



REVIEW ARTICLE

Are there any solutions for improving the cleft area hygiene in patients with cleft lip and palate? A systematic review

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Abstract

Objective: Children with cleft lip and palate (CLP) present high incidence of oral health problems, namely those associated with the accumulation of dental plaque. The objective of this systematic review was to verify if there is any solution to improve the hygiene of the cleft area.

Materials and methods: A structured systematic review was performed based on articles published in several electronic databases: PubMed, Web of Science, Cochrane Library and Scopus in the last 20 years. The *MeSH* terms used were cleft lip, palate, harelip, oral hygiene and toothbrush.

Results: From 270 potentially relevant articles, 39 articles were selected, corresponding to a total of 3226 CLP patients and 914 matched controls. The selected studies exhibited great heterogeneity regarding the type of the study, study population (average range 0–49 years old; sampling sizes 15–400 CLP patients), evaluation periods, reported variables and oral hygiene routines. Consensual outcomes were the presence of high plaque indexes, high prevalence of dental caries and worse oral hygiene patterns in CLP patients. Additionally, poor education in oral health and low motivation to integrate regular hygiene routines in the daily family life were also evident.

Conclusion: There is a clear need to improve the oral hygiene care of CLP children, but few studies were focused on specific preventive approaches. Development of devices especially designed to the hygiene of the cleft area and implementation of standardized prevention and control programmes targeting education, motivation and compliance would contribute to improve oral health in CLP children.

KEYWORDS

cleft lip palate, dental caries, dental hygiene, DMF-S/T, harelip, oral hygiene, toothbrush

1 | INTRODUCTION

Cleft lip and palate (CLP) are the most common congenital malformations of the face. Clinically, the anatomic-functional alteration of one or many structures (lip, alveolar process, hard and soft palate) is

observed. The cleft lip (CL), or cleft palate (CP), can occur alone or together, and might be complete or incomplete, uni or bilateral.

The anatomical and morphological development disturbances in children with CLP are associated with high incidence of oral health problems, namely those related to the accumulation of dental

TABLE 1 Studies that report the bacterial plaque indexes (PI), presented in descending chronological order

Reference, year of publication, period	Country & sample size	Adults/children	Study type (ST) and aim	PI	Conclusion
¹⁷ , 2017, July to Nov 2015	Poland, 15	Children: 6-18y	ST: cross-sectional Aim: periodontal status, mucogingival parameters and OH in growing patients with BCLP.	O'Leary PI CI=49.5%; LI=38.5%; C = 43.5%; 1stPM=37%	Need to include periodontal assessment and preventive treatment to a comprehensive approach as early as possible.
¹⁸ , 2017, N/I	Poland, 15	Children: 10-18y	ST: cross-sectional. Aim: differences in periodontal parameters and OH between cleft and control sides in growing patients with UCLP.	CLP vs Control CI=55% vs 43.3%; LI=67.5% vs 35.7% C = 50% vs 48.2%; 1st PM=39.3% vs 33.3%	Malformations of soft tissues and previous surgical procedures negatively affect periodontal parameters on the cleft side. Need to control development of periodontal disease.
¹⁰ , 2010, N/I	N/I, 60 (CLP: 20; CL: 20; CP: 20)	N/I	ST: cross-sectional epidemiologic. Aim: periodontal status of CLP patients.	Oral Hygiene Index - Simplified (OHI-S) Poor OH: CLP=75%; CP=30%; CL=20%.	Higher severity of periodontal disease for larger defects and involving the lip, alveolus and palate.
¹⁹ , 2010, July 2008 to July 2009	India, 120 UCLP: 60 40 ♂, 20 ♀ CP: 60 41 ♂, 19 ♀	Children: 12-18y	ST: cross-sectional. Aim: OH and periodontal status in patients with CP and UCLP.	Silness and Löe PI The poor OH status, as indicated by higher values of PI. UCLP>CP.	CLP patients had worse OH and periodontal status compared with those with CP.
³ , 2009, N/I	Greece, CLP: 41 Control: 41	Children: 4-18y	ST: case-control. Aim: OH and periodontal condition of children and adolescents with CLP.	CLP vs Control P < 0.005 PI upper front teeth 1.16 vs 0.82 Ramfjord 1.03 vs 0.76	Poorer OH (PI significantly higher): CLP>Control
²⁸ , 2009, N/I	Brazil, 400	15-49y	ST: cross-sectional. Aim: to evaluate whether teeth close to the cleft area present higher prevalence and severity of periodontal disease than teeth in other regions.	Silness and Löe PI Absence of plaque =2.60%; score 2 = 48.95%; score 3 = 40.50% Quantity of plaque =7.95%. No significant differences by dental arches, sextants and gender.	The presence of the cleft does not seem to increase the prevalence of periodontal disease.
²⁷ , 2007, N/I	Poland, 37. Germany, 63	Children: 3.5-18y	ST: cross-sectional. Aim: OH and periodontal status in children with UCLP and BCLP treated in Łódź, Poland, and Erlangen, Germany.	Optimal OH - API ≤40%: Erlangen: 60%; Łódź: 19% Poor OH Erlangen and Łódź: 57% Unsatisfactory 70%>API>40% or insufficient API >70% Erlangen: 47%; Łódź: 91%; P < 0.001	More plaque was found in the Polish population. Plaque accumulation appears not to be a key factor in causing periodontal destruction in the cleft area.
²⁰ , 1999, N/I	Austria, 80 (UCLP: 30, CP: 30, BCLP: 20)	17-24y	ST: cross-sectional. Aim: periodontal situation of patients with CLP.	API UCLP: poor OH=73%; BCLP: tendency to poor OH=69%; CP: average, but with Tendency to poor OH=65%	CLP patients presented a critical situation compared to CP patients.

