

ORIGINAL RESEARCH

Periodontal health and oral hygiene of children with orofacial clefts in Eastern China

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Abstract

To comparatively assess the periodontal condition and oral hygiene of children and adolescents at different ages presenting with different types of orofacial clefts (OFCs). A total of 1608 patients aged 6–18 years who had not previously undergone periodontal treatment were enrolled in this study. Participants were categorized into two age groups: 6–12 years (Group I) and 13–18 years (Group II). Participants in both age groups were further classified into one of the three OFC-type subgroups: cleft lip only (without or with alveolar cleft), cleft lip and cleft palate, and cleft palate only. Periodontal health was determined by evaluating plaque formation and gingival status with reference to the Silness and Loe plaque index (PI), Loe gingival index (GI), and community periodontal index (CPI). Periodontal health and oral hygiene were not significantly different between Groups I and II for cleft type ($p > 0.05$). A significant difference was not observed in PI for cleft type among the groups ($p > 0.05$). In Group II, GI and CPI were significantly higher than in Group I ($p < 0.05$). According to our results, cleft type does not influence periodontal health of children and adolescents with OFCs. Age, however, influences periodontal diseases' prevalence and severity.

Keywords

Cleft lip; Cleft palate; Periodontal index; Dental plaque; Dental plaque index

1. Introduction

Orofacial clefts (OFCs) are the most common congenital defects of the face, which may be present alone or in combination with other congenital malformations, especially congenital heart disease [1–4]. The defects are classified as cleft lip with or without cleft palate (CL/P) and cleft palate alone (CP). Approximately 70% of these defects are isolated, although some are associated with other syndromes, such as Trisomia 13 or Pierre-Robin sequence [5, 6]. OFCs occurs approximately 1.2 out of 1000 live births. Race, geographical location, and genetics are some of the contributing factors [7–9]. A meta-analysis in China (1986–2015) recorded that the overall incidence rate of OFC (including both comprehensive and noncomprehensive cleft lip and palate cases) was 1.4/per 1000 live births [10]. Asians and Native Americans have the highest incidence rates of OFCs, followed by Caucasians and Africans [7, 11, 12]. OFCs can cause delays in tooth formation and eruption, irregularities in the dental and arch segment, and mispositioning of teeth [1, 13–16]. Consequently, these defects may compromise periodontal health and exacerbate periodontal disease risk [17–20].

Treatment of OFCs requires an interdisciplinary team of medical and dental specialists, including pediatric and plastic surgeons, orthodontists, pedodontics, periodontists, prosthodontists, speech therapists and psychological counselors [21–23]. Only a few studies have been conducted

specifically on pediatric patients with OFCs in terms of periodontal health and oral hygiene. There is evidence that individuals with OFCs have an increased risk of developing periodontal diseases, including gingivitis. This is due to scar tissue in the upper lip, crowding of teeth, malformations and long-term orthodontic procedures [20, 24–28]. Periodontal disease increases with aging, and it affects the tissues that surround and support the teeth [29]. This condition usually manifests as gingivitis and is characterized by bleeding, gum swelling and pain. If left untreated, it can progress to periodontitis, leading to periodontal attachment loss and bone loss [29–32]. Previous studies have focused on the cleft area, and have not adequately examined the effects of age and different cleft types on periodontal disease. Data on periodontal diseases and oral hygiene in OFCs patients in China are almost nonexistent. Therefore, this study assesses periodontal health and oral hygiene among children and adolescents with OFCs.

2. Materials and methods

2.1 Study population

From March 2010 to August 2021, children and adolescents with OFCs aged 6–18 years at the Affiliated Hospital of Qingdao University were examined. A shortlist of study subjects was based on the following exclusion criteria: (1) syndromic

patients (*e.g.*, vela-cardio-facial syndrome, Pierre Robin sequence) or systemic abnormalities; (2) failing to cooperate with medical personnel; (3) having fixed orthodontic appliances; (4) having received periodontal treatment previously; (5) having systemic diseases; (6) and the lack of informed consent. As a result, 1608 patients were enrolled in this study. Fig. 1 illustrates detailed information about participant recruitment.

A sample size of $N = 98$ was calculated, with 80% power at 5% α -error, based on the literature [17, 33]. A final sample size of 1608 participants was selected based on recruitment parameters and exclusion criteria.

