

Oral habits in children with cleft (lip and) palate - a pilot study

Running title: Cleft palate and oral habits

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Abstract

Background/Aims: This pilot study aimed to evaluate the occurrence of oral habits and non-nutritive sucking habits in children with cleft (lip and) palate (CL/P) between 0-12 years, compared to peers without CL/P.

Methods: Children with CL/P (mean age 6.0y, SD 2.80) were recruited from the Ghent University Hospital and compared to gender- and age-matched children without CL/P (mean age 5.8y, SD 3.09). A self-report questionnaire was developed and given to the caregivers. To compare the occurrence of oral habits and non-nutritive sucking habits in children with and without CL/P, Chi-square tests or Fisher's exact tests were used.

Results: Present study revealed a significantly lower occurrence of non-nutritive sucking habits in children with CL/P. These children showed significantly less pacifier sucking; no significant difference was found regarding thumb or finger sucking nor regarding sucking on other objects. Additionally, the results showed that the occurrence of snoring was statistically significantly higher in children with CL/P.

Conclusion: Children with CL/P show less non-nutritive sucking habits but more snoring compared to a control group without CL/P. Although further research regarding oral habits in children with CL/P is necessary, cleft teams should give attention to the occurrence of oral habits.

Keywords

Oral habits, non-nutritive sucking, cleft palate, children

1. Introduction

A habit is a behavior pattern acquired by frequent repetition or physiologic exposure that shows itself in regularity [1] [2]. Related to the orofacial area, children frequently acquire oral habits. Oral habits can present temporarily or remain permanently. Although some of these habits can be considered as a normal part of development [3], they may be harmful when excessively repeated [2], resulting in a negative impact on oral postures and functions [4]. Open mouth behavior, habitual open mouth breathing, lip licking, and tongue, nail or cheek biting etc. are examples of oral habits. Bruxism (i.e. teeth grinding), drooling and snoring are frequently accompanying aspects related to these oral habits [5]. Non-nutritive sucking habits include the use of pacifiers, digit sucking (i.e. thumb and/or finger sucking) and sucking on other objects (e.g. blankets, toys).

Oral habits and non-nutritive sucking habits can interfere not only with the position of the teeth, but especially with the normal skeletal growth pattern. The specific effect depends on the nature, onset and duration of these habits (e.g. persistent non-nutritive sucking may result in unbalanced muscle function, hence affecting the stomatognathic system) [5]. Ling, et al. [6] showed that children with more than one year of daily pacifier use and/or thumb/digit sucking had higher chances of developing abnormal dental relationships (e.g. anterior open bite or increased overjet). A recent systematic review on pacifier sucking concluded that there is evidence that the use of a pacifier is associated with anterior open bite and posterior cross-bite, thus affecting harmonious development of orofacial structures [7].

Children born with a cleft of the (lip and) palate (CL/P) form a unique population when it comes to oral habits and non-nutritive sucking habits. First, a cleft of the palate impedes the creation of negative intraoral pressure during sucking [11], preventing nutritive sucking [12] and probably also reducing non-nutritive sucking acts such as pacifier use [2]. Second, surgeries in the orofacial area are frequently performed in the early stages of life (e.g. lip closure, palatal closure etc.). One may wonder whether pain, resulting from these surgical interventions, reduces the occurrence of non-nutritive sucking habits and other oral habits in this specific population. Furthermore, parents are counseled to remove the pacifier in the postoperative period of lip/palate closure, possibly resulting in further deconditioning of non-nutritive sucking habits [2].

Third, in children with CL/P, removal of the adenoids is often avoided in order to prevent the aggravation of speech problems [18]. This may result in a higher occurrence of oral habits (e.g. open mouth behavior/habitual open mouth breathing and the accompanying act of snoring) in case of hypertrophic adenoids.

