oral pathologist, 2 dentists, and an oral surgeon using mirrors, tongue blades, and a flashlight.

Results: The team examined 2,182 neonates and found a frequency of 99% for congenital oral cysts, 2% for natal teeth, 11% for ankyloglossia, 8% for commissural lip pits, and 54% for congenital vascular malformations. The male/female ratios for ankyloglossia and natal teeth were 1.5:1 and 1:2.3, respectively.

Conclusions: Babies born at the same hospital demonstrated a high rate of oral cysts, natal teeth, ankyloglossia, and commissural lip pits.

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Few published studies describe clinical findings in the newborn's oral cavity. Based on previous neonate research, a substantial prevalence of palatal and alveolar cysts was expected. Commissural lip pits, ankyloglossia, and natal teeth occur more infrequently. Although most of these anomalies are innocuous and resolve with age, recent research has shown that some of these lesions may have significant clinical relevance.

This study's purpose was to collect data on several congenital oral and perioral anomalies and other developmental conditions in 2,182 newborns from San Luis Potosi, located 356 km north of Mexico City, Mexico.

METHODS

Data were collected from newborns from low-income individual families born at San Luis Potosi Morones Prieto Hospital, San Luis Potosi, over a period of 6 months between September 1989 and February 1990. Newborns had to be full-term, breathing on their own, and no older than 20 hours.

A team consisting of an oral pathologist, a geneticist, an oral surgeon, and 2 dentists were present during each infant's examination. All oral examinations were performed exclusively by the oral pathologist, who was also the project's director. The geneticist only evaluated nonoral congenital anomalies. The oral surgeon was there in case consultation was needed. The 2 dentists were present to take notes. Data were entered on specially designed data sheets for each neonate, including date of inspection, name, identification number, and gender.

Visual examinations of the oral cavity were performed with the aid of disposable tongue depressors and natural and/or artificial light. When necessary, oral mirrors and/or disposable latex rubber gloves were used. Photographs of representative cases were also taken.

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The following terms were used to diagnose and define the conditions being investigated: gingival cysts of the newborn; ankyloglossia; birthmarks; commissural lip pits (CLP); and natal teeth.

Gingival cysts of the newborn are small, firm, white, or grayish-white lesions seen either on the palate, alveolar crest of the maxilla or mandible, or buccal and lingual aspects of the maxillary and mandibular ridges. The term “Epstein’s pearls” is sometimes used when the lesions are located along the palate’s midline. “Dental lamina cyst” is the preferred term when they are on the ridges’ crest, and “Bohn’s nodules” is preferred when they are along the ridges’ buccal and lingual aspects. These classifications are often used interchangeably.

Microscopic sections reveal that the nodules are small, superficial, keratin-containing cysts lined by stratified squamous epithelium and arising from remnants of degenerating dental lamina (rest’s of Serres). They are spontaneously shed within a few weeks. During the inspections, the number and location of cysts on the palate and on the mandibular and maxillary alveoli were noted. The location of each gingival cyst was recorded as being in 1 of 20 maxillary areas or in 1 of 30 mandibular areas.

Ankyloglossia, a thick frenulum on the tongue’s ventral surface, was diagnosed when the lingual frenum: prevented tongue protrusion; extended to the tongue’s papillated surface; or caused a fissure in the tongue tip during normal movements. Fixation results when the lingual frenum is attached too far forward toward the tongue’s tip.

CLP, a marked invagination at one or both corners of the mouth on the vermilion surface and into which a probe could be introduced, were recorded as absent, right, left, or bilateral.

Birthmarks, also known as skin lesions, were noted and defined as follows:

1. Vascular malformations: Capillary malformations (salmon patch, stork bite) that are flat pink to light red, well defined, irregularly outlined macules that blanch completely on pressure;
2. Pigmented lesions:
 - a. Café au lait macules (CALM), defined as flat, evenly pigmented, light to dark brown spots without speckles, having many different shapes and skin markings matching the adjacent skin; and
 - b. Congenital melanocytic nevi (CMN), defined as raised, light to dark brown lesions with black or blue foci with increased skin markings compared