Future Research: N/I stands for N/I regarding measures to improve dental hygiene. API, approximal plaque index; BCLP, bilateral cleft lip and palate; C, canine; CI, central incisors; DI-S, simplified debris index; LI, lateral incisor; N/I, No information; OH, oral hygiene; PI, plaque index; ST, Study type; UCLP, unilateral cleft lip and palate; 1st PM, first premolar.

plaque.¹⁻³ As a result, there is a high prevalence of dental caries in these children.^{4,5}

A number of reasons account to the difficulty in performing a proper oral hygiene, including in the cleft area, among them, fear of soft tissue trauma during brushing, concern about bleeding from inflamed gingiva, difficult to assess the teeth and the deepest area of the cleft and the decreased flexibility of surgical repaired lip.^{6,7} Appropriate hygiene is also needed for the abnormally positioned supernumerary tooth or teeth that may appear in the cleft area. This anatomical feature is an additional factor that hampers the maintenance of good local hygiene.⁸

The relevance of oral hygiene to improve oral health is highlighted in a variety of studies.^{2,6,8-10} It is also required for the successful outcome of the surgical corrective procedures. Dietary counselling and the use of antiseptic mouthwash help to minimize the problem but nothing compares with the efficacy of toothbrushing to remove dental plaque.^{8,9,11,12}

In spite of that, epidemiological studies on the oral hygiene of CLP children are scarce, and do not address the hygiene of the cleft area. The available information is quite dispersed and heterogeneous. Reported studies have notorious differences in the protocols, that is concerning the study population, evaluation period, measured variables and hygiene routines.

The aim of this systematic review is to critically select and analyse the literature information to verify if there is any solution to improve the hygiene of the cleft area. It is believed that this would fill a gap by providing comprehensive and easily accessible knowledge to the health professionals enrolled in treatment and preventive oral health programs targeting this population.

2 | MATERIALS AND METHODS

2.1 | Focused question

Are there any solutions for improving the cleft area hygiene in patients with clefts? Therefore, the main outcome of this systematic review is “instruments to improve the hygiene of the cleft area.”

2.2 | Search strategy

A structured literature search was performed on the reference bibliographic databases: PubMed, Web of Science, Cochrane Library and Scopus, from January 1997 to November 2017.

The MeSH terms used were Cleft lip, palate, harelip, oral hygiene and toothbrush, and the “AND” and “OR” Boolean operators were applied to combine keywords, respectively: ((CLEFT LIP AND PALATE) OR HARELIP) AND (ORAL HYGIENE); ((CLEFT LIP AND PALATE) OR HARELIP) AND TOOTHBRUSH. The choice of keywords was meant to be simple and since “cleft lip and palate” and “harelip” are synonyms, and most of the times are used as an alternative for one another, they were also included in the research in order to identify older studies which used this term, using the OR Boolean operator, adjunct with the main word of “cleft lip and palate.”

2.3 | Screening and selection

At first, title and abstract screening was performed. The inclusion criterion was the selection of articles in which oral hygiene in CLP patients was performed with a clinical purpose. The following exclusion criteria were applied:

- Meta-analysis and systematic reviews which are already a collection of previous studies included case reports, case series, descriptive studies, opinion articles, letters, responses to letters and articles that did not analyse oral hygiene in CLP patients;
- Studies related to patients using fixed appliances;
- Cases of bone graft;
- Single cases and syndromes;
- Articles focused on morphological teeth anomalies and nutritional health;
- Articles written in a language other than English;

The guidelines of Transparent Reporting of Systematic Reviews and Meta-analyses (PRISMA)¹³ were followed throughout the process.

Data from the selected articles were independently extracted by two researchers (RR, MCM) using a standardized form. Standard risk of bias tools was not applicable for this study type. Disagreements were resolved through discussion with a third reviewer (CCS). The extracted information was organized in three tables: characterization of bacterial plaque assessed by any quantitative index (Table 1), prevalence of dental caries assessed by dmft/dmfs and/or DMFT/DMFS (Table 2) and both issues (Table 3). Information collected on the oral hygiene routines (brushing habits, use of fluoride supplementation) is not present in the tables, but it is also described and discussed.

3 | RESULTS

3.1 | Search and selection

The literature search identified a total of 270 potentially relevant papers. Excluded studies included: duplicate studies (n = 119), studies not fulfilling the aim of this review (95), not fully accessible articles (10) and articles not written in English (6).

At the end, 39 articles were selected to be included in this systematic review. PRISMA flow diagram describing the methodology is presented in Figure 1.

3.2 | Heterogeneity

The studies included in this review exhibited extensive heterogeneity concerning the type, geographic area, age range, evaluation period and evaluated oral health parameters.