However, literature on oral habits in children with CL/P is scarce. Barsi, et al. [2] were the first to describe the prevalence of oral habits in this unique population. Results of the study showed that there was no statistically significant association between oral habits and the presence of palatal fistulae. The children with CL/P showed significantly more tongue thrusting at rest, in speech and during swallowing (i.e. forward movement of the tongue tip between the teeth) and more tongue sucking, object sucking, lip sucking, cheek sucking and nail biting compared to the group of

children without CL/P. Additionally, there was a statistically significant association between the presence of a cleft and the lack of pacifier sucking. However, the occurrence of other oral habits (e.g. habitual mouth breathing, open mouth behavior, tongue and cheek biting) and accompanying aspects (e.g. drooling, snoring, bruxism) was not assessed.

Digit sucking is often reported as a frequently occurring oral habit in children without CL/P [12]. However, in the population of children with CL/P, literature regarding digit sucking is scarce. Only two studies reported this non-nutritive sucking habit. One case report described digit sucking in a child with an unrepaired cleft living in a developing country [13]. However, the results of this study are not representative to all children with CL/P. The second study [2] reported no statistically significant association between CL/P and thumb and/or finger sucking.

Besides the limited amount of literature on the presence of oral habits in children with CL/P, generalization of these results to Dutch-speaking Flemish children is difficult as cultural (e.g. acceptance of specific habits), sociodemographic and socioeconomic differences may influence the occurrence of these habits [8, 9]. Specifically for Flanders (the northern part of Belgium), the prevalence of thumb sucking and pacifier sucking in children between 0 and 12 years old without CL/P is 10.7% and 74.8% respectively [10]. Twenty-three (0.9%) of the 2440 questioned children had a cleft of the palate. In this specific group, the occurrence of thumb sucking was 21.7%, which is remarkably higher compared to the prevalence of 10.7% in the group of children without CL/P. Unfortunately, no data regarding pacifier use were available for this subgroup.

Children with CL/P exhibit considerably more dental anomalies and problems in the orofacial area than children without clefts [14]. As the presence of a cleft may possibly influence oral habits and non-nutritive sucking habits, identification of the occurrence of these habits is important in order to prevent aggravation of already existing problems and to counsel parents.

In summary, literature considering the occurrence of non-nutritive sucking habits and oral habits in children with CL/P is scarce and results are difficult to generalize due to cultural and socioeconomic factors. Therefore, the aim of the present study was to document differences in the occurrence of non-nutritive sucking habits and oral habits in Dutch-speaking Flemish children with repaired unilateral or bilateral cleft (lip

and) palate between 0-12 years of age and to compare the results with an age and gender matched group of Dutch-speaking Flemish children without cleft (lip and) palate. Based on literature, it was hypothesized that (1) the occurrence of non-nutritive sucking habits (i.e. the use of a pacifier and object sucking) would be lower in children with CL/P, (2) the occurrence of thumb/finger sucking would be higher in children with CL/P and that (3) the occurrence of oral habits (open mouth behavior, open mouth breathing, lip licking, nail, finger or cheek biting) and accompanying aspects (drooling, snoring, bruxism) would be higher in children with CL/P.

2. Material and methods

This prospective cross sectional study was approved by the Ethics Committee of the Ghent University Hospital (2016/1228). All children and their parents participated voluntarily and the latter signed an informed consent.

2.1 Participants and data collection

Dutch-speaking Flemish children (0-12 years) were recruited in the period of September 2016 to June 2017. The group of children with CL/P was recruited from the multidisciplinary craniofacial team of Ghent University Hospital, based on convenience sampling. The native language of all children was Dutch. Inclusion criteria for this group were (1) operated cleft palate whether or not in combination with repaired cleft lip, (2) normal airway and (3) absence of cognitive and related learning disabilities or syndromes.

An age and gender matched Dutch-speaking group of Flemish children without craniofacial (including CL/P) or velopharyngeal anomalies was included. Exclusion criteria of the children without CL/P were (1) syndromes, (2) mental disabilities and (3) no age/gender matching with the children with CL/P. Children were recruited via schools using convenience sampling and snowball sampling.