Table 1. Summary of Results

	Subjects (N=2182)	%	Male (N=1111)	Female (N=1071)	Male (%)	Female (%)	Ratio (M:F)
Oral cysts	2163	99.1	1103	1060	99.3	99.0	1:1
Ankyloglossia	231	10.6	139	92	12.5	8.6	1.5:1
Commissural lip pits	184	8.4	91	93	8.2	8.7	1:1
Birthmarks	1209	55.4	569	640	51.2	59.8	1:1.1
Natal teeth	50	2.3	15	35	1.4	3.3	1:2.3

to the surrounding tissue; the epidermal ridges are more prominent, and the pattern of the ridges is not consistent with that of the adjacent skin. The textures vary and they may or may not have hair. (Note: No biopsies were performed and thus diagnoses were purely clinical.)

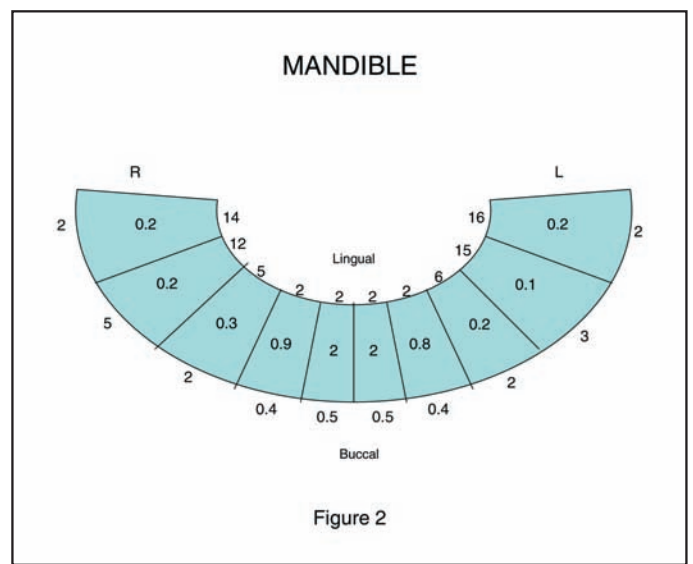
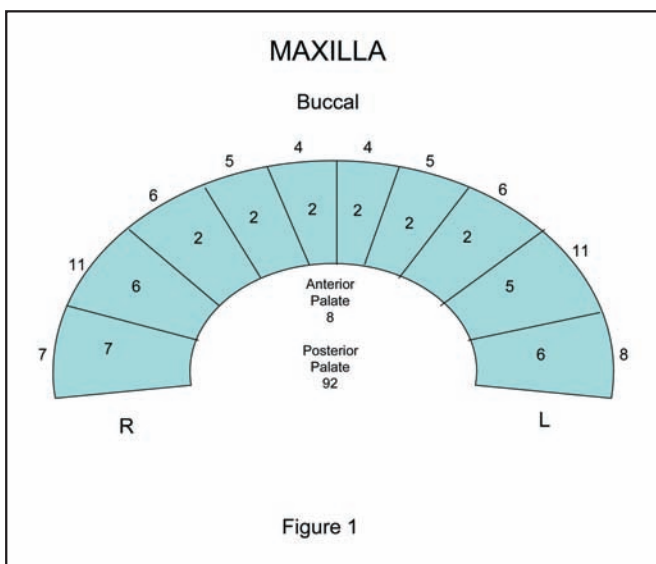
Natal teeth were defined as teeth observed within 20 hours of birth. Other intraoral and minor extraoral defects such as polydactyly, simian creases and dislocated hips were recorded in a miscellaneous category and were not part of this study.

RESULTS

A total of 2,182 newborns was examined: 1,111 (51%) were males, and 1,071 (49%) were females. Five common developmental anomalies were investigated. A summary of the results is found in Table 1.

This study found that 78% of subjects had palatal cysts, 97% had gingival cysts, and 99% had either palatal or gingival cysts. The prevalence of cysts in the maxillary area was greater than that in the mandibular area (65% vs 35%). Posterior palatal cysts were noted more often than anterior palatal cysts (92% vs 8%). On the maxilla, cysts appeared more often on the buccal aspect. On the mandible, they appeared more often on the lingual aspect (Tables 2 and 3). The prevalence of cysts occurred bilaterally and was not influenced by gender.

