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INFLUENCE DU MAINTIEN OU DE LA FERMETURE DE L'ESPACE DE L'INCISIVE LATÉRALE MAXILLAIRE SUR L'OCCLUSION ET LA SYMÉTRIE DE L'ARCADE DENTAIRE CHEZ LES PATIENTS ATTEINTS DE FENTE DU PALAIS PRIMAIRE : ÉTUDE RÉTROSPECTIVE

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# ABREVIATIONS

MLI	Maxillary Lateral Incisor
CL	Cleft Lip
CLA	Cleft Lip and Alveolus
CLP	Cleft Lip and Palate
OGC	Orthodontical Gap Closure
PR	Prosthodontic Replacement
UMMLI	Unilateral Missing Maxillary Lateral Incisor
BMMLI	Bilateral Missing Maxillary Lateral Incisor
UIIP	Upper Interincisal Point
TOBBI	Temporary Orthodontic Bonding Bridge for Implant
UC	Unilateral Cleft
BC	Bilateral Cleft

# ABSTRACT

**Introduction:** Maxillary lateral incisors (MLI) are frequently missing in cleft patients. The aim of this study was to assess how maintaining or closing orthodontically the missing MLI gap affected the occlusion and symmetry at the end of treatment. We also aimed to determine the impact of the level of oral hygiene on the decision made to manage the missing MLI gap.

**Methods:** A descriptive, retrospective cohort study was led in Nantes Cleft Centre including patients born between 1980 and 1999. Patients presenting unilateral (UMMLI) or bilateral (BMMLI) missing MLI were reviewed. Data about management of the missing MLI gap, occlusion, symmetry and level of oral hygiene were collected.

**Results:** Four hundred and eighty-two patients were reviewed, including 212 patients with UMMLI or BMMLI. When compared to orthodontic gap closure, the orthodontic opening of the MLI gap was associated with a better final occlusion (59.8% vs. 10.3%, p=0.000003) and better dental arch symmetry (88.1% vs. 44.0%; p=0.000003) for UMMLI cases but not for BMMLI cases. A better level of oral hygiene was associated with more cases with orthodontic opening of the MLI gap for UMMLI cases (p=0.03) and had no impact for BMMLI cases.

**Conclusion:** Maintaining the missing MLI space provided better functional and aesthetic results for cleft patients presenting with UMMLI.

# **KEYWORDS**

Cleft Lip and Palate; Maxillary lateral incisor; Orthodontics; Dental occlusion; Smile aesthetics; Oral hygiene

#### INTRODUCTION

Cleft lip and/or palate are the most common congenital orofacial deformities with a prevalence rate of 1 for 1000 births in Europe (1). Patients with cleft lip and/or palate need medical care as newborns, throughout their childhood and adolescence, and until early adulthood. Management of cleft lip and/or palate requires a good coordination between the multiple surgical steps and the orthodontic treatment.

Several dental anomalies are often associated with cleft lip and/or/palate (2). Maxillary lateral incisor (MLI) agenesis or hypoplasia are the most frequent for patients with cleft lip (CL), cleft lip and alveolus (CLA), and cleft lip and palate (CLP) (3,4,5,6,7,8). As a consequence, MLI are often missing due to agenesis or extraction and the orthodontist and surgeon discuss the best option to manage the MLI gap.

The first treatment option is to close the MLI gap orthodontically. With this treatment, the homolateral canine is brought mesially, adjacent to the central incisor. It re-establishes the dental arch continuity and no further prosthodontic treatment is needed. Thus, it reduces the lifetime maintenance and cost for the patient (9). However, this can lead to a hypodevelopment of the maxilla in both transversal and anteroposterior dimensions, with the need of future orthognathic surgery (10,11). It can also create ventilatory disorders by reducing the inter-canine distance and so, limiting the size of the anterior nasal apertures (12). In addition, it can have aesthetic repercussions by creating an asymmetrical dental arch and by moving the upper interincisal point (UIIP) from the midline (13).