Participants were divided into two age groups: 6–12 years (group I, $n = 936$) and 13–18 years (group II, $n = 672$). In each age group, the participants were assigned to one of three orofacial cleft-type groups: cleft lip only, without or with alveolar cleft (CL group), cleft lip and cleft palate (CLP group), and cleft palate only (CP group). In detail, the CL group included unilateral, bilateral, complete and incomplete cleft lip cases. Complete cleft lip cases involve the alveolar cleft and nostrils, while incomplete cleft lip cases have slight gaps in the upper lip. In the CP group, the spectrum of the cleft palate ranged from the submucosal cleft to primary and secondary complete cleft palate. The CLP group included both cases occurring together.

This study analyzed 1608 patients aged 6–18 without previous periodontal procedures. Fig. 2 shows the classification of children according to age and cleft type.

2.2 Data collection

To examine the periodontal health status of the participants, along with their oral hygiene habits and gingival status, a dental mirror, an explorer and a periodontal probe were used by a single experienced physician. All but the third molar, various parameters were evaluated at six different sites. A plaque index (PI) was used to measure oral hygiene habits as described by Silness and Loe [34]. The gingival status was assessed as suggested by Loe [35]. We assigned a score of 0–3 to the assessed tooth as follows: 0 = normal gums; 1 = absence of gum bleeding on probing, with signs of mild inflammation, edema and an insignificant color change; 2 = the presence of gum bleeding on probing, with signs of moderate inflammation and edema; 3 = the presence of spontaneous bleeding, with signs of severe inflammation, marked redness, edema and ulcers. The overall periodontal condition was examined using a highly reproducible community periodontal index (CPI), which is documented in the World Health Organization (WHO) Global Oral Health Data Bank. Scores 0–4 were assigned as follows: 0 = healthy; 1 = bleeding on probing; 2 = the presence of calculus on probing (black hand of the probe is visible); 3 = probing depth of 4–5 mm (gingival margin on the black hand of the probe); 4 = probing depth of ≥ 6 mm (black hand of the probe is not visible).

2.3 Statistical analysis

Statistical analysis was performed using SPSS 18.0 software (IBM, Armonk, NY, USA). A Kolmogorov-Smirnov test was applied to prove normal distribution of PI and GI values in the

two groups, ordered data were assigned to CPI values. The mean, median, and standard deviations (SDs) were calculated. Unpaired *t*-tests were performed for PI and GI at different ages, one-way analysis of variance (ANOVA) was applied for PI and GI between different types and an ordered multi-classification rank-sum test was applied for CPI at different types and ages. $p < 0.05$ was considered to indicate statistical significance.

3. Results

For Group I, the mean PI was 1.88 ± 0.58 and the mean PIs for CL, CP and CLP groups in the category of 6–12 years were 2.04 ± 0.61 , 1.73 ± 0.15 and 1.87 ± 0.56 , respectively. For Group II, the mean PI was 1.78 ± 0.76 and the mean PIs for CL, CP and CLP groups in the category of 13–18 years were 1.77 ± 0.74 , 1.95 ± 0.17 and 1.73 ± 0.80 , respectively. The mean PI for the two age groups showed no significant differences based on OFC type (Group I, $F = 1.42$, $p > 0.01$; Group II, $F = 1.20$, $p > 0.01$) (Table 1). Similarly, the mean PI did not differ significantly between the groups ($t = 0.99$, $p > 0.05$) (Table 2).

For Group I, the mean GIs for CL, CP and CLP groups in the category of 6–12 years were 1.08 ± 0.43 , 1.11 ± 0.44 and 1.25 ± 0.42 , respectively. For Group II, the mean GIs for CL, CP and CLP groups in the category of 13–18 years were 1.25 ± 0.70 , 1.28 ± 0.50 and 1.46 ± 0.50 , respectively. The mean GI was higher for CLP than CL and CP for both age groups; however, there was no significant difference between both groups based on OFC type (Group I, $F = 2.15$, $p > 0.01$; Group II, $F = 1.26$, $p > 0.05$) (Table 3). In Group I, the mean GI was lower than in Group II ($t = 2.57$, $p < 0.05$) (Table 4).

Neither age group had a CPI score of 0. For Group I, 15%, 16% and 32% of children in the CL, CP and CLP groups, respectively, were assigned a CPI score of 1. A CPI score of 2 was assigned to 6%, 5% and 19% of children in the CL, CP and CLP groups, respectively. A CPI score of 3 was assigned to 0.9%, 2% and 4% of children in the CL, CP and CLP groups, respectively. A CPI score of 4 was assigned to 0.9%, 0% and 0% of children in the CLP, CL and CP groups, respectively. CPI scores were not significantly different between CL, CP and CLP groups ($H = 1.83$, $p > 0.05$) (Fig. 3, Table 5).