Figures 1 and 2 shows the various frequencies regarding the location in the maxilla and mandible, respectively. One and a half times as many cysts were found in the area of the first and second primary molars than in the cuspid area and in the maxilla’s and mandible’s anterior regions (60% vs 40%, respectively).



The Figures indicate that 11% of the Mexican neonates examined had ankyloglossia. There was a predilection of affected males over females by a ratio of 1.5:1. CLP were present in 8% of the neonates. They were bilateral in 31% of the cases: 11 % were on the right side, and 58% were on the left side.

Fifty five percent of the neonates had at least one cutaneous lesion birthmark. The vast majority of these neonates had vascular lesions. Café au lait macules (CALM) were seen in 1.3% of the neonates and congenital melanocytic nevi (CMN) were seen in 1.4 % of the neonates with a 1.6:1 predilection for females (Table 5).

This study identified 50 subjects with natal teeth of which 1 subject had 4 natal teeth, 35 subjects had 2 natal teeth and 14 subjects had 1 natal tooth. The frequency of natal teeth in this population was 2.3% with a 2.3:1 predilection for females (Table 1).

DISCUSSION

There are few studies on the frequency and type of congenital oral lesions and developmental conditions in the neonate. Although most of these lesions are innocuous and transient, recent research has shown that some of these lesions may have significant clinical relevance. This study's results confirm some previously reported findings and counter others.

Table 2. Maxillary Cysts (N=19,836) 65%

	Number of cysts	%
Upper right buccal cysts	6598	33.3
Upper left buccal cysts	6379	32.2
Upper right ridge cysts	3669	18.4
Upper left ridge cysts	3190	16.1

Table 3. Mandibular cysts (N=10,758) 35%

	Number of cysts	%
Lower right buccal	1024	9.5
Lower left buccal	805	7.5
Lower right lingual	3804	35.4
Lower left lingual	4456	41.4
Lower right ridge	356	3.3
Lower left ridge	313	2.9

NEWBORN GINGIVAL AND PALATAL CYSTS

During the morphodifferentiation (late bell stage) of tooth development, a portion of the dental lamina fragments into numerous islands of odontogenic epithelium. These epithelial remnants of dental lamina have the capacity, at as early as 10 weeks, to proliferate, keratinize, and form small alveolar cysts in utero

In a study of 120 newborns by Donley et al,¹ it was found that a direct relationship predicts that, with increasing gestational age, increasing postnatal age, and increasing birth weight, the prevalence of cysts (palatal, alveolar, or both) increases.

Several studies have shown a racial predilection for oral cysts (Table 4). Jorganson et al² found a higher prevalence of gingival and palatal cysts in Caucasians (53%; 73%) than in African-Americans (40%; 65%). Friend et al³, had similar findings with a prevalence of gingival and palatal cysts in Caucasians as 26%; 75% versus African-Americans 11%; 55%. In a study of newborn Swedish children (98% were of Scandinavian Caucasian origin) by Flinck et al,⁴ oral cysts were found in 75% of the children, either on the palate (68%) or the alveolar ridges (22%). In 16% of the children, cysts were present on both the palate and the alveolar ridge. In a study by Ming-Hui Liu⁵ on Taiwanese

Table 4. Studies of the Frequency of Palatal and Gingival Cysts of the Newborn

	Jorganson 1982	Ikemura 1983	Friend 1990	Flink 1994	Liu 2004	Present study
Sample #	C – 596 A-A – 1568	209	C – 89 A-A – 411	1021	420	2182
Palatal cyst (%)	C – 73.0 A-A – 65.0	68.2	C – 75.0 A-A – 55.0	68.4	86.0	77.7
Gingival cyst (%)	C – 53.5 A-A – 40.0	69.6	C – 26.0 A-A – 11.0	22.0	79.0	96.9
Oral cysts (%)	NA	93.3	NA	74.9	94.0	99.1
Race	Caucasian : C African-Am : A-A	Japanese	Caucasian: C African-Am: A-A	Caucasian	Asian (Taiwanese)	Hispanic (Mexican)
Location of cysts	Pal>Max>Man	Pal>Max>Man	Pal>Max>Man	Pal>Max>Man	Pal>Max>Man	Max>Pal>Man

newborns, 86% had palatal cysts and 79% had gingival cysts, with 94% having either palatal or gingival cysts. This study of Mexican neonates found that 78% of subjects had palatal cysts and 97% had gingival cysts, with 99% having either palatal or gingival cysts. The lower prevalence of palatal cysts vs gingival cysts, as compared to previous studies, is a probable result of not including Epstein's pearls (cysts along the palate's midline) in the data. Further variance among these studies may also be due to differences in postnatal age and birth weight.