Regarding the study type, the review analysed 21 cross-sectionals^{1,14,15}; 9 case-controls^{2,3,5,6,8,12,34,35}; 2 epidemiological^{4,7}; 1 retrospective quantitative descriptive³⁷; 1 observational, cross-sectional³⁸; 1 prospective³⁹; 1 qualitative research¹¹ and 2 narrative reviews.^{9,40}

TABLE 2 Studies addressing the prevalence of caries, presented in descending chronological order

Reference, year of publication, period	Country & sample size	Adults/children	Study type (ST) and aim	Caries	Conclusion
⁵ , 2017, July 2011 to October 2012	USA, 161	Children: 1-18y 0-5y, n = 78 6-18y, n = 83	ST: cross-sectional. Aim: feasibility to implement CAMBRA during the medical visit at an institutional tertiary care centre for children with craniofacial anomalies.	Overall dental caries=57% (higher than in the general population) 0-5y vs 6-18y: 45% vs 69%, <i>P</i> < 0.01	Basic OH standards are not met. Need to implement protocols such as CAMBRA. Health care workers should to improve the understanding of OH and dental care in these patients.
³⁷ , 2017, 2009-2012	Philippines, 332	Children: 2-12y	ST: retrospective quantitative descriptive. Aim: overview on the prevalence of dental caries and caries index scores in children from the Noordhoff Craniofacial Foundation Philippines, Incorporated (NCFPI).	dmft/DMFT 2-5y vs 10-12y: 11.80 ± 6.05 vs 3.40 ± 2.72/8.80 ± 4.12, <i>P</i> < 0.0001 6-9y vs 10-12y: 11.98 ± 4.46/3.39 ± 1.91 vs 3.40 ± 2.72/8.80 ± 4.12, <i>P</i> < 0.0001	High caries prevalence in children with CL, CP and CLP.
³⁶ , 2015, N/I	India, 73 Group-I CLP: 23; Group-II control: 25 high caries risk; Group-III control: 25 caries-free	Children: 4-9y	ST: case-control. Aim: differences in risk factors of dental caries among children with CLP vs no CLP high caries risk and no CLP caries-free.	Group-I vs Group-II 8.65 ± 5.80 vs 12.64 ± 3.43, <i>P</i> < 0.01	Risk factors of dental caries: CLP ≈ Control high caries risk and Control caries-free
³⁰ , 2015, N/I	Brazil, 50 33 ♂, 17 ♀.	Children: 12y	ST: cross-sectional. Aim: relationship of dental caries and oral health with QoL in Brazilian patients with CLP.	Caries-free 26% DMFT 1.70, <i>P</i> > 0.05	The presence of CLP had a negative impact on QoL and this was also significant for tooth loss caused by dental caries.
²⁹ , 2014, July 2011 to October 2012	USA, 161 91 ♂, 69 ♀	Children: 0-5y, n = 78; 6-18y, n = 83	ST: case-control. Aim: feasibility to implement CAMBRA during the medical visit to a tertiary care centre for children with craniofacial anomalies.	Caries vs high risk of caries 0-5y: 45% vs 31% 6-18y: 69% vs 45%	N/I
²¹ , 2008, N/I	Southern Thailand, 138: CLP: 69 Control: 69	Children: 18-36 months	ST: cross-sectional. Aim: to clarify whether oral health care behaviour or oral cleft status influences ECC.	CLP vs Control DMFT: 9.19 ± 6.04 vs 6.46 ± 4.53 <i>P</i> < 0.001 Caries-free: 8.7% vs 10.1%	Greater caries experience in cleft children. Night-time feeding habit was the major factor for higher ECC in cleft children.
¹⁶ , 2006, 6 months	Brazil, Mothers +CLP children: 300	Children: 3-5y	ST: cross-sectional. Aim: oral health knowledge of CLP children mothers.	Children 77.7% of complete CLP: mean dmft 5.60 22.3% incomplete CLP: mean dmft of 6.45 <i>P</i> > 0.05	Mothers need to continue instruction and application of methods to prevention dental caries

(Continued)

TABLE 2 (Continued)