For Group II, 4% of children in each of the CL and CP groups and 18% of children in the CLP group were assigned a CPI score of 1. A CPI score of 2 was assigned to 15% of children in each of the CL and CP groups and 33% of children in the CLP group. A CPI score of 3 was assigned to 4% of children in each of the CL and CP groups and 1% of children in the CLP group. Neither group had a CPI score of 4. The CPI score did not differ significantly among CL, CP and CLP groups ($H = 1.85$, $p > 0.05$) (Fig. 4, Table 6).

None of the children had good periodontal health (*i.e.*, a CPI score of 0). For Group I, 63% of children were assigned a CPI score of 1; for Group II, 63% of children were assigned a CPI score of 2; and for Group I, 6% of children were assigned a CPI score of 3. For Group II, 10% of children were assigned a CPI score of 4. The CPI score differed significantly between Group I and Group II ($H = -13.39$, $p < 0.05$) (Fig. 5, Table 7).

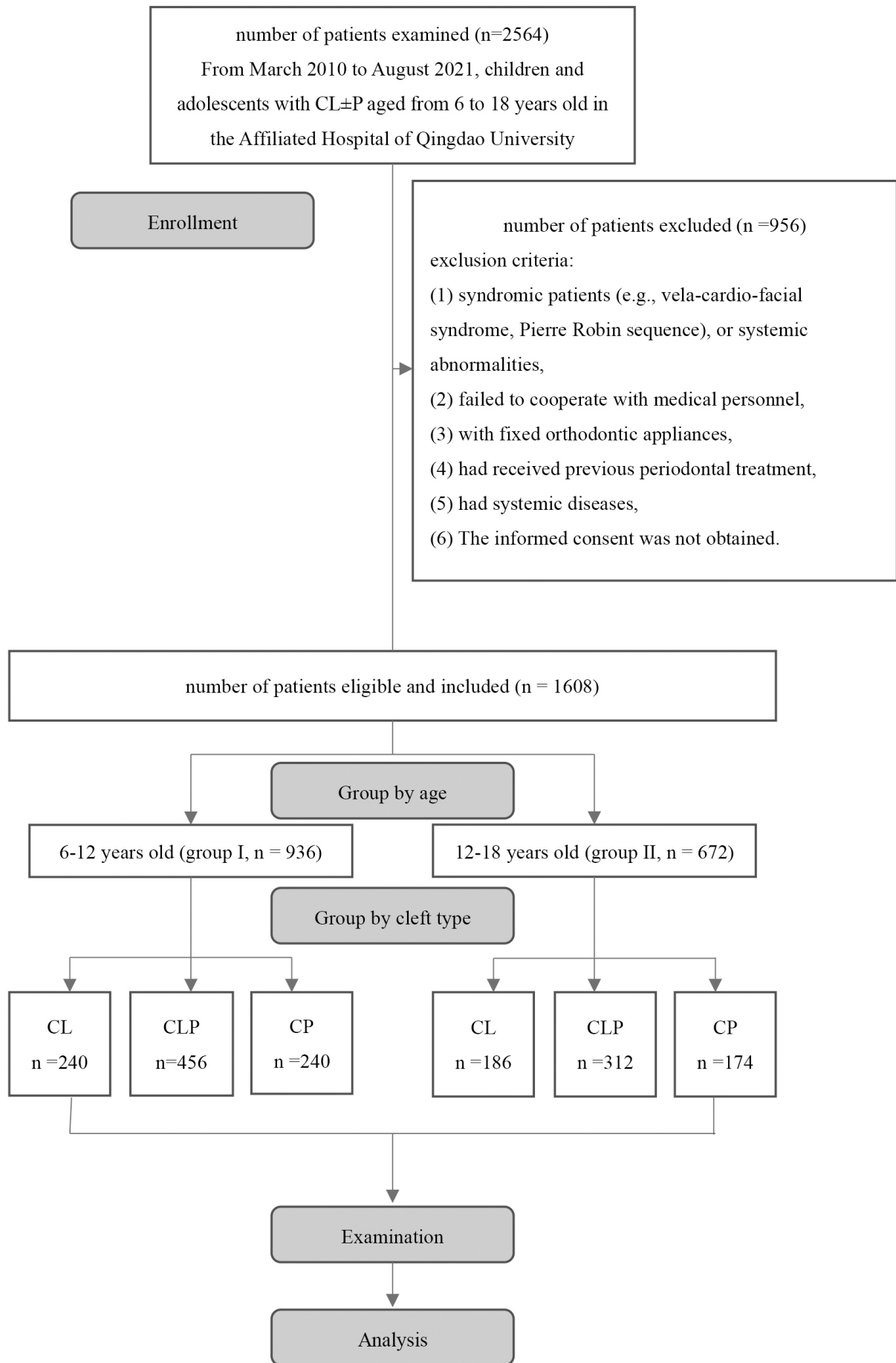


FIGURE 1. Recruitment participants' flowchart. CL: cleft lip, with or without alveolar cleft; CP: cleft palate only; CLP: cleft lip and cleft palate.

