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Prevalence of Dental Anomalies among Cleft Lip and Palate Patients: A Cross Sectional Study

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Abstract

This is a descriptive cross-sectional survey whose aim is to evaluate the prevalence of dental anomalies in a Moroccan cleft lip and palate population. The sample included 210 patients examined at the Operation Smile Morocco Center in Casablanca during the period from January 10, 2022 to March 10, 2022. The results showed that positional anomalies are the most frequent anomalies including ectopia with a number of 501 ectopias without significant predominance among males and females. Regarding number anomalies, agenesis is the most common anomaly with a number of 173 agenesis. Permanent incisors are the most affected by this anomaly with a significant male predominance with a rate of 38.72%. In permanent dentition, microdontia remains the most frequent shape anomaly with no significant predominance among males and females. Our study showed that amelogenesis imperfecta is the most frequent structural anomaly with a male-dominant character with a rate of 58.43%. The other dental abnormalities were very rare or even absent, therefore, they didn't have a significant rate in our survey. In conclusion, given the frequency of these dental abnormalities in patients with cleft lip and palate, early multidisciplinary management is necessary.

Subject Areas

Dentistry

Keywords

Prevalence, Tooth Abnormalities, Cleft Lip, Cleft Palate

1. Introduction

Cleft lip and palate are common craniofacial malformations, with an incidence

of 1 to 2 per 1000 births worldwide [1], 2/1000 in Northern Europe and 1/1000 in Italy [2].

Higher rates are reported in Africa [2].

Clefts of the secondary palate are common in females, while clefts of the primary palate are two times more common in males [3].

The face is derived from five buds: an odd median bud, the nasofrontal process, and two maxillary and mandibular processes. During the sixth embryonic week, the left and right maxillary buds fuse back to front with the medial nasal bud to form the primary palate. Around the seventh week, the secondary palate is formed by the front-to-rear fusion of the palatine processes of the maxillary buds [4].

In the case of the palate, two blades lower, come together and fuse in the middle, around the 40th day of intrauterine life. This closure progresses from front to back, until the uvula is formed at the posterior end. If these two blades fail to fuse, the result is a cleft palate.

The cleft may be unilateral on the right or left, or bilateral, in which case it may be symmetrical or asymmetrical [3]. Anatomo-clinical forms can therefore be very varied.

The actual cause of the fusion defect between the buds is still unknown. A genetic and environmental origin is likely [5] [6].

Smoking and the mother's use of medication (anti-epileptic drugs) may also be responsible for the appearance of a cleft [3] [7].

Several studies have demonstrated a high prevalence of dental anomalies in patients with cleft lip and palate [8] [9] [10].

The cleft lip and palate prevents the invasion of preodontoblasts and myocytes, which explains the dental anomalies and lip volume defects found [3].

Early cleft repair surgery can also induce iatrogenic dental anomalies [11] [12].

The whole dentition may be affected, with a high prevalence of teeth bordering the cleft, particularly the maxillary incisors [13] [14].

The lateral incisor bud is located at the naso-palatal fold, and the creation of the cleft at this level divides the bud in two, giving rise to an additional tooth.

An etiopathogenic similarity between the origin of the cleft and the appearance of a defect in mineralization of the adamantine matrix, by definition an amelar hypoplasia, is assumed [14].

Because of its clinical appearance, this oral dysmorphosis disrupts the functions of the orofacial sphere: breathing, swallowing, chewing, and phonation, and has psychosocial repercussions [11] [15].

Practical management is complex and may involve dental, maxillofacial, orthodontic plastic surgery, speech therapy and, of course, psychological services to improve patient's quality of life [16].

To diagnose and treat cleft lip and palate patients in a way that allows for effective management while taking into account their special dental and occlusal characteristics, it is critical to investigate the prevalence of these dental anoma-

lies in these patients.

The aim of this study is to assess the prevalence of dental anomalies in a Moroccan population of cleft lip and palate patients.

2. Materiel and Methods

Our study is a cross-sectional descriptive survey that took place at the Opération Smile Morocco center.

The survey targeted patients with cleft lip and palate attending the Opération Smile Morocco center in Casablanca.

The investigation lasted 2 months from the 10th of January 2022 to the 10th of March 2022. A total of 210 patients were examined and consequently included in this study.

2.1. Survey Support

The request form had four sections:

- First section:

It provides patients' socio-demographic variables (gender, age, socio-economic level).

- Second section:

It treats the type of cleft, either uni- or bilateral, total or partial, right or left.

- Third section:

This includes a full clinical examination to look for dental anomalies of number, shape, structure and position.