Reference, year of publication, period	Country & sample size	Adults/children	Study type (ST) and aim	Caries	Conclusion
²² , 2005, N/I	India, 83, 46 ♂, 37 ♀	Children: 2-5y	ST: descriptive and cross-sectional. Aim: primary dentition status and treatment needs in CLP children.	dmft 2y vs 3y: 6.57 ± 5.59 vs 3.95 ± 4.36, P > 0.05 4y vs 5y: 4.05 ± 5.43 vs 6.27 ± 3.80, P > 0.05 3y vs 4y: 3.95 ± 4.36 vs 4.05 ± 5.43, P > 0.05 Breast fed vs bottle/spoon fed: 5.11 ± 5.19 vs 5.67 ± 4.36, P > 0.05 CL vs CP: 2.40 ± 1.24 vs 5.67 ± 4.66, P < 0.05 CP vs CLP: 5.67 ± 4.66 vs 5.63 ± 5.45, P > 0.05	Prevalence of dental caries CL < CLP or CP
¹ , 2004, N/I	Brazil, 77	1-32y	ST: cross-sectional. Aim: oral health of children with CLP.	1-5y: dmft = 2.91 ± 3.99 6-12y: dmft = 2.77 ± 3.15. 6-12y: DMFT = 1.87 ± 1.78 13-18y: DMFT = 6.46 ± 3.11 19-32y: DMFT = 13.62 ± 6.51	The dental and periodontal condition of CLP patients was similar to the general population. Children 1-5y have a lack of healthy lifestyle and higher treatment needs.
³² , 2004, N/I	Vietnam, 154 54% ♂, 46% ♀	aged 4-6; 11-13; 14-16y	ST: cross-sectional. Aim: prevalence of dental caries in South Vietnamese children with a known oral cleft.	4-6y: dmft = 11.20 ± 6.45 6-12y: dmft = 2.77 ± 3.15 11-13y: DMFT = 5.06 ± 3.34 14-16y: DMFT = 7.07 ± 3.99	CLP patients have a high number of teeth affected by dental caries.
¹⁴ , 2001, N/I	China, CLP: 104 (CL: 25; CP: 79) 66 ♂, 38 ♀	Children: 3-6y	ST: cross-sectional. Aim: caries' levels in CL vs CLP children.	dmft: CLP = 75%/3.7 ± 3.3; CL = 68%/2.7 ± 2.5; CP = 77%/4.1 ± 3.8	CLP patients have higher levels of dental caries compared to those with CL or CP.
³¹ , 2001, N/I	UK, 91	Children: 4, 8, and 12y	ST: cross-sectional. Aim: prevalence of dental caries, developmental defects of enamel, and related factors in children with clefts.	4y - dmft = 1.3; caries-free: 63% 8y - DMFT = 1.8; caries-free: 36% 12y - DMFT = 1.8; caries-free: 34%	Dental disease is prevalent in clefts' patients. Need for a more aggressive preventive approach and optimize clinical outcome.
⁹ , 2000, N/I	N/I	N/I	ST: narrative review. Aim: to review the predisposing factors contributing to dental caries in CLP patients	CLP patients have a higher incidence of dental caries than patients without oral clefts.	Poorer OH and higher susceptibility to caries: CLP > Controls.
²⁴ , 1999, N/I	Taiwan, 123 68 ♂, 55 ♀	Children: 2y	ST: cross-sectional. Aim: caries prevalence in CLP children under 2y, and parental attitudes towards bottle-feeding, dental care and their relationship to baby bottle tooth decay.	Baby bottle tooth decay Bottle fed: 25% No bottle fed: 10.3% P = 0.019	Taking a bottle to bed increased the risk for developing baby bottle tooth decay. The parents showed a lack of motivation to perform regular preventive dental home care for their children.

CAMBRA, Caries Management By Risk Assessment; CL, central incisors; ECC, early Childhood caries; N/I, No information; OH, oral hygiene; QoL, quality of life; ST, Study type.

TABLE 3 Studies that report the bacterial plaque index (PI) and the prevalence of caries, presented in descending chronological order

Reference, year of publication, period	Country & sample size	Adults/children	Study type (ST) and aim	PI	Caries	Conclusion
⁴ , 2017, N/I	Romania, 64 54.5% ♂	Children: 3-6y	ST: epidemiological survey. Aim: extension of caries risk and decays incidence of primary teeth in CLP children.	OH=presence/absence. OHI index 1 = 22.7% OHI index 2 = 31.8% OHI index 3 = 45.5%	Decay of central upper incisors causes the decay of lateral incisors (80.6%).	Need to increase the motivation of parents to achieve a correct and complete OH.
⁸ , 2015, N/I	Sweden, CLP: 133 Control: 297	CLP 5y, n = 77; 10y, n = 56 Control 5y, n = 133 10y, n = 164	ST: case-control. Aim: compare the caries risk profiles: CLP vs controls using a computerized caries risk assessment model.	Modified Quigley-Hein PI CLP vs Control Low=45% vs 57% Medium=51% vs 42% High=4% vs 1%	Increased caries scores in the high-risk group (no significant difference for the cleft children, $P > 0.05$) dmfs High-risk CLP vs Control 5y (mean values): 1.2(2.5) vs 1.0(3.0) 10y: 1.6(2.3) vs 3.2(4.1)	CLP patients displayed increased odds of high caries risk due to impaired OH and elevated salivary lactobacilli counts.
²⁵ , 2015, N/I	UK, 264 67.4% ♂	Median age 5.5y	ST: cross-sectional. Aim: oral health and hearing outcomes from the Clinical Standards Advisory Group (CSAG, 1998) and the Cleft Care UK (CCUK, 2013) studies.	CCUK None=69.2% <1/3 of teeth=28.8% >1/3 of teeth=2%	CCUK vs CSAG with dmft=0 47.7% vs 45.2%, $P > 0.05$	Overall, the centralization of cleft services in the UK had little impact on oral health of children with UCLP.
²³ , 2014, N/I	Thailand, CLP: 68 34♂; 34♀ Control: 118 48♂; 70♀	Children: 10-14y	ST: cross-sectional. Aim: levels of dental caries, periodontal disease and oral health-related quality of life in children with CLP.	CLP vs Control 0.99 ± 0.47 vs 0.85 ± 0.55, $P = 0.02$	CLP vs Control Prevalence of caries 48.5% vs 20.3%, $P < 0.01$ DMFT: 0.82 ± 1.23 vs 0.38 ± 0.93, $P < 0.01$ dmft: 0.66 ± 1.38 vs 1.05 ± 1.72, $P = 0.08$	Levels of dental caries and gingivitis and poorer OH: CLP>Controls
³⁵ , 2013, March to December 2010	Brazil, CLP: 30 Control: 30 36.7%♂, 63.3%♀	12-21y	ST: case-control. Aim: caries experience of adolescents with CLP vs controls.	Ainamo and Bay PI CLP vs Control 75.60 ± 22.08 vs 50.09 ± 24.71 $P < 0.001$	Prevalence (CLP)=100% Prevalence (control)= 86.67%	The caries experience CLP>Controls
² , 2011, N/I	Jordan, CLP: 98 Control: 98-51♂, 47♀	Mean age 11.7 ± 6.3y	ST: case-control. Aim: dental health and OH status of children with UCLP and BCLP.	Silness and Løe PI BCLP vs UCLP 1.89 ± 0.73 vs 1.56 ± 0.65, $P = 0.02$	CLP vs Control dmft: 4.28 ± 4.19 vs 1.66 ± 1.440, $P < 0.001$ DMFT: 4.58 ± 5.37 vs 2.25 ± 2.04, $P < 0.001$ Caries experience: ♀>♂	Higher plaque accumulation and gingivitis, higher prevalence of dental caries in children with CLP in both permanent and deciduous teeth.