Another treatment option, which aims to avoid these complications, is opening orthodontically the gap between the central incisor and the canine. It gives better functional and aesthetic results with a better projection of the upper lip and a symmetrical dental arch (13). This option implies a longer orthodontic treatment and requires a prosthodontic solution. To do so, different solutions are available such as dental implant, fixed partial denture or removable partial denture. Dental implant is the most conservative option, with a good survival rate (14,15) and satisfactory results in terms of quality of life (16,17,18). The main disadvantages are that they can only be placed once the facial growth is completed and often require additional bone graft (15). Several options for fixed dentures are available such as resin-bonded fixed partial denture, cantilever fixed partial denture and conventional full-coverage fixed

partial denture (19). Removable denture is the cheapest option and also acts as a retainer to maintain maxillary transverse dimension (13). However, young patients are usually not satisfied with removable options.

The criteria used to decide for closing or opening the MLI gap are unclear in the literature. This decision seems to be made for each individual, based on occlusion, aesthetics or level of oral hygiene. Only two studies compared the aesthetic results obtain with orthodontic gap closure (OGC) and prosthodontic replacement (PR) for cleft and non-cleft patients (20,21). Patients benefiting from OGC were overall more satisfied with their aesthetics than patients benefiting from PR but these studies didn't evaluate separately the patients with UMMLI or BMMLI cases, which can be a problem as the later don't have symmetry issues.

The aim of this study was to assess whether maintaining or closing the MLI gap affected the dental arches relationship at the end of treatment for CL, CLA and CLP patients with missing MLI at Nantes tertiary referral cleft centre. Secondly, we aimed to determine if the management of the MLI gap had an influence on dental arch symmetry and whether or not the level of oral hygiene impacted the decision made to manage the MLI gap.

#### METHODS

This was a single centre, descriptive, retrospective cohort study. In this retrospective study, no change to the current clinical practice or randomization was performed. No ethics committee approval was required for the use of the data in this epidemiologic study, as per French legislation article L. 1121-1 paragraph 1 and R1121-2 of the Public Health Code.

#### **Participants**

Patients born between 1980 and 1999 who had surgery at Nantes Cleft Centre for CL, CLA or CLP were included. Patients with median cleft were excluded. Data, including demographic information, cleft type and laterality were collected from medical records. Medical records with missing information about the cleft were excluded.

#### Management of the MLI gap

Data about patients presenting missing MLI were collected. Patients with missing central incisor or canine adjacent to the cleft were excluded.

• Aetiology of the absence of MLI

MLI anomalies were categorised as "congenitally missing" if the MLI was missing and no information suggested that it existed, or as "anomaly of size and/or shape".

#### Orthodontic management

Patients were categorised in two groups based on if the MLI gap was orthodontically opened or closed. Cases with a MLI gap incompletely opened and narrower than the crown of an MLI with no further radiographs available were classified as "missing information".

# Prosthodontic management

Patients with a MLI gap opened orthodontically were categorised in one of the following, based on the prosthodontic solution they benefited from: removable partial denture, dental implant, tooth-supported restauration, Temporary Orthodontic Bonding Bridge for Implant (TOBBI), or no prosthodontic rehabilitation.

# Impact on dental occlusion

The main outcome of the study was to evaluate the influence of the orthodontic closure or opening of the MLI gap on dental arches relationship at the end of treatment.

# Photographs

Dental occlusion was evaluated on the most recent photograph in the medical record by a single practitioner. If the orthodontic treatment was not completed, the patient was classified as "missing information". Photographs were taken by a professional photographer and were standardised including extra-oral and intra-oral photographs with frontal and lateral views.

# • Evaluation of dental arches relationship

Dental arches relationship was evaluated regarding the following criteria: Angle class dental relationship, anterior or lateral open bite, edge to edge incisors relationship, over bite and anterior or lateral cross bite. Patients were considered with no malocclusion when they presented with bilateral Angle class I for molar and canine relationship, no anterior or posterior open bite, no edge to edge position of the incisors, no over bite and no anterior or lateral cross occlusion.

# Secondary outcomes

Position of the interincisal point

We then evaluated which management solution of the MLI gap had the greater aesthetic impact by measuring the distance between the upper interincisal point (UIIP) from the facial midline for the two groups. This measure was done by two practitioners on standardised smiling photographs of patients after the orthodontic treatment was achieved. For divergent results, photographs were re-evaluated by both practitioners and a joint decision was made.