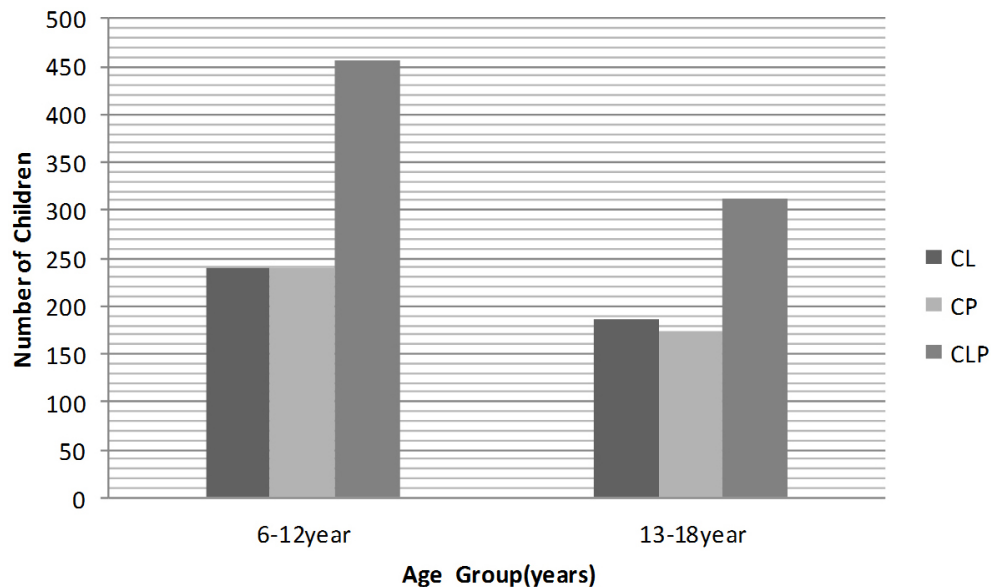


FIGURE 2. Proportions of participants according to age and cleft type. CL: cleft lip, with or without alveolar cleft; CP: cleft palate only; CLP: cleft lip and cleft palate.

TABLE 1. Comparison of PI score among 1608 patients in the age group using one-way ANOVA.

Age groups (yr)	Mean	CL	CP	CLP	F	p
6-12 (n = 936)	1.88 ± 0.58	2.04 ± 0.61 (n = 240)	1.73 ± 0.15 (n = 240)	1.87 ± 0.56 (n = 456)	1.42	0.242
13-18 (n = 672)	1.78 ± 0.76	1.77 ± 0.74 (n = 186)	1.95 ± 0.17 (n = 174)	1.73 ± 0.80 (n = 312)	1.20	0.302

CL: cleft lip, with or without alveolar cleft; CP: cleft palate only; CLP: cleft lip and cleft palate. *p*-value < 0.05 significant difference.

TABLE 2. Comparison of PI score among 1608 patients between age groups using unpaired *t*-tests.

Age groups (yr)	CL	CP	CLP	t	p
6-12 (n = 936)	2.04 ± 0.61 (n = 240)	1.73 ± 0.15 (n = 240)	1.87 ± 0.56 (n = 456)	0.99	0.322
13-18 (n = 672)	1.77 ± 0.74 (n = 186)	1.95 ± 0.17 (n = 174)	1.73 ± 0.80 (n = 312)		

CL: cleft lip, with or without alveolar cleft; CP: cleft palate only; CLP: cleft lip and cleft palate. *p*-value < 0.05 significant difference.

TABLE 3. Comparison of GI score among 1608 patients in the age group using one-way ANOVA.