(Continued)

TABLE 3 (Continued)

Reference, year of publication, period	Country & sample size	Adults/children	Study type (ST) and aim	PI	Caries	Conclusion
³⁹ , 2011, April to June 2007	Nigeria, 52	N/I	ST: prospective. Aim: oral health status of individuals with CLP in Lagos	OH Good=38.6% Fair=45.5% Poor=15.9%	Decayed permanent teeth=15.9% Missing/filled in permanent dentition=0 Mean number of teeth affected by Caries: $5y=0.06$; $11-15y=0.5$; $26-30y=0.7$; $6-10y$ and $21-25y$ were caries-free	Poor oral health status of patients with CLP deformities.
¹² , 2009, N/I	Greece, CLP: 41 23♂, 18♀ UCLP: 26 BCLP: 10 CP: 5 Control: 41	Children: 4-18y	ST: case-control. Aim: health profile, more specifically, the prevalence of carious and hypoplastic lesions in CLP children and adolescents.	Silness and Løe PI CLP vs Control: Total dentition 1.03 ± 0.35 vs 0.76 ± 0.29 , $P = 0.0003$ Teeth adjacent to the cleft 1.11 ± 0.52 vs 0.82 ± 0.40 , $P = 0.0027$	Cavitated carious lesions were similar in both groups. Initial/white spot lesions, especially at the area of maxillary incisors, CLP=85% $P < 0.001$	The increased number of initial/white spot lesions combined with poor OH in the CLP group predisposes for a higher risk to develop carious cavitated lesions.
¹⁵ , 2007, 2004-2005	Germany, 100 (Polish: 37; German: 63)	Children: 3.5-18y	ST: cross-sectional. Aim: oral, dental status and level of oral health care among Polish and German CLP patients treated in Łódź and Erlangen.	Optimal OH (API $\leq 40\%$) DMF=2.175 \pm 2.986; dmf=1.500 \pm 2.640 Insufficient (70%>API>40%) DMF=3.917 \pm 2.74 dmf=4.000 \pm 2.944 Dissatisfying (API>70%) DMF=5.257 \pm 3.898 dmf=2.063 \pm 3.255 dmf $P < 0.001$ & DMF $P < 0.0001$	Caries prevalence CLP>general population, and worse in Polish patients. Need of programs for early plaque control.	
³⁸ , 2005, N/I	Jordan, CLP: 32 Control: 32	10-28y	ST: observational, transversal. Aim: levels of periodontal disease and dental caries of CLP vs controls.	Silness and Løe PI CLP vs Control 10-15y 1.79 ± 0.57 vs 1.28 ± 0.38 , $P = 0.016$ 16-28y 1.73 ± 0.59 vs 0.96 ± 0.30 , $P = 0$	CLP vs Control 10-15y: DMFT =4.76 \pm 5.11 vs 2.08 \pm 1.12, $P = 0.04$ 16-28y: DMFT=5.42 \pm 5.94 vs 2.11 \pm 1.10, $P = 0.008$	Increased risk for dental caries and periodontal disease in CLP patients.
⁶ , 2004, N/I	UK, CP: 81 Control: 61	Children: 6-16y	ST: case-control. Aim: to test if CP children have increased food oral clearance times, thus, higher levels of caries and caries-associated microorganisms.	CLP>Control CP: poorer OH $\chi^2 = 48.58$; $P < 0.001$ plaque scores: $\chi^2 = 63.44$; $P < 0.001$	CP vs Control dmft: 2.38 \pm 0.28 vs 0.62 \pm 0.17, $P < 0.001$ DMFT: 1.56 \pm 0.18 vs 0.48 \pm 0.17, $P < 0.001$	Caries>CP. This was associated with poorer OH and higher levels of caries-associated microflora.

(Continued)

TABLE 3 (Continued)