# • Oral Hygiene

Further, we assessed if oral hygiene could influence the decision to open or close the MLI gap. Oral hygiene was assessed by a single practitioner and based on the most recent photograph available. In the absence of recent photograph, the evaluation was based on medical records. Oral hygiene was subjectively categorised from "good" for patients with no dental plaque and no sign of gingivitis, "intermediate" if there was a small of dental plaque, calculus or mild gingivitis, to "poor" in the presence of an important amount of dental plaque, moderate to severe gingivitis, multiple dental decays or tooth loss.

# • Influence of the surgical team

We finally evaluated if having any surgical step of the cleft repair performed in another centre influenced the decision on the closure or the opening of the MLI gap.

# Statistical analysis

Statistical analysis was performed using GraphPad Prism 5.0 for Mac (GraphPad Software, La Jolla, CA, USA). Quantitative data were analysed using a chi-squared test. A p-value of less than 0.05 (p < 0.05) indicated statistical significance.

## RESULTS

## Participants

Four hundred and eighty-six patients were included in this study. Four patients were excluded: two because of median cleft and two because of missing information about the cleft type. Demographic data and cleft details are presented in Table 1. Syndromic patients are presented in Table 2.

Table 1: Demographic data and cleft details

	Bilateral cleft	Right side	Left side	Male	Female	Total	Syndromes
CL	7 (12.1%)	20 (34.5%)	31 (53.4%)	36 (62.1%)	22 (37.9%)	58 (12.0%)	1 (1.7%)
CLA	11 (13.4%)	22 (26.8%)	49 (59.8%)	42 (51.2%)	40 (48.8%)	82 (24.0%)	2 (2.4%)
CLP	109 (31.9%)	88 (25.7%)	145 (42.4%)	241 (70.5%)	101 (29.5%)	342 (70.5%)	25 (7.3%)
Total	127 (26.3%)	130 (27%)	225 (46.7%)	319 (66.2%)	163 (33.8%)	482 (100%)	28 (5.8%)

CL: Cleft Lip, CLA: Cleft Lip and Alveolus, CLP: Cleft Lip and Palate

Syndrom Number of patients		Characteristics of the cleft			
Optiz	9	6 bilateral CLP, 1 right CLP, 1 left CLP, 1 left CLA			
Kallman Morsier	1	Bilateral CLP			
Branchio-oculo-facial	1	Right CLP			
Branchio-oto-facial	1	Bilateral CLP			
Van der Woude	4	1 bilateral CLP, 2 right CLP, 1 left CLP			
Fragile X	1	Bilateral CLP			
Amniotic Band Syndrom	1	Right CLP			
CHARGE	1	Right CLP			
Fetal alcohol	3	1 right CLP, 2 left CLP			
Goldenhar	2	1 bilateral CLP, 1 right CLP			
Gorlin	1	Right CL			
	3	1 bilateral CLP, 1 left CLP, 1 left CLA			

CL: Cleft Lip, CLA: Cleft Lip and Alveolus, CLP: Cleft Lip and Palate

#### Anomalies of the MLI

Among the 482 included patients, 344 (71.4%) had an anomaly of at least one MLI. Details of data are presented in Figures 1 and 2. 42.1% of patients with unilateral cleft (UC) presented with an agenesis of at least one MLI. Among bilateral cleft (BC) patients, 55.8% had an agenesis of at least one MLI.

Information about MLI anomalies were missing for 19 patients (12 with one missing MLI and 7 with two missing MLI).

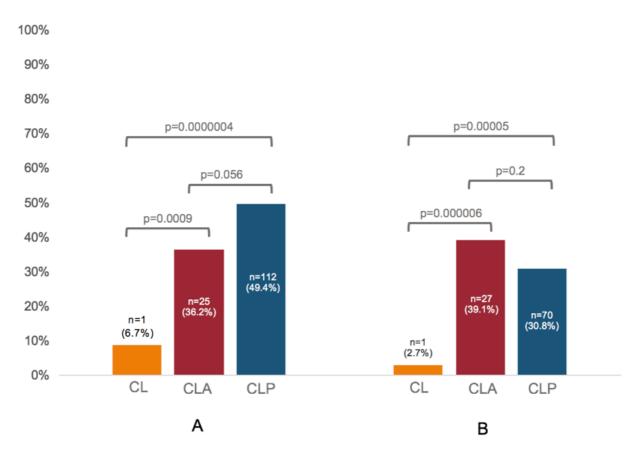


Figure 1: MLI anomalies on patients presenting with UC. A.  $\ge$  1 congenitally missing MLI. B.  $\ge$  1 anomaly of size and/or shape of MLI

