FACTORS ASSOCIATED WITH BREASTFEEDING ADHERENCE IN INFANTS WITH OROFACIAL CLEFTS: A SCOPING REVIEW*

HIGHLIGHTS
1. The anatomical complexity of the cleft influences breastfeeding.
2. The absence of negative intraoral pressure is the main limiting factor.
3. Most of the limiting factors are amenable to intervention.

ABSTRACT
Objective: To map the factors associated with breastfeeding adherence in infants with orofacial clefts. Method: Scoping review according to the recommendations of the Joanna Briggs Institute, with a search carried out in March 2023 in the PubMed, CINAHL, Web of Science, SCOPUS, EMBASE, LILACS, ProQuest and BDTD databases. Results: A total of 31 documents were selected. Factors associated with adherence to breastfeeding included: anatomical complexity of the cleft, pre- and post-natal guidance/training, desire to breastfeed, previous experience, knowledge of the benefits of breastfeeding, use of palatal obturators, use of feeding tube, social, family, and professional support, milking and offering by utensils and early performance of cheiloplasty. Conclusion: Although the complexity of the cleft is a factor that cannot be changed early on, the other variables that influence adherence to breastfeeding can be intervened upon. Thus, the support of all those involved in the breastfeeding process is fundamental for its optimization.

KEYWORDS: Breastfeeding; Cleft palate; Cleft lip; Infants; Review.
INTRODUCTION

Breast milk is the best and most complete food and the main source of nutrition for infants. In this sense, the World Health Organization (WHO) recommends its exclusive use until six months of age.

It is defined as the supply of milk straight from the breast or milked, and is classified as exclusive, predominant, supplemented or mixed. It provides numerous benefits to the infant and mother, including reduced morbidity and mortality, financial burdens, prevention of malnutrition and obesity, strengthening the bond between mother and child, development of the stomatognathic system, quality of sleep, environmental sustainability due to the reduction in the use of packaging, as well as reducing the occurrence of infectious diseases due to its immunological properties.

However, numerous factors are associated with adherence to this practice, including the mother's previous experience, the desire to breastfeed, receiving qualified guidance during prenatal and postnatal care, knowledge about the benefits of breast milk, the mother's age and work activity, sufficient milk production, social and family support, among others.

In addition to these, there are conditions related to infants, such as malformations, including orofacial clefts, which can affect the lip, alveolar ridge, and palate, either alone or in combination. In fact, individuals with this malformation are 18 times more likely not to be breastfed than those without the anomaly.

In short, it is an anatomical discontinuity that hinders lip sealing and the establishment of negative intraoral pressure, which are essential for proper suction. However, even if the limitations are recognized, they can be worked around, meaning that breastfeeding is possible and desirable in infants with this malformation, especially in less complex cases.

It should be noted, however, that direct breastfeeding is contraindicated in cases where orofacial clefts are associated with syndromes, usually due to impaired respiratory and neurological functions and oropharyngeal dysphagia. In these cases, breast milk should be milked and given via the stomach.

In summary, knowledge of the factors associated with breastfeeding adherence can support interventions and public policies to encourage, protect and support this practice, since breastfeeding and the use of human milk should be prioritized and supported so that infants with orofacial clefts can fully enjoy its benefits.

Therefore, this study sought to map the factors associated with adherence to breastfeeding in infants with orofacial clefts.

METHOD

A scoping review, developed and structured according to the recommendations of the Joanna Briggs Institute (JBI) and the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA-ScR), registered on the Open Science Framework (OSF) platform under DOI: 10.17605/OSF.IO/359HC.

This research was constructed in five stages: formalization of the research question, identification of relevant studies, selection of studies, data analysis, synthesis, and results presentation.
In order to identify the existence of records of similar reviews, a preliminary search was carried out in the electronic databases: OSF, Figshare, JBI and The Cochrane Library, in which no similar studies were found.

The research question considered the population, the context and the concept, i.e. it was developed from the acronym PCC, where the population consisted of infants with orofacial clefts; the context, breastfeeding; and, as a concept, the factors associated with adherence. Thus, the following question was posed: what factors are associated with breastfeeding adherence in infants with orofacial clefts?