Age groups (yr)	CL	CP	CLP	F	p
6-12 (n = 936)	1.08 ± 0.43 (n = 240)	1.11 ± 0.44 (n = 240)	1.25 ± 0.42 (n = 456)	2.15	0.117
13-18 (n = 672)	1.25 ± 0.70 (n = 186)	1.28 ± 0.50 (n = 174)	1.46 ± 0.50 (n = 312)	1.26	0.284

CL: cleft lip, with or without alveolar cleft; CP: cleft palate only; CLP: cleft lip and cleft palate. *p*-value < 0.05 significant difference.

TABLE 4. Comparison of GI score among 1608 patients between age groups using unpaired *t*-tests.

Age groups (yr)	CL	CP	CLP	t	p
6-12 (n = 936)	1.08 ± 0.43 (n = 240)	1.11 ± 0.44 (n = 240)	1.25 ± 0.42 (n = 456)	2.57	0.010
13-18 (n = 672)	1.25 ± 0.70 (n = 186)	1.28 ± 0.50 (n = 174)	1.46 ± 0.50 (n = 312)		

CL: cleft lip, with or without alveolar cleft; CP: cleft palate only; CLP: cleft lip and cleft palate. *p*-value < 0.05 significant difference.

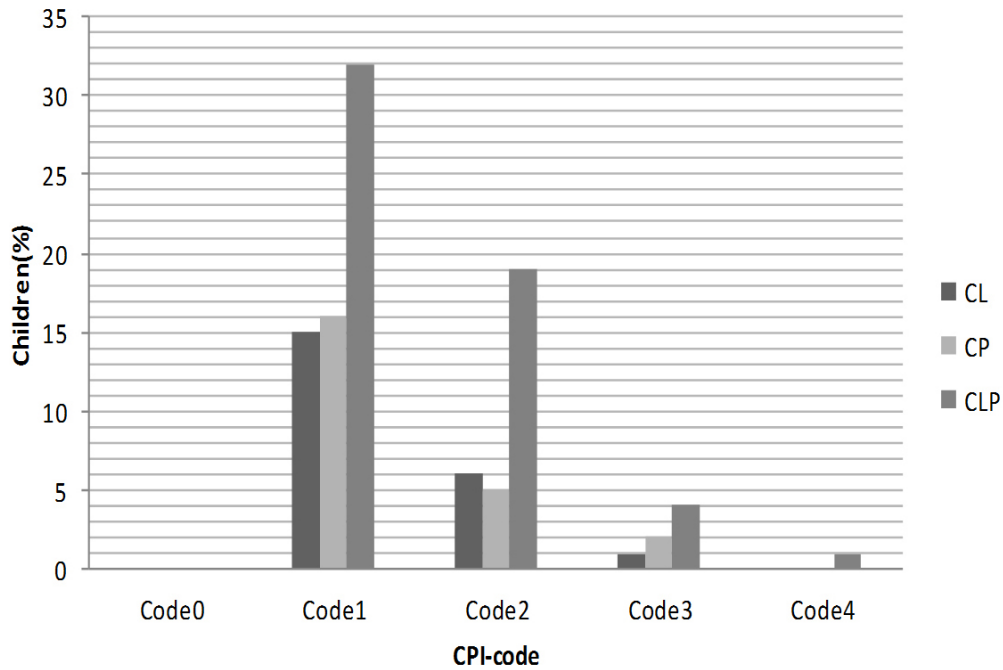


FIGURE 3. Proportions of participants aged 6–12 years old according to the CPI and cleft type. CL: cleft lip, with or without alveolar cleft; CP: cleft palate only; CLP: cleft lip and cleft palate; CPI: community periodontal index.

TABLE 5. Comparison of CPI score among 936 patients in 6–12 group using rank sum test.

Cleft type	M (P25, P75)	H	Rank Sum
CL (n = 240)	1 (1, 2)		
CP (n = 240)	1 (1, 2)	1.83	0.482
CLP (n = 456)	1 (1, 3)		

CL: cleft lip, with or without alveolar cleft; CP: cleft palate only; CLP: cleft lip and cleft palate. p-value < 0.05 significant difference.