Reference, year of publication, period	Country & sample size	Adults/children	Study type (ST) and aim	PI	Caries	Conclusion
26, 1998, N/I	UK, 114 61.4% ♂, 38.6% ♀	Children: 3-18y	ST: cross-sectional. Aim: oral and dental health status of children with CLP.	OH index (Green and Vermillion) DI-S = 0.9 ± 0.7 CLP=1.1 ± 0.8 DI-S = CL: 0.7 ± 0.7 DI-S = CP: 0.7 ± 0.7 DI-S anterior vs DI-S posterior P < 0.001 DI-S upper vs lower P < 0.005 DI-S bilateral clefts vs unilateral clefts P < 0.05	No past caries=53.5% Active decay=20% dmfs: 2.3 ± 5.1 DMFS: 0.9 ± 1.7 Primary dentition: 3-5y year: dmfs = 1.9 ± 6.5 6-12y: dmfs = 2.8 ± 3.7 Permanent dentition: 6-12y: DMFS=0.4 ± 0.9 13-18y: DMFS=1.9 ± 2.2 According to cleft type: No previous caries CP=61.4%; CL=63.7%; CLP=45.8% DMFSs vs cleft types or laterality, P > 0.05	Active decay, poor OH and gingival health>anterior segment, possibly due to reduced access for cleaning the upper anterior teeth following surgical repair.
33, 1998, N/I	Russian, 89 62 ♂, 27 ♀	Age range: 5-9y	ST: cross-sectional. Aim: oral and dental health of Russian children who underwent Frolova primary palatoplasty.	OH: excellent=6%; good=28%; fair=44%; poor=22%	Caries-free=6% Low caries rate=2% Moderate caries rate=37% High caries rate=45% dft=7.31 ± 1.70 DFT=1.45 ± 3.69	The poor OH and high % of dental caries is likely due to unavailability of dental hygiene products and its high cost, limited parents' education on the importance of OH and appropriate diet to prevent dental disease.
34, 1997, N/I	Germany, 158 CLP: 81 49 ♂, 31 ♀ Control: 77 35 ♂, 42 ♀	Children: 0-2.5y 0-4y	ST: case-control. Aim: incidence of dental caries in the primary dentition in CLP vs controls	Poor OH CLP vs Control 59.3% vs 37.7% (P < 0.05)	Caries in the CLP group: ≈ 50% maxillary incisors, teeth nearby the oral cleft and in primary molars. CLP vs Control 30.9% vs 6.5% (P < 0.05)	CLP patients have higher risk for dental caries.

Future Research: N/I, no information; API, approximal plaque index; BCLP, Bilateral cleft lip and palate; OH, oral hygiene; PI, plaque index; ST, Study type; UCLP, unilateral cleft lip and palate.

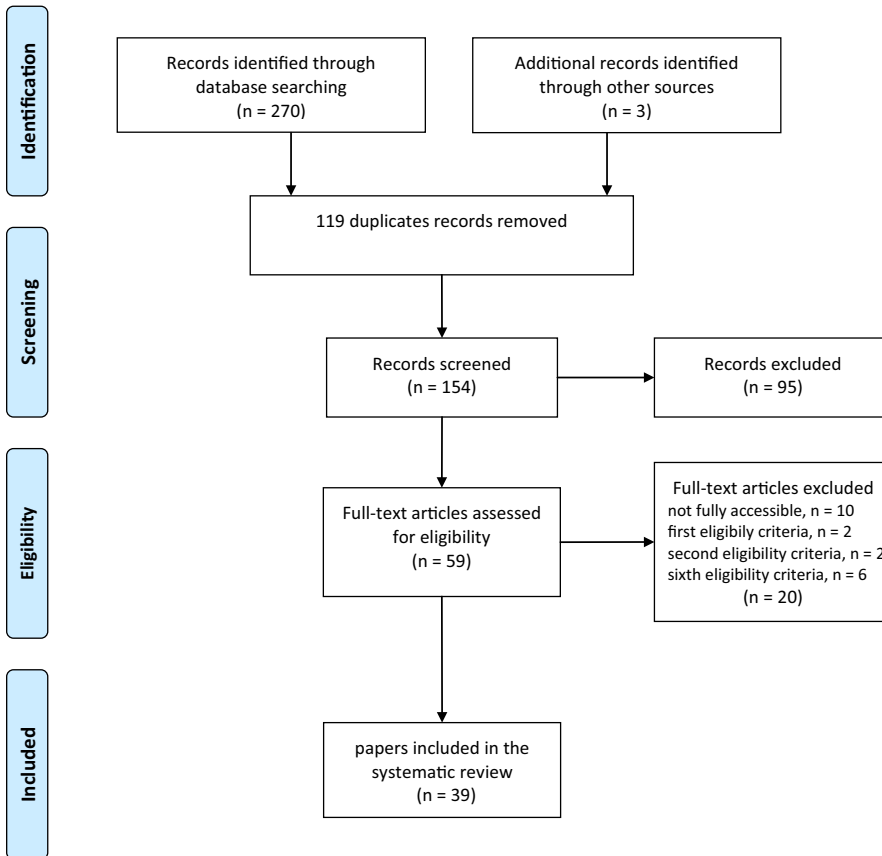


FIGURE 1 Information flow with the different phases of the search and selection process

Fifteen studies were conducted in European populations,^{3,4,6,8,11,12,15,17,18,20,25-27,31} thirteen from Asia,^{2,7,14,19,21-24,32,33} five from South America,^{1,16,28,30,35} two from North America,^{5,29} one from Africa³⁹ and three studies do not mention the country.^{9,10,40}

The age range varied from 0 to 49 years old, and the studies addressed a wide^{1,3,5,6,12,15,17,26-29,35} or a narrow^{4,7,8,11,14,16,17,19-23} age range, and some of them refereed only the mean age.^{2,25}

Most of the studies do not report on the evaluation period.^{1-4,6-12,14,17} About the oral health indexes, the plaque index was not reported in a variety of studies.^{1,5,7,9,11,14,21,22,24,29-32,36} Also, some of them address caries lesions other than those related to bacterial plaque^{1,3,5,10,14,16,17,19-22,24,27-29} and others investigate both issues.^{2,4,6,8,12,15,23,25,26,33-35,38} Information on brushing frequency and/or fluoride supplementation is lacking in some studies.^{1,2,9,10,17,19,20,24-28,30}

3.3 | Outcomes

The 39 studies included in this review involved a total of 3226 CLP patients and 914 matched control patients, sampling sizes of 15-400 CLP patients and were published between 1997 and 2017. It was possible to perceive that the main outcome "instruments to improve the hygiene of the cleft area" was referred as what is considered to be conventional toothbrushes. Regarding that, only one study was found⁹ and stated that the use of a baby-sized toothbrush supplemented with an interspace brush is recommended in some situations to improve oral hygiene of the CLP children.