This study, as well as several others, has shown a significantly higher prevalence of cysts in the maxilla than in the mandible.^{4-6,8} On the maxilla, cysts appeared more often on the buccal side, while the mandibular gingiva demonstrated a lingual predominance.^{5,6} A notably higher frequency of palatal cysts were located posteriorly than anteriorly (92% vs 8%). According to Friend et al,³ who reported similar results, this may be due to the premaxilla being the first portion of the palate to fuse and the fact that cysts in this area may already have degenerated by birth.

One and a half times as many cysts were found in the area of the primary first and second molars than in the cuspid area and anterior regions both in the maxilla and the mandible (60% vs 40%, respectively). No other studies were found in the dental literature that defined the specific areas where the cysts predominate.

The present study's results confirm those of earlier neonate studies—namely, that oral cysts are a common finding. Practitioners should be aware of the particularly high frequency of these cysts. In 2001, Wan⁷ demonstrated that there was an association of *Streptococcus mutans* infection and Bohn's nodules in preterm infants. Risk analyses suggested that those with oral nodules were over seven times more likely to be colonized with *S mutans* at an earlier age,

thereby possibly increasing caries risk. The Wan study suggested an involved relationship between the presence of oral nodules, preterm/fullterm birth, and maternal salivary levels. To clinically apply Wan's conclusion, further studies following children from infancy to early childhood are necessary.

ANKYLOGLOSSIA

The frequency of ankyloglossia has been reported to vary from 0.4% in a study by McEnery⁹ to 4.8% cited by Messner¹⁰ with a predilection for males to females varying from 1.5:1 (Ballard JL et al.)¹ to 4:1 (Flinck A et al.)⁴. In this study, 10.6% of the neonates had ankyloglossia and it was 1.5 times more common in males than females, a difference between the genders that confirms previous findings. Some authors^{12,13} have postulated that the short frenum can

lengthen with age and use. The relatively high frequency may be due to such factors as diagnostic criteria and age-related differences.

Ankyloglossia is an oral anomaly that can cause difficulty with breast-feeding,¹⁴ speech articulation,¹⁰ and tasks such as licking lips and using the tongue to sweep the teeth free of debris. Practitioners should educate parents about the possible effects of "tongue tie" so they can make an informed choice regarding possible therapies, such as a frenotomy or frenuloplasty.^{11,15}

COMMISSURAL LIP PITS

Commissural lip pits (CLP), first described by Epstein,¹⁶ may result from a failure of mesodermal masses, from embryonal maxillary and mandibular processes to penetrate the epithelial walls completely.¹⁷ Everett and Wescott¹⁸ observed that CLP were often inherited as an autosomal dominant trait.

The prevalence of CLP in the dental literature covers a wide range. There were differences found between younger and older subjects: 2% among neonates,² <1% to 5% among children,^{18,19} and 7% to 20% among adults.²⁰ These differences would suggest that lip pits become more accentuated with increasing age.

Variance in the prevalence of CLP have been noted among various ethnic groups.^{2,21} Baker²⁰ studied US Air Force Hospital male patients and found CLP in 7% of Asians, 12% of Caucasians, and 20% of African Americans.

The relatively high frequency of CLP found in the present study may be due to diagnostic criteria and the selected population studied. The present study showed a significant predilection for CLP to occur unilaterally (69%) vs bilaterally (31%). Of the unilateral lip pits, 16% were found on the right side and 84% were found on the

left side. This increased occurrence on the left side correlates with isolated unilateral clefts, which occur twice as frequently on the left side as on the right. No other studies of unilateral pits ratio in neonates could be found in the dental literature.

BIRTHMARKS

Most birthmarks consist of vascular and pigmentary lesions. For birthmarks to provide meaningful information, the incidence and manner of presentation in a population needs to be documented.