- Fourth section:

Radiographic examination of the patient, whether panoramic and or retroalveolar.

2.2. Source of Data

A total of 210 patients' files were examined.

Our study is based on data collected during:

- Interviews with cleft lip and palate patients and/or their parents.
- Clinical examination of the oral cavity.
- Radiological examination.

2.3. Study Parameters

Socio-demographic variables:

Sex, age, residence, socio-economic status (low, medium, high).

Clinical variables:

The cleft type:

It is a qualitative variable that provides information on the type of cleft and the structures affected by this malformation.

Based on the extent of the cleft, we can distinguish between a cleft lip, a cleft lip-alveolar, and a cleft lip-palate.

Depending on the side affected, a distinction is made between a right-sided unilateral cleft, a left-sided unilateral cleft, a symmetrical bilateral cleft, and an asymmetrical bilateral cleft.

Types of dental anomalies: It is a quantitative variable that provides information on the presence of: **Number anomalies**: Agenesia, Supernumerary tooth.

Shape anomalies: Microdontia, macrodontia, fusion, gemination, taurodontism, dens in dente, Coronary-radicular dilaceration.

Position anomalies: Ectopy, rotation, transposition, version.

Structural anomalies: Amelogenesis imperfecta, dentinogenesis imperfecta, hypoplasia, dyschromia.

X-ray variable:

This is a qualitative variable that indicates the type of radiograph taken.

It may be a retroalveolar and/or a panoramic.

2.4. The Interviewer

One investigator was in charge of this study.

2.5. Survey Procedure

The investigation took place in three phases:

1) Identification of the cleft patient through questioning and medical history.

2) Clinical examination of the entire oral cavity was carried out on cleft patients using disposable gloves, a probe and a mirror under a light source.

3) A radiological examination allowed us to confirm the data collected during the clinical examination.

The information was recorded on the questionnaire reserved for this purpose. Statistical analysis was performed using Epi info version 7.0 software.

The Chi^2 test was used to study correlations between the different variables.

A 5 per cent risk of error was adopted. A p-value less than 0.05 was considered significant.

3. Results

3.1. Sample Distribution by Gender, Age and Socio-Economic Level

The sample consisted of 210 patients, including 114 girls with an average age of 12.7 and 94 boys with an average age of 14.6.

Distribution by socio-economic level showed that 78 boys (37.14%) were in categories 0 to 4, corresponding to a low to medium socio-economic level. Whereas 18 (8.57%) were in categories 5 and 6, corresponding to significantly higher socio-economic status.

As for the girls, the distribution showed that 98 girls (46.66%) were in categories 0 to 4, corresponding to a low to medium socio-economic level, and only 16 girls (7.61%) were in categories 6 to 8, corresponding to high socio-economic level (Table 1).

Table 1. Sample distribution by gender, age and socio-economic level.

	Socio-economic level			
	Low	Mid	High	
	N	%	N	%
Sex				
Female	98	44.66	16	7.61
Male	78	37.14	18	8.57
Total	176	83.8	34	16.19
	Female	**Male**		
Age	12.7 years	14.6 years		
Total	114	96		

3.2. Distribution of the Sample According to Gender and Anatomical/Clinical Type of Cleft

Right unilateral clefts are the most frequent, with a rate of 37.61% (79 clefts), including 20% in girls and 17.61% in boys.

Bilateral clefts account for 72 (34.28%), of which 38 (18.09%) are in girls and 34 (16.19%) in boys.

Unilateral left-sided clefts accounted for 59 (28.09%), of which 34 (16.19%) were in girls and 25 (11.90%) in boys (Table 2).

Table 2. Distribution of the sample according to gender and anatomical/clinical type of cleft.

Cleft type	Male	Female	Total
	N (%)	N (%)	
Right unilateral cleft	37 (17.61%)	42 (20.00%)	79 (37.61%)
Left unilateral cleft	34 (16.19%)	38 (18.09%)	72 (34.28%)
Bilateral cleft	25 (11.90%)	34 (16.19%)	59 (28.09%)
Total (%)	96 (45.71%)	114 (54.28%)	210 (100%)

3.3. Frequency Distribution of Number Anomalies in Girls and Boys

The total number of dental agenesis in our sample was 173, with 94 among boys (54.33%) and 79 among girls (45.66%).

Statistical comparison showed no significant difference between boys and girls ($p = 0.242$).

Supernumerary teeth were rare, with a total of only 14 supernumerary teeth, including 8 supernumerary teeth (57.14%) among boys and 6 supernumerary teeth (42.85%) among girls.