CL: Cleft Lip, CLA: Cleft Lip and Alveolus, CLP: Cleft Lip and Palate

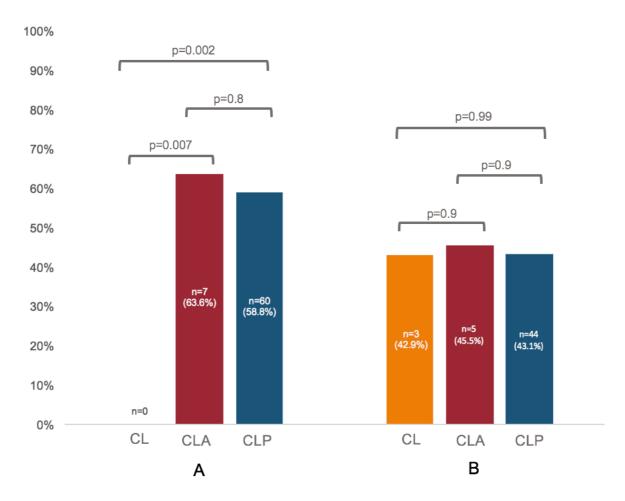
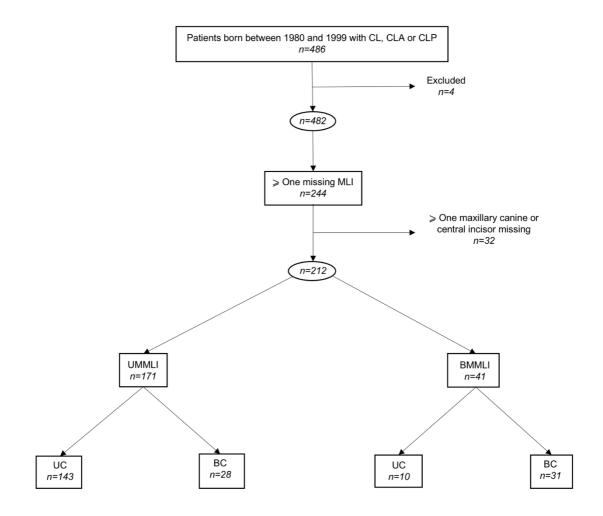


Figure. 2: MLI anomalies on patients presenting with BC. A.  $\ge$  1 congenitally missing MLI. B.  $\ge$  1 anomaly of size and/or shape of MLI

CL: Cleft Lip, CLA: Cleft Lip and Alveolus, CLP: Cleft Lip and Palate

Among the 482 patients, 244 had at least one missing MLI. Thirty-two patients also had a missing central incisor or canine homolateral to the cleft and were excluded, leaving 212 patients for analysis (Figure 3).

Figure 3: Flow-chart

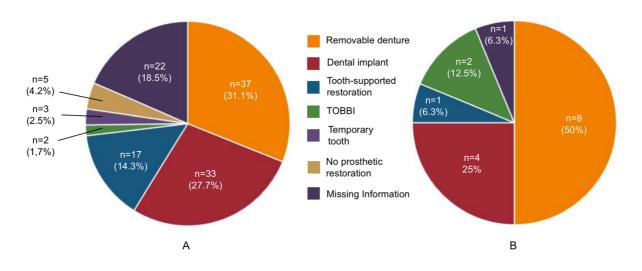


CL: Cleft Lip, CLA: Cleft Lip and Alveolus, CLP: Cleft Lip and Palate, MLI: Maxillary Lateral Incisor, UMMLI: Unilateral Missing Maxillary Lateral Incisor, UC: Unilateral Cleft, BC: Bilateral Cleft

#### Orthodontic management of the MLI gap

Among patients presenting at least one missing MLI, 139 benefited from MLI gap opening (71.6%), including 119 patients with UMMLI (77.3%) and 20 patients with BMMLI (52.6%). Information was missing for 20 patients (17 with UMMLI and 3 with BMMLI). Prosthodontic solutions are presented in Figure 4.