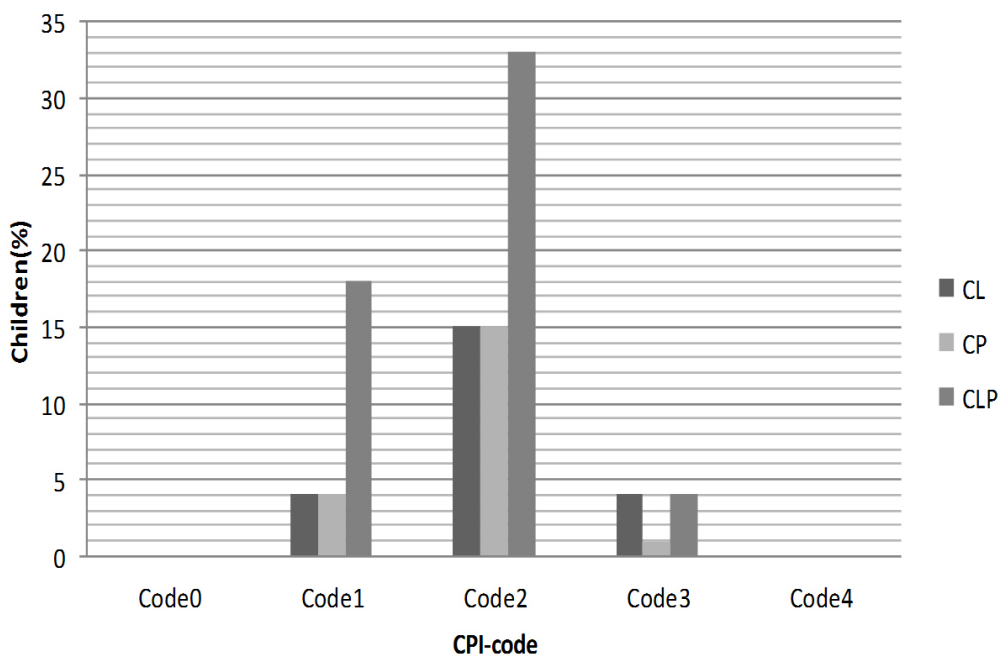
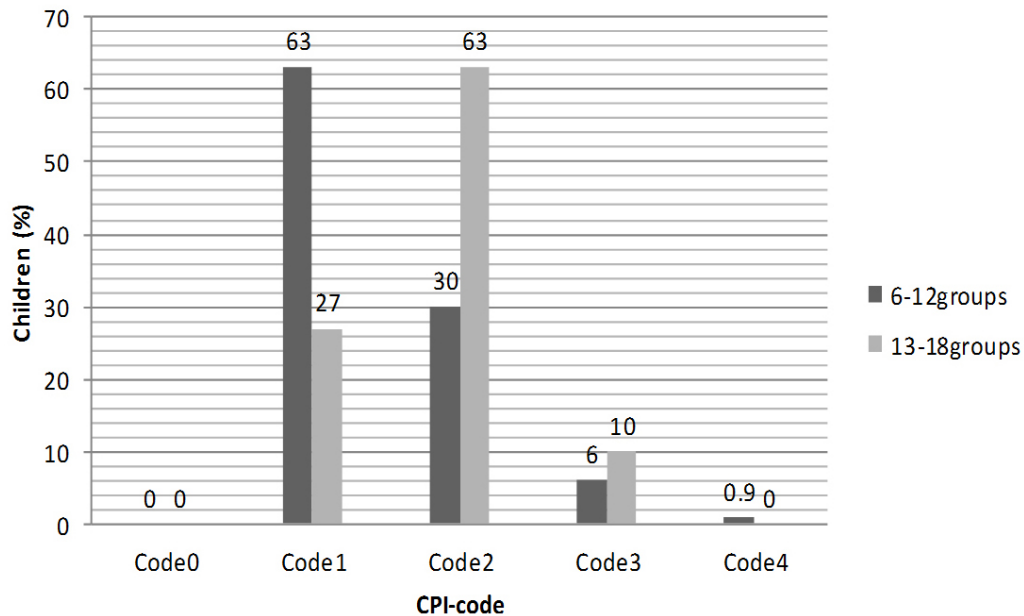


FIGURE 4. Proportions of participants aged 13–18 years old according to the CPI and cleft type. CL: cleft lip, with or without alveolar cleft; CP: cleft palate only; CLP: cleft lip and cleft palate; CPI: community periodontal index.

TABLE 6. Comparison of CPI score among 672 patients in the 13–18 group using rank sum test.

Cleft type	M (P25, P75)	H	Rank Sum	<i>p</i>
CL (n = 186)	2 (2, 2)			
CP (n = 174)	2 (2, 2)	1.85		0.478
CLP (n = 312)	2 (1, 2)			

CL: cleft lip, with or without alveolar cleft; CP: cleft palate only; CLP: cleft lip and cleft palate. *p*-value < 0.05 significant difference.