The search was carried out with the help of a librarian in the following databases: National Library of Medicine and National Institutes of Health (PubMed), Cumulative Index to Nursing and Allied Health Literature (CINAHL), Web of Science, SCOPUS, EMBASE and Latin American and Caribbean Health Sciences Literature (LILACS). For the gray literature, the electronic databases were consulted: ProQuest Dissertations & Theses Global and the Brazilian Digital Library of Theses and Dissertations (BDTD). The descriptors extracted from DeCS/MeSH were used: breastfeeding, cleft lip and cleft palate, in English and Portuguese, with their respective synonyms, as well as the AND and OR Boolean operators.

Documents available in full in Portuguese, English or Spanish were included, which covered the concept of breastfeeding advocated by the WHO, whose target audience includes children aged between zero and 24 months, with isolated orofacial clefts (of the lip, palate or lip and palate), i.e. without association with clinical syndromes or comorbidities (neurological, metabolic, renal, respiratory, or cardiovascular problems). No time limit was set. Editorials, theoretical essays, blogs, web pages, abstracts published in the proceedings of scientific events and books were excluded. The search took place in March 2023.

In the analysis of the studies, the first stage consisted of excluding duplicate literature and organizing the documents in temporal order. Based on the research question and inclusion criteria, the titles and abstracts were assessed. Subsequently, the full texts were read. This whole process was carried out independently and simultaneously, manually, by two researchers. Divergent cases were analyzed and defined by the supervising researcher.

In order to extract and summarize the results, an instrument was created with the following variables: publication year, country, sample and factors influencing adherence to breastfeeding. The selected studies were tabulated using Microsoft Excel 2013® and analyzed using descriptive statistics.

As this was a methodological study using a secondary source of data, ethical appraisal was waived.

**RESULTS**

Initially, a total of 897 publications were found. After excluding duplicates and analyzing the titles and abstracts, 90 remained to be read in full. Of these, 28 were selected. Three documents were added from the reverse search. Finally, 31 documents were included: 27 articles, two dissertations and two theses (Figure 1).
Some studies carried out in Brazil predominated (n=11, 38%). The oldest was published in 1990 and the most current in 2023. The factors associated with adherence to breastfeeding included: the anatomical complexity of the cleft, pre- and post-natal guidance/training, the desire to breastfeed, previous experience, knowledge of breastfeeding’s benefits, the use of a palatal obturator, the use of a feeding tube, social, family, and professional support, the milking and offering of breast milk by utensils and the early performance of cheiloplasty (Chart 2).
<table>
<thead>
<tr>
<th>Identification</th>
<th>Publication year and country</th>
<th>Sample</th>
<th>Factors related to breastfeeding adherence</th>
</tr>
</thead>
<tbody>
<tr>
<td>ID18</td>
<td>1990, Brazil.</td>
<td>105 mothers of infants with cleft lip and/or palate, aged between zero and 18 months old.</td>
<td>Anatomical complexity of the cleft, guidance and encouragement from professionals, family, and social support.</td>
</tr>
<tr>
<td>ID19</td>
<td>1996, England.</td>
<td>25 mothers of neonates with cleft lip and/or palate</td>
<td>Guidance provided by trained professionals during hospitalization and support after discharge.</td>
</tr>
<tr>
<td>ID20</td>
<td>1997, Japan.</td>
<td>10 infants with cleft lip and palate.</td>
<td>Use of a palatal obturator.</td>
</tr>
<tr>
<td>ID21</td>
<td>2000, Brazil.</td>
<td>Not reported.</td>
<td>Complexity of the cleft, health education and the mother’s desire to breastfeed.</td>
</tr>
<tr>
<td>ID23</td>
<td>2003, Brazil.</td>
<td>200 infants with cleft lip and/or palate.</td>
<td>Anatomical complexity of the cleft.</td>
</tr>
<tr>
<td>ID24</td>
<td>2004, Australia.</td>
<td>55 articles.</td>
<td>Use of a palatine obturator, pre- and post-natal guidance, and adaptive positions.</td>
</tr>
<tr>
<td>ID25</td>
<td>2004, Brazil.</td>
<td>31 infants with cleft lip, lip and palate, and palate.</td>
<td>Anatomical complexity of the cleft, pre- and post-natal guidance by qualified professionals.</td>
</tr>
<tr>
<td>ID27</td>
<td>2010, Brazil.</td>
<td>23 children with cleft lip, palate and/or lip and palate.</td>
<td>Anatomical complexity of the cleft.</td>
</tr>
<tr>
<td>ID28</td>
<td>2010, Thailand.</td>
<td>20 infants with cleft lip and palate.</td>
<td>Pre- and post-natal counseling by qualified professionals and family support.</td>
</tr>
<tr>
<td>ID29</td>
<td>2011, Brazil.</td>
<td>137 children with cleft lip and/or palate.</td>
<td>Anatomical complexity of the cleft.</td>
</tr>
<tr>
<td>ID30</td>
<td>2011, Brazil.</td>
<td>215 parents of children with cleft lip and/or palate, from eight institutions.</td>
<td>Anatomical complexity of the cleft.</td>
</tr>
<tr>
<td>ID31</td>
<td>2013, India.</td>
<td>Newborn with cleft lip and palate.</td>
<td>Use of a palatal obturator.</td>
</tr>
<tr>
<td>ID34</td>
<td>2017, United States.</td>
<td>110 infants with isolated cleft lip or lip and palate.</td>
<td>The desire to breastfeed, pre- and post-natal guidance and professional support.</td>
</tr>
<tr>
<td>ID35</td>
<td>2017, Czech Republic.</td>
<td>104 newborns with isolated cleft lip or lip and palate.</td>
<td>Professional support and early cheiloplasty.</td>
</tr>
</tbody>
</table>
Several studies\textsuperscript{6,10,18,21,23,25-27,29-30,37-38,40-42,45} have shown that infants with cleft lip do not have any difficulties in breastfeeding directly. On the other hand, those with cleft palate, or cleft lip and palate, had significant difficulties, linked to the absence of lip sealing and, above all, the formation of negative intraoral pressure, which is considered essential for successful breastfeeding\textsuperscript{7,12,43}.