Statistical comparison showed no significant difference between boys and girls ($p = 0.213$) (Table 3).

Table 3. Distribution of number anomalies among girls and boys.

	Male CLP	Female CLP	Total	p-value	Significance
	N (%)	N (%)			
Agenesis	94 (54.33%)	79 (45.66%)	173	0.242	NS
Supernumerary teeth	8 (57.14%)	6 (42.85%)	14	0.213	NS

3.4. Distribution of Agenesis According to Tooth Type in Girls and Boys

Teeth affected by agenesis were mainly permanent incisors (64.73%), with a statistically significant predominance for boys (p = 0.03).

However, girls showed a significant difference for premolar agenesis.

Permanent molars came third, accounting for 12.13% of the total number of agenese teeth, with a statistically significant predominance for girls (p = 0.04).

Permanent canines are rarely agenese. In our study, only 6 permanent canines were agenese, 3 in girls and 3 in boys.

Finally, we found agenesis of 11 temporary incisors, equally distributed according to gender (Table 4).

Table 4. Distribution of agenesis according to tooth type among girls and boys.

Tooth type	Male CLP	Female CLP	Total	p-value	Significance
	N (%)	N (%)			
Permanent incisors	67 (38.72%)	45 (26.01%)	112 (64.73)	0.03	S
Temporary incisors	5 (2.89%)	6 (3.46%)	11 (6.35)	0.83	NS
Permenanent canines	3 (1.73%)	3 (1.73%)	6 (3.46)	0.23	NS
Permanent molars	6 (3.46%)	15 (8.67%)	21 (12.13)	0.04	S
Premolars	13 (7.51%)	10 (5.78%)	23 (13.29)	0.12	NS

3.5. Distribution of Shape Anomalies by Gender

Regarding shape anomalies, microdontia is the most frequent. It affects 78 teeth, 34 in females (43.58%) and 44 in males (56.41%).

Macrodontia comes second with 15 teeth affected, 6 in girls (40%) and 9 in boys (60%).

Corono-radicular dilacerations were rare, with only 4 incisors affected, all in boys. Girls showed no coronal-radicular dilaceration.

Two permanent central incisors were affected by gemination, and only one boy's permanent lower incisor showed fusion (Table 5).

Table 5. Distribution according to shape anomalies by gender.

Anomaly type	Male	Female	Total	p-value	Significance
	N (%)	N (%)	N (%)		
Macrodontia	44 (56.41%)	34 (43.58%)	78	0.724	NS

Continued

Microdontia	9 (60.00%)	6 (40.00%)	15	0.467	NS
Gemination	1 (50.00%)	1 (50.00%)	2	0.654	NS
Coronary-radicular dilaceration	4 (100%)	0 (0.00%)	4	0.02	S
Fusion	1 (100%)	0 (0.00%)	1	0.04	S

3.6. Distribution of Shape Anomalies in Temporary and Permanent Dentition

We noted a total of 100 shape anomalies, with 17 anomalies in temporary dentition and 86 anomalies in permanent dentition.

The difference is statistically significant according to gender.

Indeed, boys are more prone to shape anomalies than girls in both dentitions (Table 6).

Table 6. Distribution of shape anomalies in temporary and permanent dentition.

Dentition type	Male CLP	Female CLP	Total	p-value	Significance
	N (%)	N (%)	N (%)		
Primary dentition	12 (12.00%)	5 (05.00%)	17 (17.00%)	0.02	S
Permanent dentition	50 (50.00%)	36 (36.00%)	86 (86.00%)	0.04	S

3.7. Distribution of Structural Anomalies According to Sex

Amelogenesis imperfecta was the most frequent structural anomaly in our sample, with a total of 166 teeth.

A statistically significant predominance in favor of boys was noted, with an amelogenesis rate of 58.44%.

36 teeth were affected by dentinogenesis imperfecta, with a statistically significant predominance in favor of boys.

30 teeth were affected by hypoplasia with or without loss of substance, with a non-statistically significant difference between boys and girls (see Table 7).

Table 7. Distribution of structural anomalies according to gender.

Anomaly	Male	Female	Total	p-value	Significance
	N (%)	N (%)			N (%)
Amelogenesis imperfecta	97 (58.43%)	69 (41.56%)	166	0.01	S
Dentinogenesis imperfecta	22 (61.11%)	14 (38.887%)	36	0.03	S
Hypoplasia without substance loss	8 (44.44%)	10 (55.56%)	18	0.32	NS
Hypoplasia with substance loss	5 (41.66%)	7 (58.33%)	12	0.05	NS

3.8. Distribution of Structural Anomalies According to Tooth Type

Table 8 shows the distribution of structural anomalies in temporary and permanent dentition in girls and boys.