**FIGURE 5. Proportions of patients with cleft lip and cleft palate according to the CPI and age. CPI: community periodontal index.****TABLE 7. Comparison of CPI score among 1608 patients between age groups using rank sum test.**

Age groups (yr)	n	Mean Rank	Rank Sum	H	<i>p</i>
6–12	936	686.64	642,694.50		
13–18	672	968.66	650,941.50	-13.39	<0.001

p-value < 0.05 significant difference.

4. Discussion

There is a limited understanding of periodontal disease and oral hygiene among Chinese children with OFCs. This is the first to systematically investigate periodontal health and oral hygiene among children and adolescents in China. When partial teeth are studied, the periodontal health and oral hygiene of teeth near the OFC in children and adolescents may be overestimated or underestimated [24, 26, 28, 36]. Our study differs from previous studies in that all teeth except the third molars were examined. Considering the patient's willingness to cooperate with the examination and the focus on children and adolescents, a study age range of 6–18 years was chosen. As a dividing line, a median of 6 and 8 (12) was used, with 6–12-year-old children constituting one group and 13–18-year-old adolescents constituting the other. Age trends in children and adolescents with CL ± P concerning periodontal status were studied; hence, grouping in this manner was feasible.

Several studies have reported that periodontal disease is more likely to develop in children with OFCs than in the general population [17, 36, 37]. However, other studies have shown no differences in disease course [38, 39]. Sundell *et al.* [40] found no significant difference in PI scores and gingivitis proportions between the cleft and the control group in approximately 5-year-old patients. However, Salvi *et al.* [41] opined that individuals with cleft lip, palate and alveolar cleft were more likely to suffer from gingivitis and periodontal diseases than those with only cleft palate. A Hungary study reported similar results [42]. It is possible to explain the contradictory findings by the fact that children and adolescents with OFCs participated in an individualized preventive dental program that was different from the standard preventive program and benefited from it. According to Perdikogianni *et al.* [17], individuals with cleft lip-cleft palate had poorer oral hygiene (*i.e.*, higher PI) than those with cleft lip or cleft palate. Another study found that cleft palate individuals had similar

levels of periodontal disease to the general population. On the contrary, cleft lip-cleft palate patients were predisposed to deep periodontal tissue destruction [27]. Studies conducted in India found a greater severity of periodontal disease in individuals with OFCs than in those with cleft lip or cleft palate [19, 43]. Moreover, palatal fistulae may affect children's oral hygiene. A study of 89 children with OFCs found oral hygiene differed among different cleft types; children without palatal fistulae had better oral hygiene than children with this defect [44]. Poor oral hygiene is significantly more prevalent in patients with fistulae could be attributed to periodontal microorganisms' imbalance in the oral cavity. This explanation agrees with the relevant mechanisms of periodontitis and CLP, as summarized by Wu *et al.* [18]. Furthermore, Kenta *et al.* [45] noted functional dysbiosis in the plaque microbiota of CLP patients compared to controls. Briefly, fistulas drain nasal flora into the oral cavity, and nasal fluid's tenacious nature may exacerbate plaque adhesion to teeth [46–49]. Other investigations have also reached the same conclusion [17, 20, 50–54]. In the present study, the mean GI score did not differ significantly between cleft types, and a similar conclusion was drawn in another study [55]. Of children with cleft lip-cleft palate in other countries, those with OFCs had a higher mean GI score than those in Greece [17], but lower than those in Jordan [37]. In addition, children with OFCs had no increased incidence of periodontal disease. This could be related to their younger age, as age is a risk factor for periodontal disease [29, 56, 57].

The mean PI score of patients with OFCs did not differ significantly by age [58]. Sundell *et al.* [40] revealed no significant difference in PI scores between children with facial cleft and unaffected children. As well as Khan *et al.* [47]. In a study performed on Grecian children and young adults with clefts whose ages ranged from 4 to 20 years, Perdikogianni *et al.* [17] observed that the mean PI began increasing at the age of 16 years. Furthermore, Salvi *et al.* [41] noted a significant increase in plaque area in children with OFCs >14 years of age. These findings may have been altered by periodontal procedures, general oral cleanings, interventions in the form of restorative treatments, and orthodontic appliances given to the children. Our study found the highest prevalence of bleeding on probing (63%) in children with OFCs aged 6–12 years, whereas the highest prevalence of dental calculus formation (63%) was in those aged 13–18 years. A significant difference was also noted in the mean GI score, which increased with age, consistent with previous studies [17, 38]. An assessment of 41 Grecian children with OFCs conducted by Perdikogianni *et al.* [17] revealed that most patients had dental calculus formation. In this study, a score of 3, which corresponds to probe depths of 4–5 mm, was assigned to 6% and 10% of children in Group I and Group II, respectively. As a result, the mean PI score obtained in this study was higher than that obtained in a previous report [59] (1.87 ± 0.56 vs. 1.82 ± 0.3 , respectively). This phenomenon might be explained by the age difference between both studies [59]. In contrast to the previous study, which examined children aged 5–6 years, the present study assessed children aged 6–18 years. Therefore, OFCs do not seem to increase the prevalence of periodontal disease. These observations may be attributed to the importance of early assessment of cleft lip and palate and

permanent dentition in preventing periodontal diseases.