\[\text{DISCUSSION}\]

\begin{table}[h]
\centering
\begin{tabular}{|c|c|c|}
\hline
ID\textsuperscript{35} & 2017, Brazil. & 150 mothers of children with cleft lip and/or palate. & Guidance received during the prenatal period. \\
\hline
ID\textsuperscript{6} & 2018, Brazil. & 121 caregivers of children with isolated cleft lip, lip and palate and palate. & Cleft complexity and the information received by health professionals during prenatal care. \\
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ID\textsuperscript{36} & 2020, India. & 32 informal caregivers of infants with unilateral or bilateral isolated cleft lip and palate, aged between zero and six months. & Orientation/training through audiovisual education modules. \\
\hline
ID\textsuperscript{37} & 2020, United States. & 150 biological mothers of infants aged between eight and 14 months who had an isolated unilateral or bilateral cleft lip and palate. & Anatomical complexity of the cleft, prenatal guidance/training, and supply of milked milk. \\
\hline
ID\textsuperscript{38} & 2020, Nigeria. & 65 mothers of infants with isolated cleft lip or lip and palate, aged between one and 16 months. & Anatomical complexity of the cleft, pre- and postnatal orientation/training. \\
\hline
ID\textsuperscript{39} & 2020, Chile. & 36 infants with isolated cleft palate, aged between zero and six months. & Knowledge of breastfeeding benefits, use of feeding tubes, breast milk milking and professional support. \\
\hline
ID\textsuperscript{40} & 2020, Brazil. & 162 caregivers of children with cleft lip; lip and palate; and isolated palate. & Anatomical complexity of the cleft. \\
\hline
ID\textsuperscript{41} & 2021, Brazil. & 210 children with isolated cleft lip; lip and palate; or palate. & Anatomical complexity of the cleft. \\
\hline
ID\textsuperscript{42} & 2021, South Africa. & 32 children with cleft lip, lip and palate or palate in isolation, aged between zero and 24 months. & Anatomical complexity of the cleft, milking and supply using utensils, family, and social support. \\
\hline
ID\textsuperscript{43} & 2021, United Kingdom. & A child with an isolated unilateral cleft lip and palate, monitored from birth to 11 months. & The desire to breastfeed, previous experience of breastfeeding, social and professional support. \\
\hline
ID\textsuperscript{44} & 2022, Indonesia. & 11 articles. & Use of a palatine obturator and baby bottles. \\
\hline
ID\textsuperscript{45} & 2022, Colombia. & 28 mothers of children with unilateral, non-syndromic cleft lip and/or palate. & The mother's desire to breastfeed. \\
\hline
ID\textsuperscript{10} & 2023, Brazil. & 29 studies. & Anatomical complexity of the cleft. \\
\hline
\end{tabular}
\caption{Factors associated with breastfeeding adherence in infants with orofacial clefts: A scoping review.}
\end{table}
In this direction, some strategies have favored adherence to breastfeeding, including adaptive positions, mainly facilitating lip sealing. In fact, changing the children’s position was the way that mothers most often used to breastfeed their children with clefts. These positions can be taught by health professionals or emerge from the mothers’ experience.