The data extracted from the other 40 studies include oral hygiene, not instruments, and were conveyed as bacterial plaque index and dental caries. These were considered as secondary outcomes and were organized in three tables, each addressing, respectively, the bacterial plaque assessed by any quantitative index (Table 1), the prevalence of dental caries assessed by dmft/dmfs and/or DMFT/DMFS (Table 2) and both issues (Table 3).

Plaque indexes in CLP patients (Table 1) were evaluated in eight studies conducted from 1999 to 2017, most of them cross-sectional (one case-control), and involving sample sizes from 15 to 400 individuals within the age range 3.5 to 49 years old. All studies showed that CLP patients presented high plaque indexes.^{3,10,17-20} These were worse in CLP patients than in controls^{3,18} and more serious in CLP type than in CP type.^{10,19,20}

Sixteen studies were reported from 1999 to 2017 on the prevalence of dental caries in CLP individuals (Table 2), differing on the study type (systematic reviews and meta-analysis, narrative reviews, cross-sectional, retrospective, descriptive and case-control), geographic area, sample size and measured plaque indexes. All studies reported high prevalence of dental caries in the CLP population. Several studies showed that this prevalence was higher compared to common matched individuals,^{5,9,21} and two articles presented evidence of differences by cleft type (CL<CLP or CP) on the caries prevalence.^{14,22}

Table 3 includes the articles that addressed oral hygiene based on these two subjects—plaque index and dental caries. Fourteen studies, including an epidemiological survey, and cross-sectional,

case-control, prospective and observational studies, showed the same pattern compared to those that analysed these parameters separately. Oral hygiene (plaque index and prevalence of caries) is worse in CLP patients than in controls.^{6,12,23,35,38} This was observed in patients in different phases of oral development, and it was similar in the primary, mixed and permanent dentitions, suggesting that the increased risk of oral problems persists. Just one article showed evidence of differences by gender, that is females had higher caries experience than males, and this was worse in patients with bilateral clefts.² One study demonstrated differences by cleft type (CLP or CP).²⁶

Many studies also conducted a questionnaire addressing several variables related to oral hygiene. Data regarding the brushing habits were present in fourteen studies,^{4,5,7,8,11,14-16,21-23} and about half of them^{4,5,15,21,23,29} reported a frequency ≥ 2 daily brushings that is higher in CLP patients than in controls.^{16,35} The use of fluoride products was reported in a variety of studies,^{4-6,8,12,16,21,29,34} and some of them showed that this was higher in CLP patients than in controls.^{6,12,21,34,35} When specified, toothpastes,^{5,35,36} mouthrinses,^{6,16,21,35} supplements, professional application or fluoridation programs^{4,12} were used. The type of feeding in young children is referred in one study²⁴ and showed that those taking a bottle to bed presented an increased risk of developing tooth decay. In several studies, the questionnaire addressed the relevance of the education/motivation of the parents and the importance of parental supervision for the oral health of their CLP children.^{11,12,16,24} Two studies showed that the mother supervision on toothbrushing is higher in CLP patients than that observed in the controls.^{12,16} One of these studies, dated from 1999²⁴ and involving CLP children under 2 years of age ($n = 123$), observed that parents of bottled fed and non-bottled fed children showed no difference in knowledge regarding oral health of their children. Meanwhile a study performed in 2006,¹⁶ involving 300 mothers and CLP children, 3-5 years old, observed that a percentage of mothers had some knowledge on the causes and prevention of dental caries in their children. Regarding parents' motivation, the old study, from 1999,²⁴ detected a lack of mothers' motivation to perform regular preventive dental home care for their children. Interestingly, a recent qualitative research (2017) from United Kingdom involving 22 parents and 16 CLP children aged 2-11 years old revealed that parents need support to maintain and prioritize regular toothbrushing within the demanding family life.¹¹ Related to this, some studies stressed the relevance of care and prevention counselling to improve the oral health of CLP children.^{4,7,11,16,40} Others highlight the significance of including a dentist in the cleft team.⁷

4 | DISCUSSION

This systematic review selected and analysed the available information that deals with aspects related to oral hygiene in patients with CLP. At the beginning of the search, it was noted that most studies presented data mainly associated to aetiological, epidemiological and orthodontic aspects, and only few were specifically focused on

oral hygiene and related issues, namely plaque indexes, prevalence of dental caries and oral hygiene routines. Thus, applying the inclusion criterion and the exclusion criteria, 39 studies were selected from a total of 270 potentially relevant articles published between 1997 and 2017.

Bone and dental development abnormalities hinder the removal of the bacterial plaque in children with CLP, which is a major predisposing factor for the development of dental caries. Thus, the selected research outcomes were related to bacterial plaque indexes, prevalence of dental caries and oral hygiene routines.