Periodontal disease is more likely to progress in patients with oral clefts [41, 58]. Inflammatory responses were observed in periodontal tissues in children with OFCs [28, 36, 41]. In patients with OFCs, periodontal clinical indicators may be worse than those without; however, inconsistent approaches and factors other than the presence or absence of cleft palate may influence the development of periodontal disease. These factors include but are not limited to, age, diabetes and other systemic diseases, immune response, oral flora, oral hygiene habits, salivary flow and composition, orthodontic treatment, prosthodontic appliances and periodontal maintenance therapy [36]. Race, diet, feeding habits and socioeconomic status also influence periodontal health and oral hygiene [28]. Oral hygiene problems in children with OFCs are caused by the stiffness of the upper lip due to scar tissue formation, cleft site, orthodontic retention appliance, reduced gingival width, and crowding and malformation of teeth [60]. Psychological factors, such as concerns about soft tissue damage, gingival inflammation and bleeding during brushing, exacerbate the difficulty of maintaining proper oral hygiene [61]. Moreover, pain during tooth brushing affects children's oral hygiene management [18, 62]. Plaque formation is therefore difficult to control in these children [27, 63, 64]. Neither age group of children with OFCs presented with severe periodontal disease, but its prevalence increased with age. Therefore, cleft lip-cleft palate patients should receive comprehensive oral examinations and follow-up care, including periodontal therapy. Dental specialists should instill routine oral health habits in patients to help establish and maintain good oral care habits.

As research continues to shed light on the impact of periodontal health on overall health, people have started paying increased attention to oral health. Periodontal health affects not only the oral cavity but also overall health [65–67]. Patients with OFCs require special attention. Our study focused on clinical data. For further studies, we intend to incorporate confounding factors associated with poor periodontal conditions to comprehend the correlation between oral hygiene habits, dietary economy and oral health. As a highly variable environment, certain alterations in the periodontal microbiota of the oral cavity can help in better understanding the reasons for periodontal health and oral hygiene changes in children with cleft lip and palate in East China from a microbial standpoint.

5. Conclusions

According to the PI, GI and CPI scores, children with OFCs had periodontal disease commonly. However, the types of clefts did not influence the prevalence of periodontal diseases. Periodontal parameters are not influenced by OFC type, despite age being a key risk factor in periodontal disease development and oral hygiene. As children age, they should be guided to pay more attention to oral hygiene by dental specialists.

Stringent and early supportive periodontal therapy should be administered to patients with oral clefts to maintain stable periodontal conditions.

AVAILABILITY OF DATA AND MATERIALS

Not applicable. Due to privacy protection and informed consent of patients' parents or primary guardians, the datasets generated and/or analyzed during the study are not publicly available.

AUTHOR CONTRIBUTIONS

XWL—designed and administrated the research study. CL, LFX and YXX—performed the research and supervised the data collection. JY—analyzed the data. CL and LFX—wrote the manuscript. JZZ—reviewed the draft of the manuscript. All authors contributed to editorial changes in the manuscript. All authors read and approved the final manuscript.

ETHICS APPROVAL AND CONSENT TO PARTICIPATE

The research was conducted with the approval of the Ethics Committee of the Affiliated Hospital of Qingdao University (number: QDFY-09-12). Written informed consent has been obtained for all patients' parents or primary guardians prior to the start of the study.

ACKNOWLEDGMENT

Not applicable.

FUNDING

This research was supported by the Shandong Province Natural Science Foundation of China (grant no. ZR2015HM022).

CONFLICT OF INTEREST

The authors declare no conflict of interest.

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How to cite this article: Cong Li, Ling-fa Xue, Yao-xiang Xu, Jin Yue, Jin-ze Zhao, Wen-lin Xiao. Periodontal health and oral hygiene of children with orofacial clefts in Eastern China. *Journal of Clinical Pediatric Dentistry*. 2024; 48(5): 86-94. doi: 10.22514/jocpd.2024.107.