Table 8. Distribution of structural anomalies in temporary and permanent dentition in girls and boys.

Dentition type	Amelogenesis imperfecta		Dentinogenesis imperfecta		Hypoplasia with substance loss		Hypoplasia without substance loss	
	Male	Female	Male	Female	Male	Female	Male	Female
Primary dentition	32 (19.27%)	26 (15.66%)	9 (25%)	5 (13.88%)	2 (11.11%)	2 (11.11%)	0 (0.00%)	0 (0.00%)
Permanent dentition	65 (39.15%)	43 (25.9%)	13 (36.11%)	9 (25%)	6 (33.33%)	8 (22.22%)	5 (41.66%)	7 (58.33%)

3.9. Distribution of Position Anomalies by Gender

Concerning positional anomalies, we noted that ectopia was the most frequent anomaly, with a total of 501 ectopias.

A total of 369 teeth were tipped, accounting for 47.69% in males and 52.30% in females. We noted 18 transpositions (6 in boys and 12 in girls).

Statistical comparison of positional anomalies between boys and girls showed a non-significant difference (Table 9).

Table 9. Distribution of positional anomalies by gender.

Anomaly type	Male	Female	Total	p-value	Significance
	N (%)	N (%)	N (%)		
Ectopia	234 (46.70%)	267 (53.29%)	501	1.423	NS
Rotation	253 (56.09%)	198 (43.90%)	451	0.745	NS
Version	176 (47.69%)	193 (52.30%)	369	0.386	NS
Transposition	6 (33.33%)	12 (66.66%)	18	0.237	NS

4. Discussion

The present study is a descriptive cross-sectional study designed to assess the prevalence of dental anomalies in patients with cleft lip and palate.

Diagnosis of anomalies was based on direct clinical examination and radiological examination using panoramic radiography and, if necessary, retro-alveolar radiography on the involved tooth.

In all, 114 girls and 94 boys were examined among the 210 participants. The age range of the patients was 6 to 14 years, with a majority of 176 patients of low socio-economic status.

The precarious economic situation and non-proximity of dental care centers could explain the delayed diagnosis and non-early management of these patients, which justifies the average age of 10.9 years.

A repetitive pattern of right unilateral clefts was detected (37.61%), followed by bilateral clefts (34.28%) and left unilateral clefts (28.09%).

These data are in line with the analysis of Namdar *et al.* who observed that unilateral clefts are more frequent than bilateral clefts, but with a higher prevalence of left unilateral clefts in the Iranian Sari population [17].

This is in agreement with a study conducted by Sahim *et al.* in 2021 on a moroccan sample which also concluded a frequent occurrence of left unilateral clefts [18].

In contrast to our study, where a higher incidence of right unilateral clefts was found.

In our study, a non-significant predominance of females was found, with a rate of 54.28%. This is in agreement with a study carried out in Madagascar and Sudan, which reported a female predominance for no apparent reason [19] [20].

It has been reported that 96.7% of patients affected by this condition are more likely to develop at least one dental anomaly [19] [21]. With no distinction between male and female, both can be affected [17] [22].

Shape anomalies are the most frequent, followed by number and position anomalies [9].

In contrast to our research, positional anomalies are the most frequent, followed by number anomalies, then structural anomalies and finally shape anomalies.

The results of our study noted a high incidence of agenesis with a rate of (82.39%), which is in agreement with the data in the literature [8] [9] [10] [12] [16].

A study carried out in Pakistan in 2021 demonstrated an increased risk of dental anomalies in the presence of cleft lip and palate, notably agenesis, with the most affected tooth being the premolar followed by the lateral incisors [23].

At the same time, Schwartz *et al.* demonstrated a high incidence of number anomalies affecting maxillary second premolars in first place, followed by maxillary lateral incisors and mandibular second premolars without distinction between genders [24].

This was not the case in our study, where the teeth most affected with agenesis in descending order were the permanent incisors, followed by the premolars and then the molars, with a predominance of agenesis of the permanent incisors noted in boys (38.78%).

Canine agenesis was rare, with only 6 canines found, divided equally between the genders, 3 in girls and 3 in boys. Similarly, a study conducted at CLAPP (cleft lip and palate association of Pakistan) recorded only 2 canine agenesis out of 100 patients [23].

Agenesis mainly affects the permanent dentition. In our study, we quantified 11 missing temporary incisors.