Fig 4: Prosthodontic restoration. A. Patients presenting UMMLI. B. Patients presenting BMMLI



TOBBI: Temporary Orthodontic Bonding Bridge for Implant

#### Dental arches relationship and UIIP shift from facial midline

In patients with UMMLI, a satisfying dental arches relationship was found in 55 patients (59.8%) with PR, and in 3 patients (10.3%) with OGC (p=0.000003). Information was missing for 33 patients. The UIIP was on the facial midline for 85 patients (78.0%) including 74 patients (88.1%) with PR and 11 patients (44.0%) with OGC (p=0.000003).

The UIIP was shifted toward the cleft side (or toward the missing MLI for subjects with BC with UMMLI) for 92.9% of patients benefiting from OGC versus 40% for those benefiting from PR (p=0.005). Information was missing for 45 patients.

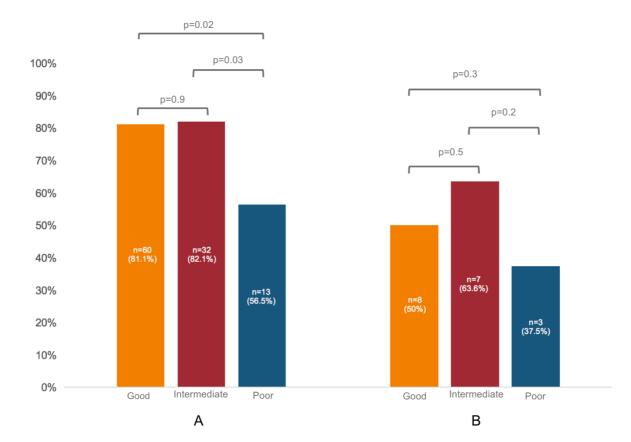
In patients with BMMLI, a satisfying dental arches relationship was found in 9 patients (47.4%) with PR and in 6 patients (42.9%) with OGC (p=0.8). Information was missing for 5 patients. The UIIP was on the facial midline for 17 patients

(56.7%), including 10 patients (58.8%) with PR and 7 patients (53.8%) with OGC (p=0.8). Information was missing for 8 patients.

# Oral hygiene

The results about the influence of oral hygiene on the rate of PR and final dental arch relationships are presented in Figures 5 and 6. Information about level of oral hygiene was missing for 22 patients (18 with UMMLI and 4 with BMMLI).

Figure 5: Rate of PR regarding oral hygiene. A. Patients presenting UMMLI. B. Patients presenting BMMLI



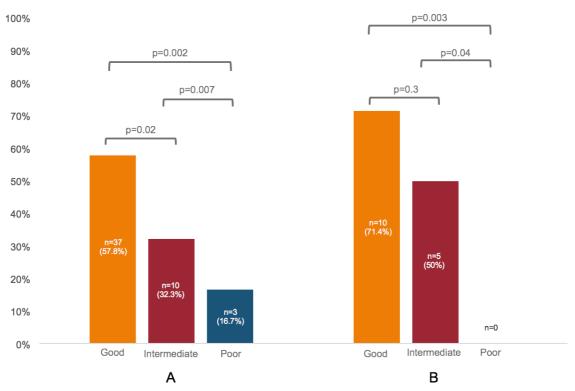


Fig 6: Rate of satisfactory dental arch relationship regarding oral hygiene. A. Patients presenting UMMLI. B. Patients presenting BMMLI

## Influence of the surgical team

Patients with UMMLI whose medical follow-up was performed in Nantes Cleft Centre from birth benefited statistically significantly more from PR (82.5%) than the ones treated in another centre for any surgical step (66.7%) (p=0.03). For patients with BMMLI, the MLI gap opening rate was of 57.1% for patients treated at Nantes Cleft Centre exclusively and 40% for the others (p=0.35).

#### DISCUSSION

This study described the management of the MLI gap in cleft patients with missing MLI treated at Nantes Cleft Centre between 1980 and 1999. It aimed to compare the results obtained with OGC or opening by evaluating the final dental arches relationship and symmetry. In addition, we tried to identify factors that could have influenced the decision of the best option for each patient.

In this cohort, 41.2% and 55.8% of patients with UC and BC respectively presented at least one MLI agenesis. In addition, the prevalence of agenesis was correlated with the severity of the cleft. This was comparative with previous result reported for both UC and BC (3,7,8,22,23). Demographic data and cleft characteristics of our cohort were similar to previous studies, confirming the fact that our cohort was representative.