2.2 Questionnaire

The used parental questionnaire indicated the several types of oral habits: (1) non-nutritive sucking habits (i.e. the use of a pacifier, thumb sucking or sucking on other objects), (2) oral habits (i.e. mouth breathing, open mouth behavior, lip licking, nail, tongue or cheek biting) and (3) accompanying acts (i.e. drooling, snoring and

bruxism). The caretakers had to choose between 'yes, the habit is still going on', 'the child had a habit earlier but it has stopped', 'no' or 'I don't know'.

Parents were given the self-administered questionnaire during the consultation at the cleft team in the Ghent University Hospital. They were asked to answer the questions as completely as possible and their anonymity was guaranteed.

In order to prevent the possible problem of "socially desirable" answers, the questionnaire indicated that there were no right or wrong answers and additionally, no leading questions were used.

Before the questionnaire was handed to the caregivers of the children with and without CL/P, it was tested to 5 individuals (parents of children without CL/P not familiar with research and not participating in present study) to verify clearness of the questions and responses.

2.3 Statistical analysis

Statistical analysis was performed using SPSS version 24.0 (SPSS Inc., Chicago, Illinois) to compare the occurrence of oral habits and non-nutritive sucking habits between children with and without CL/P. Significance level was set at $p \leq 0.05$. Categorical variables were compared using a Pearson Chi-square test (2x2 table) with $p \leq 0.05$ considered statistically significant. If 20% or more of the cells had an expected count less than 5 or the smallest expected count was less than 2, a Fisher's exact test was used.

3. Results

3. Results

3.1. Participants

In total, 26 patients with CL/P between 1 and 11 years old (mean age 6.0y, SD 2.80) were included. The group consisted of 15 boys and 11 girls. Twelve patients had a unilateral cleft lip and palate (UCLP), three had a bilateral cleft lip and palate (BCLP) and eleven had a cleft palate (CP). Median age of lip closure was 3 months (interquartile range (IQR): 3-5m), median age of palatal closure was 12 months (IQR: 8-14m).

The initial group of children without CL/P consisted of 49 children of whom 23 were excluded because there was no age and/or gender matching. Finally, a group of 26 children between 1 and 12 years old (mean age 5.8y, SD 3.09) without CL/P was established. Based on an independent samples t-test, mean age of both groups demonstrated no statistically significant difference ($t(50) = -0.376, p = 0.708$).

None of the children suffered from acute ear, nose or laryngeal disorders and all of them received normal education. Data were obtained by questioning the parents and available medical records.

3.2. Non-nutritive sucking habits

Table 1 provides descriptive information regarding the oral habits and accompanying aspects. Comparing the two groups, children with CL/P showed significantly less sucking habits ($X^2(1) = 11.337, p = 0.002$) (Table 1). Further analysis revealed that children with CL/P showed significantly less pacifier sucking ($X^2(1) = 24.923, p < 0.001$), although no significant difference was found regarding thumb or finger sucking ($X^2(1) = 0.433, p = 0.743$) nor regarding sucking on other objects (Fisher's Exact test, $X^2(1) = 1.486, p = 0.419$). Furthermore, no significant difference was found for the actual presence of a sucking habit at the moment of completing the questionnaire ($X^2(1) = 0.262, p = 0.730$).

3.3. Oral habits and accompanying aspects

Children with CL/P did not show significantly more presence of habitual open mouth behavior (Fisher's exact test, $X^2(1) = 0.165, p = 1.000$), nor open mouth breathing (Fisher's exact test, $X^2(1) = 0.361, p = 0.764$) compared to children without CL/P. Furthermore, no significant differences were found for lip licking (Fisher's exact test, $X^2(1) = 0.591, p = 0.703$), nail, tongue or cheek biting ($X^2(1) = 1.981, p = 0.291$), bruxism ($X^2(1) = 1.981, p = 0.291$) and drooling (Fisher's Exact test, $X^2(1) = 2.364, p = 0.248$). However, significantly more children with CL/P did snore compared to children without CL/P at the time of completing the questionnaire (Fisher's exact test, $X^2(1) = 4.952, p = 0.050$).