VASCULAR LESIONS

Vascular birthmarks are classified into 2 major groups: (1) vascular malformations; and (2) vascular tumors.^{22,23} For this study's purposes, only vascular malformations were noted, since vascular tumors may present after birth in the neonatal period.

Vascular malformations are errors in morphogenesis that may affect any branch of the neonatal vasculature. These may include arterial, capillary, venous, and lymphatic tissues.²² Previous studies have shown that the most frequent vascular malformation is the salmon patch or medial telangiectatic nevus. The dental literature reveals a 20%²⁴ to 69%²⁵ variance in the prevalence range. In this study, 55% of the neonates displayed this midline capillary malformation, of which 77% were at the nape, 57% were on the eyelids, and 30% were on the glabella. These commonly involved sites strongly correlate with previous studies by Rohr²⁶ and Jacobs.²⁷

In most cases, salmon patches are asymptomatic and pose minimal cosmetic problems. They usually disappear within 2 years, although nape lesions are often persistent. It has been suggested that all infants born with capillary hemangiomas, in any location other than the glabella or nape of the neck, be followed clinically for several years to determine whether or not a systemic angiomatous disorder will develop.²⁵ No other vascular malformations were noted in this study.

PIGMENTED LESIONS

Hyperpigmented lesions at birth may be macular, papular, plaque-like, evenly colored, or speckled or spotty. It is also known that the prevalence of pigmented lesions increases during childhood and decreases in adult life. Pigmented nevi, not present at birth, develop in early infancy, complicating the definition of "congenital" nevi.^{28,29}

Even colored, light brown to dark brown macules at birth are usually Café au lait macules (CALM).²⁸ The border may resemble either a smooth "coast of California" or a jagged and shaggy "coast of Maine" appearance, both of which are

Table 5. Prevalence of CALM and CMN in various populations.

SOURCE	Total No. of patients in cohort	Prevalence of CALMs in the cohort	Prevalence of CMNs in the cohort
Alper & Homes	4641 newborns	2.7% in general 0.3% Caucasian infants 18.2% African-Am infants	1.1% in general 1.0% Caucasian infants 1.8% African-Am infants Female predilection
Hidano et al.	5387 newborns	1.7% Japanese infants	2.7% Japanese infants
Tsai & Tsai	3345 newborns	0.4% Chinese infants	1.0% of Chinese infants
Kahana et al.	1672 newborns	0.3% in general 0.11% Jewish infants 0.48% Arab infants	0.6% in general 0.35% Jewish infants 0.84% Arab infants
Osburn et al.	830 newborns	2.8% in general 0.6% Caucasian infants 8.3% African-Am infants 1.3% Hispanic infants	2.8% in general 1.0% Caucasian infants 7.5% African-Am infants 1.3% Hispanic infants Female predilection
Present study	2182 newborns	1.3% Hispanic infants	1.4% Hispanic infants 1.6:1 female predilection

nondiagnostic of a specific condition.³⁰ In newborns, the size ranges from 0.2 to 4.0 cm in diameter and increases proportionately with body growth.²⁵ Light microscopic examination of CALM shows increased melanin content in both melanocytes and basal keratinocytes, without melanocytic proliferation.³¹

A racial variation exists in the prevalence of CALM in newborns (Table 5). In a study by Alper and Holmes,²⁵ CALM appeared in <1% of Caucasian newborns in the first hours after birth, whereas 18% of black newborns demonstrated these lesions. They were also recorded more commonly in Arab newborns (0.48%) compared with Jewish newborns (0.11%).²⁴ In a cohort of 3,345 Chinese infants younger than 48 hours of age, CALM were pre-sent in <1%.³² In a study of Japanese neonates by Hidano et al.,³³ CALM were noted in 2%. In the current study, CALM were observed in 1% of the newborns. Most of the infants had a single lesion, three infants had 2 lesions, and 1 had 4 lesions. Solitary CALM are common in the general population, but multiple lesions are rare, particularly in the Caucasian population, and may indicate disorders such as neurofibromatosis, Watson syndrome, and McCune-Albright syndrome.