## Orthodontic decision: Missing MLI gap closure vs. opening

In the present study, 71.6% of patients benefited from MLI gap opening (77.3% with UMMLI and 52.6% with BMMLI). These are higher than the rates previously reported in the literature for different populations: 20% to 35.1% for unilateral cases (3,9) and 37 % for bilateral cases (24). Lee et al. reported a rate of 58.5% of missing MLI gap opening, including cases of both UMMLI and BMMLI (10).

The high rate of missing MLI gap opening in this cohort is due to the intuition that it allows better functional and aesthetic outcomes. The local protocol includes a maxillary enlargement at the age of 4 to correct the collapses of the lateral maxilla which maintains the MLI gap opened at an early stage. This explains why patients with UMMLI treated exclusively within our centre benefited more of gap opening (81.7%) than other cases (66.7%). As suggested before, the socioeconomic status could play a role in the decision for low-income patients leaving far from our cleft unit, as it requires to attend multiple appointments.

#### Prosthodontic solution for missing MLI gap opening

The results showed that the majority of the patients benefited from a removable partial denture. This denture was specifically designed and reinforced to prevent the maxillary arch from collapsing. This option is easy and associated with minimal cost.

Dental implants are a good option but are associated with higher expenses, a frequent need for additional bone graft and also the need to maintain a maxillary contention (13,15,25). In addition, lower level of satisfaction on aesthetic outcomes were previously reported by cleft patients (17). If dental implants showed a good survival rate for cleft patients (14,15), few studies explored other important criteria such as absence of implant mobility or peri-implant radiolucency, no bone loss or asymptomatic function of the implant following restoration and occlusal loading (17,26,27,28). Salvi et al. also showed periodontal attachment loss adjacent to the cleft for patients not enrolled in a supportive periodontal therapy program (29). The lower rate of dental implant for BC patients can be explained by the fact that they showed more periodontal diseases in our experience, and this technique tended to be avoided for this population.

# Impact of the management of missing MLI gap on dental arches relationship

This is the first study showing the consequences of OGC or opening for the MLI gap on dental arches relationship for cleft and non-cleft patients. In our cohort, patients with UMMLI who had MLI gap opening had statistically less malocclusion than patients with OGC (p=0.000001). This result shows that MLI gap opening should be privileged over OGC for cleft patients to improve final occlusion. In addition, some authors reported the long term need for additional orthodontic treatment or orthognathic surgery for patients who benefited from OGC for the MLI gap (10,11). This occurs at a late stage, where patients are less inclined for extra procedures, regarding the multiple treatments they previously had.

Similarly, no previous study reported final occlusal results for cleft patients with BMMLI. In this cohort, final dental arches relationship tended to be better if the MLI gap was opened with not statistical significance. In a non-cleft population with a majority of BMMLI, Robertsson et al. observed small differences in the occlusion between OGC and opening (21).

It would have been interesting to explore the influence of the initial dental relationships on the decision made to manage the MLI gap. However, this information was not available in medical records. According to Kokich and Kinzer, OGC by bringing the canine mesially is only a suitable solution for patients presenting with Class II malocclusion with no crowding at the mandibular arch and Class I patients with important mandibular arch crowding requiring dental extractions (30).

#### Impact of the management of missing MLI gap on aesthetic outcomes

Regarding the symmetry of the maxillary dental arch, it was impossible to measure precisely the shifting of the UIIP as there was no scale on the photographs used. Nonetheless, Kokich et al. explained how dental aesthetics and the perception of symmetry are highly variable between lay people, general dentists and orthodontists (31). Moreover, the asymmetry of the nose and the upper lip for cleft patients can influence the perception of the facial midline.

Nevertheless, this is the first study evaluating the symmetry and the aesthetic results based on the orthodontic decision of OGC or opening of the MLI gap for cleft patients with UMMLI. It shows much better outcome for patients who benefited from MLI gap opening, which could impact the decision on orthodontic strategy. This could be explained by the fact that when applying orthodontic forces to close the MLI gap, posterior teeth won't be as mobile as anterior teeth and anterior teeth will be attracted distally (10). We also found that for patients benefiting from orthodontic gap opening, the UIIP was more often shifted towards the opposite side of the cleft than to the cleft side. This may lead to non-aesthetic results with the need of further orthodontic treatment or surgery.