Table 1: comparison of the occurrence of non-nutritive sucking and oral habits in children with and without CL/P

	Occurrence of non-nutritive sucking and oral habits			
	Children with CL/P	Children without CL/P	X ² (df)	p
Sucking habits – total**	50% (13/26)	92.3% (24/26)		0.002*
Pacifier sucking	15.4% (4/26)	84.6% (22/26)	24.923 (1)	<0.001*
Thumb/finger sucking	26.9% (7/26)	19.2% (5/26)	11.337 (1)	0.743
Object sucking	7.7% (2/26)	19.2% (5/26)	1.486 (1)	0.419
Oral habits				
Open mouth behavior	15.4% (4/26)	11.5% (3/26)	0.165 (1)	1.000
Open mouth breathing	34.6% (9/26)	26.9% (7/26)	0.361 (1)	0.764
Lip licking	19.2% (5/26)	11.5% (3/26)	0.591 (1)	0.703
Nail/finger/cheek biting	11.5% (3/26)	26.9% (7/26)	1.981 (1)	0.291
Accompanying aspects				
Bruxism	26.9% (7/26)	11.5% (3/26)	1.981 (1)	0.291
Drooling	23.1% (6/26)	7.7% (2/26)	2.364 (1)	0.248
Snoring	61.5% (16/26)	30.8% (8/26)	4.952 (1)	0.050*

*Pearson chi-square test, $p \leq 0.05$ ** Sucking habits – total (i.e. no distinction between pacifier sucking/thumb sucking/finger sucking/object sucking).

4. Discussion

The aim of present pilot study was to document differences in oral habits in Dutch-speaking Flemish children with CL/P compared to a group of age and gender matched children without CL/P. It was hypothesized that (1) the occurrence of non-nutritive sucking habits (i.e. the use of a pacifier and object sucking) would be lower in children with CL/P, (2) the occurrence of thumb/finger sucking would be higher in children with CL/P and that (3) the occurrence of oral habits (open mouth behavior, open mouth breathing, lip licking, nail, finger or cheek biting) and accompanying aspects (drooling, snoring, bruxism) would be higher in children with CL/P. In summary, present study revealed a statistically significant lower occurrence of non-nutritive sucking habits (especially the use of a pacifier) in children with CL/P

compared to children without CL/P. Additionally, it was found that children with CL/P showed a statistically significant higher occurrence of snoring when compared to children without CL/P.

Santos, et al. [15] in Brazil showed that pacifier sucking was the most prevalent non-nutritive sucking habit in children without CL/P. In Flanders (Belgium), similar results were found [10]. The results for children without CL/P in the current study were in line with these findings [Table 1]. As hypothesized, pacifier use in children with CL/P was significantly lower. As mentioned in the introduction, the inability to create intraoral pressure before palatal closure, which is needed during (pacifier) sucking, may cause this difference. Additionally, pacifier use is discouraged in the postoperative period of lip/palate closure, resulting in further deconditioning of non-nutritive sucking habits. The latter hypothesis was previously suggested and highlights the need for further research [2]. Given the low occurrence of pacifier sucking in children with CL/P, it is remarkable that there is no higher occurrence of other non-nutritive sucking habits (i.e. thumb or finger sucking and sucking on other objects). In other words, one may assume that children with CL/P do not compensate their decreased ability to use a pacifier by performing other non-nutritive sucking habits. Further research including prospective randomized studies are interesting to assess influence of oral habits on postoperative outcomes.

Furthermore, a comparable percentage of occurrence for thumb and finger sucking in children with CL/P was found in the current study (26.9%) and the sample of children with CL/P included in the prevalence study of Delobelle and Verbrugge in Flanders (21.7%) [10]. However, despite non-significant results for thumb and finger sucking between children with and without CL/P in present study, the occurrence of these particular habits was higher (19.2%) compared to Delobelle and Verbrugge (10.7%) [10]. This may be due to the smaller sample size and different sampling strategies and is therefore subject for further research.