According to Namdar *et al.*, supernumerary teeth are more common in patients with unilateral than bilateral clefts, with a slight prevalence in girls [17]. This is in agreement with Ajami *et al.*'s report [25].

In our sample, we found 14 (6.66%) supernumerary teeth, with 57.14% in boys and 42.85% in girls. A low rate (1.25%) was found by Sahim *et al.* in another study also [18].

Contrary to one study on german population in which we found a significantly high percentage of 33% [26].

Concerning shape anomalies, the most affected tooth is the lateral incisor, with a riziform shape [5]. Sahim *et al.* found that microdontia was the most frequent anomaly, followed by macrodontia [18].

According to our results and in order of frequency, microdontia comes first, macrodontia second, and then corono-radicular dilaceration, gemination and fusion are rarer.

According to Al-Kharboush *et al.*, microdontia is the second most common anomaly, with a rate of 45.6% higher than the 37.14% found in our patients [22].

For macrodontia, an identical rate has been calculated, with 60% in boys and 40% in girls for the Moroccan and Iranian populations [17].

In line with the same study, no significant difference in the distribution of dental anomalies between males and females was detected [17].

This finding is consistent with the literature demonstrating that the type of dental anomalies in cleft patients is not gender-specific, but is widespread across both sexes [16] [21] [22] [27].

This contradicts our study. We noted a statistically significant difference between the two genders, which means that males are more prone to shape anomalies than females, in both temporary and permanent dentition.

In terms of structural anomalies, amelogenesis imperfecta was the most common, followed by dentinogenesis imperfecta than hypoplasia with loss of substance.

According to our data, males are more predisposed to structural anomalies, in particular amelogenesis imperfecta with a rate of 58.43% and dentinogenesis imperfecta with a rate of 61.11%. Both dentitions were affected, but the permanent teeth were the most affected.

However, Sá *et al.* recorded 18.9% of amelar hypoplasia, with a slight predisposition in patients with cleft lip (21.6%) compared with patients with unilateral and bilateral cleft lip and palate, with a rate of 16.7% and 17.7% respectively [11].

In the same line, Rakotoarison *et al.* claim that enamel dysplasia is common and that all teeth are susceptible to structural anomalies, especially incisors (22.22%) followed by canines (11.11%) [20].

The evidence in the literature is consistent with the dominance of positional anomalies in patients with cleft lip and palate [16] [20].

Our statistical analysis revealed a considerable incidence of dental ectopia, followed by rotation, version and, lastly, a lower rate of transposition.

Similar to our finding, Amady *et al.* found a predominance of ectopias followed by dental rotations [28].

According to Fox *et al.*, the average population with ectopic teeth is between 2% and 6% for maxillary first molars and between 1.5% and 2% for permanent canines [29].

This conflict in results can be explained by the genetic and racial differences specific to each population.

We noted 56.09% in males and 43.90% in females of tooth rotation, a higher rate in our population than in the Iranian population, of which 36.6% in males and 63.3% in females, according to the same study [17].

According to Meazzini *et al.*, tooth rotation may be a consequence of cleft repair surgery in early childhood. At this stage, gingivoplasty would reduce the anterior alveolar space and thus limit the space available for tooth alignment [30].

We intercepted a total of 369 dental versions in our patients. No studies evaluating dental versions in cleft lip and palate patients were found.

According to Tortora *et al.*, the notion of a dental version is rarely mentioned, as it is a false version due to the lack of space incurred by the tooth during eruption [31].

According to Chateau, lack of balance in the tooth corridor leads to version [32].

A total of 18 transpositions were noted, 6 in males and 12 in females. Like dental versions, this malposition is usually related to lack of space and insufficient development of the alveolar processes at the cleft [20].

There was no difference in the distribution of positional anomalies between males and females.

5. Conclusions

Patients with cleft lip and palate have a high prevalence of dental anomalies.

Dental ectopia is the most frequent anomaly in our study.

Agenesis of the permanent incisor comes second, followed by amelogenesis imperfecta and microdontia.

Supernumerary teeth, coronal-root dilaceration, gemination, fusion, transposition and hypoplasia are very rare in our sample.

Dental anomalies can have repercussions on the functional, aesthetic and psychological aspects of cleft lip and palate patients, hence rigorous early management is very important.

In order to provide the best odontological care for patients with cleft lip and palate, further research in this field is required as the results obtained cannot be applied to the entire population of patients with this condition.

This work highlights the importance of multidisciplinary collaboration for optimal dental, occlusal and aesthetic rehabilitation of cleft lip and palate patients, all of this to improve the quality of life of these patients.

Conflicts of Interest

The authors declare no conflicts of interest.

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