In patients with BMMLI, aesthetics results tended to be better when the gap was orthodontically opened with no statistically significant difference. This was expected as there are less concerns about dental arch symmetry for BMMLI cases than for UMMLI cases. These results were comparative with previously published study comparing the impact of OGC or opening for a small sample of patients with BCLP (24).

#### Impact of oral hygiene on the orthodontic decision

When looking for factors that could have influenced the orthodontic management of the missing MLI gap, the level of oral hygiene was found statistically significant. OGC was privileged for patients with bad level of oral hygiene. This can be easily explained by the need for orthodontists to adopt a shorter strategy for these patients. As the canine on the cleft side tends to migrates spontaneously into the MLI gap during facial growth, OGC is an easier solution than trying to move the canine distally and maintain the MLI gap opened. However, this easier and shorter strategy seems to lead to worst final occlusal outcome.

This shows the importance for dental practitioners to be involved in cleft care to improve the level of oral hygiene and so, the occlusal results. This was further confirmed by Keribin et al. who showed the benefits of strict periodontal care and hygiene protocol on the overall treatment for cleft patients (32).

Different studies led on several communities suggested than the level of oral hygiene was correlated to a low socioeconomic status (33,34,35). Therefore, oral health education should be globally reinforced, especially for such patients.

However, in this study, the assessment of oral hygiene was done subjectively and retrospectively on the most recent photograph available, without following international guidelines. A better assessment method used in a prospective study would be needed to confirm our findings.

# CONCLUSION

This study is the first to show that orthodontic opening of the missing MLI gap provides better long-term occlusal and aesthetic outcomes for cleft patients, with different prosthodontic solutions available that can suit each individual. The level of oral hygiene appears to be one of the main criteria in the therapeutic decision.

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### INFLUENCE DU MAINTIEN OU DE LA FERMETURE DE L'ESPACE DE L'INCISIVE LATÉRALE MAXILLAIRE SUR L'OCCLUSION ET LA SYMÉTRIE DE L'ARCADE DENTAIRE CHEZ LES PATIENTS ATTEINTS DE FENTE DU PALAIS PRIMAIRE : ÉTUDE RÉTROSPECTIVE

# RÉSUMÉ

**Introduction :** Les incisives latérales maxillaires (ILM) sont fréquemment absentes chez les patients atteints de fente. L'objectif de cette étude était d'une part d'évaluer dans quelle mesure le maintien ou la fermeture orthodontique de l'espace de l'ILM pouvait influencer l'occlusion et la symétrie de l'arcade dentaire en fin de traitement et d'autre part de déterminer si le niveau d'hygiène orale pouvait influencer la décision prise quant à la gestion de cet espace.

**Méthode** : Il s'agit d'une étude de cohorte rétrospective réalisée au CHU de Nantes sur des patients nés entre 1980 et 1999. Les patients présentant une ou deux incisives latérales maxillaires (ILM) absente(s) ont été analysés. Les données concernant la gestion de l'espace de l'ILM, l'occlusion, la symétrie de l'arcade dentaire et le niveau d'hygiène orale ont été recueillies.

**Résultats** : Quatre cent quatre-vingt-deux patients ont été analysés, dont 218 présentant une ou deux ILM absente(s). Le maintien de l'espace de l'ILM était associé à une meilleure occlusion (59,8% vs. 10,3% ; p=0,000003) et à une meilleure symétrie de l'arcade dentaire (88,1% vs. 44% ; p=0,000003) en fin de traitement pour les patients présentant une ILM absente mais pas pour ceux présentant deux ILM absentes. Un meilleur niveau d'hygiène orale était associé à un taux de décision de maintien de l'espace plus important chez les patients présentant une ILM absente (p=0,03) mais pas chez ceux présentant deux ILM absentes.

**Conclusion** : Maintenir l'espace de l'ILM permet d'obtenir de meilleurs résultats fonctionnels et esthétiques chez les patients présentant une ILM absente.

#### **MOTS-CLES**

Fente labio-palatine ; Incisive latérale maxillaire ; Orthodontie ; Occlusion ; Esthétique du sourire ; Hygiène orale