In children with cleft palate, although lip sealing and sucking are preserved and there appears to be an effective breastfeeding process, adequate intraoral pressure is not formed. Therefore, special attention should be paid to excessive swallowing of air, frequent regurgitation, satiety and, above all, weight gain and growth, which should be age appropriate. In addition, for children with more complex clefts, such as those of the lip and palate, the milking of breast milk and feeding through utensils, such as a bottle, should be encouraged.

In fact, breastfeeding directly at the breast is not always possible, and one of the alternatives is to express milk by hand or electric pump, offered by cup, spoon or bottle. In this situation, although the benefits in terms of strengthening the stomatognathic system are not possible, others, such as nutritional and immunological benefits, are guaranteed. Various studies have shown that milking promotes breastfeeding.

In addition, the extraction of breast milk, by manual or electronic milking, favors production through the sucking stimulus, as well as promoting comfort, physical and psychological well-being for the mother. However, they need to be properly trained, supported, and monitored prospectively.

Guidance, training, and support from experienced health professionals, received during prenatal care, postnatal care, or both, positively influenced adherence to breastfeeding. To this end, different approaches have been used, including the use of audiovisual tools such as videos, books, and pamphlets, as well as educational courses and technologies.

Guidelines include assessing the infant’s ability to suck, demonstrating proper breastfeeding positioning, supporting mothers to establish and maintain their milk supply, considering adaptable feeding equipment such as specialized bottles and nipples, and raising awareness about the benefits of breast milk. However, knowledge about orofacial clefts is scarce among health professionals, making it a challenge to overcome.

In addition to support from experienced health professionals, it was found that social, family, and spousal support were essential for breastfeeding adherence and maintenance. In fact, support favors breastfeeding, including lactation consultants, nurses, feeding therapists and support groups with remote meetings.

In another study carried out in Uganda, mothers said that support from family members, communities and hospitals was essential. The fact is that in addition to the anatomical difficulties in breastfeeding, those involving social, emotional, economic, and cultural relationships must also be taken into account.

In addition, the mother’s desire to breastfeed and knowledge of the breastfeeding benefits influenced adherence. These variables are made up of previous successful experiences, quality information acquired throughout life, before and after the birth of the infant. One of the main information tools is the internet, so it is important that websites, apps, and other educational materials contain quality information. In fact, mothers with previous positive experiences, both personal and familial, tend to breastfeed again.

Some studies have described the benefits of using palatal obturator plates. In one case report, the use of an acrylic plate improved the infant’s ability to suck; however, the...
design was modified with the placement of an orthodontic wire in order to retrieve the obturator intraorally in the event of airway obstruction.

It should be noted, however, that the bottle itself is the utensil most often used to feed children with clefts, especially those with long, soft nipples, which favor sucking without the need for negative intraoral pressure, thus dispensing with the use of a palatine prosthesis. In another study carried out in Japan, the palatine obturator was used to feed 10 infants with cleft lip and palate; however, supplementary bottle feeding was necessary to provide sufficient nutrition, meaning that the effectiveness of the palatine prosthesis was limited.