Congenital melanocytic nevi (CMN) are composed mostly of melanocytes, the pigment-producing cells that colonize the epidermis. Melanocytes are derived from neural crest and migrate during embryologic development to selected ectodermal sites (primarily the skin and the central nervous system). CMN are thought to represent an anomaly in embryogenesis and could be considered a malformation or a hamartoma. Lesions typically grow proportionately with the individual.³⁴ Several studies have documented the prevalence of CMN

in infants from a variety of countries and racial groups (Table 5). In a cohort of 4,641 newborns, CMN were found in 1% of Caucasians, 2% of African Americans, and <1% of Hispanics.²⁵ A study by Osburn et al³⁵ examined 830 infants and found nevi in 1% of Caucasians, 7% of African Americans, and 1% of Hispanics. Both studies found a female predilection for CMN.

In this study, CMN were diagnosed clinically in 1% of the neonates, with a 1.6:1 predilection for females. Three of 31 nevi were hairy nevi. Regarding size of the CMN, 27 were small (≥ 1.5 cm) and 4 were medium (2 x 3 cm, 4 x 2 cm, 6 x 3 cm, and 6 x 7 cm). None were over 8 cm. Variance of prevalence can be attributed to racial differences and to the fact that none of the neonates examined were over 20 hours old.

CMN are clinically significant because of their association with malignant melanoma. Some data suggest³⁶ that small (≤ 1.5 cm) and intermediate (1.5-8 cm) CMN do not pose a significantly increased risk for melanoma, but reports of childhood melanoma in patients with intermediate lesions exist.^{36,37}

NATAL TEETH

According to a 1950 study by Massler and Savara,³⁸ the term "natal teeth" is defined as the presence of teeth at birth. Morphologically, the teeth show normal size and shape, although they exhibit enamel hypoplasia and small root formation.³⁹ On the basis of dental literature data, Hebling (1997)⁴⁰ classified natal teeth into 4 clinical categories:

1. shell-shaped crown poorly fixed to the alveolus by gingival tissue and absence of root;
2. solid crown poorly fixed to the alveolus by gingival tissue and little or no root;
3. eruption of the crown's incisal margin through gingival tissue; and
4. edema of gingival tissue with an unerupted but palpable tooth.

Most natal teeth are normal primary dentition (95%), while a small percentage are supernumary.^{41,50} A majority (85%) of natal teeth are mandibular incisors, except in the case of cleft lip and palate, where they occur mostly in the maxillary cleft areas. Natal and neonatal teeth have also been found associated with certain developmental abnormalities and syndromes.⁴²⁻⁴⁴

Radiographic examination should be utilized for differential diagnosis between supernumerary primary teeth and normal dentition teeth and to rule out other oral manifestations that may be confused with the dental condition in question, such as Bohn's nodules.

Treatment of natal teeth should be conservative. If the tooth's incisal edges are sharp, they may be smoothed out or covered with composite resin to prevent the development of ulceration on the tongue's ventral surface, also known as Riga-Fede disease.^{45,49} If the teeth show excessive mobility, immediate extraction should be performed.⁴¹

The etiology of natal teeth is rather unclear, though it seems to have a hereditary component.^{38,41,46,47} Bodenhoff and Gorlin⁴⁸ demonstrated that 15% of children with natal and neonatal teeth had parents, siblings, or close relatives with a history of having the same condition. In a study by Mayhall (1967),⁴⁷ it was shown that 9% of newborn Alaskan Tlinget Indians had natal or neonatal teeth, and 62% of them had affected relatives.

The prevalence of natal teeth, as reported in the recent dental literature, varies from 1 in 3,392⁴⁴ to 1 in 376,² and a predilection for females was cited by some authors.^{41,42,44} The present study of Mexican neonates identified 1 subject with 4 natal teeth, 35 subjects with 2 natal teeth, and 14 subjects with 1 natal tooth. The frequency of natal teeth was 2%, with a predilection for females to males of 2.3:1. All teeth were located in the mandibular anterior area. The relatively high frequency found in this study might be due to diagnostic criteria and the selected population studied.

CONCLUSIONS

Based on this study's results, the following conclusions can be made:

1. There was a high prevalence of gingival cysts. The upper arch's buccal aspects and the lower arch's lingual aspects had a higher prevalence of cysts than other areas.
2. Gingival cysts were significantly more prevalent in the oral cavity's posterior area.
3. Unilateral lip pits were found more on the left side than the right side.
4. There was a high prevalence of ankyloglossia, commissural lip pits, and natal teeth.

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