Barsi et al. [2] reported significantly more nail biting and object sucking in children with CL/P; present study did not show these associations. This may be explained by differences in inclusion criteria and terminology. More specifically, we included a broader age range (1-12 years vs. 3-6 years) and expanded the definition of 'biting' to nail, tongue or cheek biting instead of nail biting only. Furthermore, cultural (e.g.

acceptance towards habits), sociodemographic and socioeconomic differences may explain the contrasting results [8, 9].

No study has yet investigated the occurrence of accompanying habits (i.e. drooling, snoring and bruxism) in children with CL/P, nor the difference when compared to children without CL/P. Present study revealed a higher occurrence of drooling, although not statistically significant, in children with CL/P [Table 1]. The higher occurrence of drooling may result from the swallowing problems children face before (and after) their surgeries [16]. Another possible explanation for the higher occurrence of drooling is the perioral stiffness associated with the scarred upper lip and altered sensation [16]. Furthermore, difficulties in lip coordination can possibly cause difficulties in orofacial function thus resulting in drooling. As drooling often results in social-emotional problems in children [17], clinicians should be aware of higher occurrence of this habit in children with CL/P and its possible consequences for the child and his/her environment. The impact of drooling on quality of life in children with CL/P is a topic for further research. Additionally, SLP's and pediatric dentists can help these children by increasing their lip mobility, lip strength and sensibility.

Adenoid hypertrophy can cause symptoms such as snoring [18]. In children with CL/P, adenoidectomy is often avoided to prevent aggravation of speech problems [19]. In the present study, adenoidectomy was performed in none of the children with CL/P. This may be a possible explanation for the statistically significant higher occurrence of snoring in these children. The presence of midfacial hypoplasia in children with CL/P resulting in a more narrow pharyngeal airway might also be responsible for impairment of breathing and occurrence of snoring [20] [21]. However no statistically significant difference in oral habits (e.g. open mouth breathing/open mouth behavior) was found in the current study and this was in contrast with the hypothesis. A limitation of present study was that only a self-report questionnaire was used. Parents do not always have a full and accurate idea of the occurrence of open mouth breathing and open mouth behavior nor the difference between these two habits. Therefore, future studies may include an orofacial myofunctional assessment in addition to a dental and jaw examination and the self-report questionnaire.

Another limitation in this pilot study was the small sample size resulting from difficulties in recruiting patients in this specific population. A heterogeneous

population of patients with CL/P was included making it impossible to draw conclusions per cleft type. A possible solution for this problem would be to stratify patients by the type of cleft in order to draw conclusions of more homogenous groups in future studies. Furthermore, no information about socioeconomic status (SES), sociodemographic [8] and socioeconomic [9] aspects was included in the present study. More specifically, as the secondary education level of parents is related to habit persistence [15], we suggest to include this information in further studies. Moreover, the development of a validated questionnaire to assess the occurrence of oral habits and non-nutritive sucking habits in a more reliable way, is a topic for further research. A possible solution is setting up focus groups to contribute to the development of the questionnaire [22]. First, the focus group data can be the foundation for the questionnaire (e.g. wording, item development) [22]. Second, focus groups can provide an understanding of what the research project means to members of the study population [22].

To the best of our knowledge, this pilot study was one of the first conducted on the topic of oral habits in children with CL/P. Findings of present study in children between 1-12 years show the need of further research on this topic and additionally, this report contributes to the limited literature documenting oral habits in this specific population.

Conclusion

Present pilot study revealed that the occurrence of non-nutritive sucking habits (i.e. use of a pacifier) was significantly lower in children with CL/P when compared to children without CL/P. Additionally, it was found that children with CL/P showed a significantly higher occurrence of drooling and snoring when compared to children without CL/P. Although further research regarding oral habits in children with CL/P is necessary, ear, nose and throat specialists and speech-language pathologists should give attention to the possible prevalence of oral habits when diagnosing or treating this specific population.

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