Furthermore, the use of an acrylic feeding plate was considered useful by only one of nine mothers of infants with isolated cleft palates, for whom the benefits of creating negative intraoral pressure would be more desirable.

Among the limitations of using palatine prostheses are the difficulty of fixation, adaptation, asphyxiation risk and the need for periodic changes, considering the growth of facial and intraoral structures. In summary, to date there has been limited scientific evaluation of the use of palatal prostheses to support breastfeeding in newborns with clefts.

Among the factors that negatively influenced adherence to breastfeeding was the use of a feeding tube. For children with isolated orofacial clefts, i.e. without associated clinical syndromes or comorbidities, oral feeding is advocated from birth, as the necessary reflexes are preserved. In this sense, the use of tube feeding is associated with misinformation.

In summary, the use of tube feeding without precise indications makes breastfeeding unfeasible, as the transition time to oral feeding is long and costly. In addition, feeding methods other than the usual ones cause high levels of stress for parents, especially mothers, which can even influence breast milk production and the desire to breastfeed.

Early cleft lip repair surgery, cheiloplasty, was reported as a factor associated with breastfeeding adherence, while breastfeeding adherence in infants with cleft lip and palate remained low even after surgery. In a study carried out in Africa, of 36 neonates referred to a referral hospital, 23 could not be satisfactorily breastfed and underwent cheiloplasty. However, the main benefit was improved maternal confidence and interaction with the baby, rather than adherence to breastfeeding per se.

Although there is no consensus on the ideal age for lip repair surgery, it is widely used from the age of three months, taking into account certain factors such as facial growth. In addition, an isolated cleft lip does not contraindicate breastfeeding, including direct breastfeeding.

In summary, performing cheiloplasty strictly to benefit breastfeeding should be viewed with caution, considering that every surgical procedure has risks, especially for newborns. In addition, performing cheiloplasty early can limit maxillary growth, leading to the need for other surgical interventions later on.

Although this study used the main databases and the classic language of the publications, the existence of other databases, as well as investigations in languages other than those considered, are limitations.

**CONCLUSION**

The factors associated with adherence to breastfeeding in infants with orofacial clefts included: the anatomical complexity of the cleft, pre- and post-natal guidance/training, the desire to breastfeed, previous experience of breastfeeding, knowledge of the breastfeeding
benefits, the use of palatal obturators, feeding tubes, professional, social and family support, milking and offering by utensils, and early cheiloplasty.

Although the complexity of the cleft is a factor that cannot be changed early on, the other variables that influenced adherence to breastfeeding can be intervened upon. In short, it was clear how much health professionals can do to encourage the practice of breastfeeding in children with orofacial clefts.

The contributions of this research are evident and include a detailed and in-depth report on the factors associated with breastfeeding adherence in infants with orofacial clefts, which could serve as a basis for interventions, protocols and/or public policies aimed at encouraging this practice in this specific population, for whom the benefits of breast milk are essential for establishing an effective rehabilitation process. Future research testing the effectiveness of interventions through properly controlled studies is essential.

REFERENCES


Factors associated with breastfeeding adherence in infants with orofacial clefts: A scoping review

Batista NT, Antonio CT, Bom GC, Farinha FT, Von Kostrisch LM, Mazzo A, Trettene A dos S


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Associate editor: Dra. Tatiane Trigueiro
Corresponding author:
Armando dos Santos Trettene
Hospital de Reabilitação de Anomalias Craniofaciais, Universidade de São Paulo
E-mail: armandotrettene@usp.br

Contribuição dos autores:
Substantial contributions to the conception or design of the work; or the acquisition, analysis, or interpretation of data for the work - Antonio CT, Bom GC, Farinha FT, Trettene A dos S. Drafting the work or revising it critically for important intellectual content - Von Kostrisch LM, Mazzo A, Trettene A dos S. Agreement to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved - Batista NT, Trettene A dos S. Orientation - Trettene A dos S. All authors approved the final version of the text.

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