The relevance of oral hygiene in the bacterial plaque indexes and prevalence of dental caries is well known. In this review, the selected articles included information on the bacterial plaque indexes,^{3,10,17-20} the prevalence of dental caries^{1,5,9,14,16,21,22,24,29-32} and on both plaque indexes and dental caries.^{2,4,6,8,12,15,23,25,26,33-35,38} Data on bacterial plaque and/or dental caries and oral hygiene were reported in almost half of the selected studies, but only eight studies addressed the three issues.^{4,6,8,12,15,23,34,35}

Most studies reported a high prevalence of dental caries in children with CLP^{8,10,14,33,34} regardless the cleft type; however, few studies^{14,22,26} relate the type of cleft to the incidence of caries. Others^{2,26} report a direct link between the severity of the cleft and the experience of caries. This high risk of caries may be correlated with the anatomy of the region, as it may retain food in the cleft^{36,40} as well as malocclusion.^{10,20,36} Compared to the general population, the incidence of dental development abnormalities is greater in CLP patients, in which anomalies in the shape (dental fusion, geminations and conoids),^{8,35} the enamel structure (dental hypoplasia's)^{9,12,15,36} and the number (agenesis or supernumeraries)⁸ are among the most frequently observed. The predisposing factors to dental caries in CLP children are also associated to the cariogenic microbiota,^{3,12,14,16} the type,^{14,22,26} the gender,² the breathing,³⁵ the fear of brushing the cleft area,⁸ the mother's education^{14,22} and a greater concern with the corrective surgeries than with the prevention or the early treatment of caries lesion.^{2,8,35,36} Also, following the surgical procedures, the presence of the healing tissues poses additional difficulties to the local hygiene and favours the retention and/or hinders the control of bacterial plaque, resulting in high plaque indexes^{12,23} and increased risk of periodontal disease.¹⁹ This is especially relevant in the area adjacent to the cleft^{3,17,20} and in patients with oronasal communication.^{8,16}

Oral hygiene routines (toothbrushing, use of fluoride products) were evaluated through a questionnaire, and although the relevance of this issue in the oral health of CLP children, these data were missing from many studies.^{1-3,9,10,17,19,20,25-28,30} Additionally, this did not improve over time. Even in the last decade, more than half of the studies focused on plaque indexes and/or dental caries (Tables 1-3). This is an important observation given the role of the oral hygiene in CLP children. Thus, much remains to be done to raise the awareness to the preventive dental care, both to health care workers and children/parents. In line with this, two studies conducted in USA to evaluate the feasibility of implementing Caries Management By Risk Assessment (CAMBRA) during the medical visit at an institutional tertiary care

centre for children with craniofacial anomalies concluded that the basic oral hygiene standards were not met in this high-risk population.^{5,29}

Other pertinent observation is related to the education in oral hygiene and the motivation of the parents to perform regular preventive dental home care for their children.^{4,7,11,16} The few studies that included this information noted that parents have great limitations concerning the importance of oral hygiene^{4,11} and, additionally, they need support to maintain their intention to accomplish regular toothbrushing and to prioritize it within the context of demanding family life.¹¹ Thus, continuous support and educational programmes on oral hygiene care, emphasizing its relevance to oral health and successful rehabilitation of CLP patients, are also essential for effective compliance. The importance of including a dentist in the cleft team is also emphasized.⁷

Perhaps the most unusual observation is that although the consensus on the poor oral hygiene of CLP children, no specific measures have been advanced to minimize this situation. For instance, toothbrushing and removal of dental plaque are strictly related, but poor attention has been given to the toothbrush. Only one of the studies⁹ mentions that a regular brush may not be effective in the hygiene of the cleft area. The authors propose that the use of a baby-sized toothbrush and an inter-space brush might be valuable to clean hard to reach areas.

This review has strengths and limitations. The selection process was rigorous and extensive. The inclusion criterion was well defined— inclusion of studies dealing with the oral hygiene in CLP children performed with a clinical purpose. Studies comprising data involving subjects with fixed appliances and bone grafts were excluded as these hamper clinical observation and brings an additional difficulty in maintaining the oral hygiene due to the fear of hurting the area undergoing surgery. Studies dealing with teeth anomalies and nutritional health were also excluded as they introduce confounding variables. Following the established selection criteria, the most relevant studies were included. However, the review did not consider articles that were not fully accessible or in another language other than English, which eventually might add some specific information on the subject. Another limitation was the great heterogeneity of the included studies regarding the geographic area, study population, study type, evaluation of the bacterial plaque and differences in the oral hygiene routines, which hinders the emerging of consensual patterns.

This review highlights some consensual information regarding the oral hygiene status of children with CLP. The commitment of paediatric dentistry professionals with the continuous dissemination of preventive measures and associated benefits is undoubtedly a challenge to be followed in order to improve the oral health status of these children. Preventive oral health programmes targeting this population and improving parent's knowledge on oral health are needed. It was also clear that there is a need of identifying a better solution to improve the hygiene of the cleft area. There are no studies dealing with specific strategies to minimize this problem, namely concerning the implementation of measures that effectively ensure a better oral hygiene. It is suggested that the development of a device/toothbrush with high efficacy in removing the dental plaque in the cleft area would minimize the complications associated with poor oral hygiene.

5 | CONCLUSION

The poor oral hygiene of CLP individuals is associated with high plaque indexes and prevalence of dental caries.

There is a clear need to improve the oral hygiene care of CLP children, but few studies were focused on specific preventive approaches. Development of devices especially designed to the hygiene of the cleft area and implementation of standardized prevention and control programs targeting education, motivation and compliance would contribute to improve oral health in CLP children.

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DISCLOSURES

None.

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