

Relationship between Ankyloglossia and Breastfeeding: A Bibliometric Review

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ABSTRACT

Objective: To assess global trends in the publication of studies investigating the association between ankyloglossia and breastfeeding. **Material and Methods:** An electronic search was performed in the Scopus database without restrictions. Observational studies and clinical trials were included. Bibliometric indices such as publication year, authors, co-authors, journals, field of knowledge, countries, and the most cited keywords were analyzed using the VOSviewer program. **Results:** The search retrieved 350 studies, and 68 were selected. The first article was published in 2000 in the United States. The United States presented the highest number of publications (n=21), followed by Brazil (n=9) and the United Kingdom (n=9). An increase in publications on this theme was observed in 2013; 2021 was the year with the highest number of publications (n=14). The most common word was “frenulum”. The authors with the highest number of publications were Botze and Dollbert from Israel (n=3), Ghaheri, and Mace from the United States (n=3). Among the journals, “Breastfeeding Medicine” presented the highest number of publications (n=7), followed by the “International Journal of Pediatric Otorhinolaryngology” (n=6), “CODAS” (n=5), “Journal of Human Lactation” (n=4) and “Pediatrics” (n=3); the latter published the top-cited studies, with 412 citations. **Conclusion:** There has been an increase in recent articles evaluating the correlation between ankyloglossia and breastfeeding, indicating the growing interest of researchers in this field.

Keywords: Breast Feeding; Ankyloglossia; Lingual Frenum; Infant, Newborn.

Introduction

For years, whether ankyloglossia can negatively interfere with newborn breastfeeding has been discussed in the literature; however, no consensus has been reached [1-4]. The literature suggests that frenulum function guides the movement of the tongue to allow swallowing, speaking, and phonation; therefore, a frenulum with an altered insertion could limit the mobility of the tongue, resulting in breastfeeding problems such as pain in the breast and cracked or ulcerated nipples [5].

The classification of the lingual frenulum is based on anatomy, which is still very subjective, so several instruments have been created to standardize this classification. In 1993, an instrument for the assessment of the lingual frenulum was designed before being modified in 2010, known as the Hazelbaker [6] Assessment Tool for Lingual Frenulum Function (HATLFF). In 2015, based on the HATLFF and daily clinical practice, the Bristol Tongue Assessment Tool (BTAT) was developed, which proposed a simplified method of identifying ankyloglossia containing four items. This method demonstrated measuring the severity of a tongue-tie with a solid correlation to HATLFF, which is also available in English [7]. Recently, Ingram et al. included a version of this tool containing 12 images of the tongue, its attachment to the gum, and the limits of tongue mobility, known as the Tongue-tie and Breastfed Babies (TABBY) assessment tool, which was based on BTAT [8]. In 2012, in Brazil, a protocol for the Assessment of the Frenulum of the Tongue of Babies was created to identify frenula that could present tongue-tie, leading to limitations of tongue movements, preferably in the first 48 hours of life [9,10]. Based on this protocol, neonatal screening of the frenulum became mandatory in public and private Brazilian maternity hospitals and hospitals through Law No. 13.002/2014 [11].

Since 2016, the technical-scientific report of the Ministry of Health of Brazil [12] adopted the use of the Bristol Protocol as a recommendation, modified by images developed in partnership with British researchers [8,13], considering that there is no gold standard instrument for ankyloglossia diagnostic testing. In addition, in 2018, they added that the assessment of breastfeeding is essential, together with the evaluation of the tongue, to determine the existence of ankyloglossia that may be associated with breastfeeding difficulty and, based on this information, to determine the need for surgical intervention, this recommendation was confirmed in the current technical-scientific report [12,14].

Even though the classification is subjective and there is no gold standard protocol for the diagnosis of ankyloglossia, this test for neonatal screening has been mandatory in Brazil since 2014; to the best of our knowledge, in countries such as the United States and the United Kingdom, the tests are recommended only in the case of difficulties with breastfeeding [8,15]. However, there are references in the literature to suggest that frenotomy/frenectomy can improve some aspects that may be related to breastfeeding in children with ankyloglossia [16-19]. No scientific evidence supports the benefit of breastfeeding or the child's weight gain in this practice [1,5,17,20-25].

Due to the mandatory assessment of tongue-tie in newborns in Brazil [26] and the still controversial association between ankyloglossia negatively interfering with breastfeeding, the objective was to carry out a bibliometric review of the literature to research the publication trends on the theme to date to gather data on academic production and publications on the subject.

Material and Methods

Search Strategy

A systematic search strategy was developed and applied in the Scopus database until Feb 2022, without any restrictions. The search strategy used a combination of MeSH and the free terms most frequently cited in

the published studies referring to “ankyloglossia,” “breastfeeding,” and its variants. A preliminary search in the specialized literature was used to determine the terms related to the topic. The following MeSH and free terms were used: “breast-feeding,” “breastfeeding,” “lactation,” “lactating,” “ablactation,” “breastfeed,” “ankyloglossia,” “tongue tie,” and “lingual frenulum.” Complete strategies for literature search syntax rules are described in Table 1. The Boolean operators (“OR” and “AND”) were applied in the syntax rules of the Scopus database.

Table 1. The search strategy used in the electronic search.

Database	Search Strategy	Total
Scopus	(TITLE-ABS-KEY (breast-feeding) OR TITLE-ABS-KEY (breastfeeding) OR TITLE-ABS-KEY (lactation) OR TITLE-ABS-KEY (lactating) OR TITLE-ABS-KEY (ablactation) OR TITLE-ABS-KEY (breastfeed) AND (TITLE-ABS-KEY(Ankyloglossia) OR TITLE-ABS-KEY(tongue tie) OR TITLE-ABS-KEY(lingual frenulum))	350

The studies retrieved by the search were exported in BibTeX format to the Mendeley reference manager program to apply eligibility criteria. At this stage, the selection was performed by two independent researchers (DNS and TKSF) who selected the studies by titles and abstracts. In the second stage, the studies were fully accessed for the final decision on their inclusion. In cases where a consensus decision was not reached, a third researcher was consulted (F.B.F.).

Observational studies and clinical trials evaluating the relationship between ankyloglossia and breastfeeding were included without restricting language, date, or publication status.

Studies unrelated to the topic and articles that used secondary data to obtain results were excluded. In addition, comments, abstracts, animal studies, case reports, case series, opinion articles, letters to the editor, and studies with participants with systemic diseases were excluded.

Data Analysis

The records were exported to the VOSviewer program (Version 1.6.16) [27] for data mining. The main results of the bibliometric analysis describe information such as the number of articles, citations, authors, institutions, countries, sources, and keywords. Collaboration analysis was used to identify co-authorship and determine author collaboration networks. Keyword co-occurrence analysis was performed to map and group the terms extracted from the keywords of the analyzed collection. In addition, the field of knowledge related to the topic was also extracted.

Results

A total of 350 studies were retrieved from Scopus. After applying the eligibility criteria, 71 articles were selected. After reading the complete text, 68 remained in the bibliometric revision, of which 49 (72%) were observational studies and 19 (28%) were clinical trials.

Figure 1 shows that the first studies appeared in 2000, with one publication, and there was a more significant trend of publications from 2013, with the first Brazilian article that composed this search published in 2015. In 2020, there was a more substantial number of publications on the topic (n=10), with 2021 being the year with the highest number of publications (n=14). In the first two months of 2022, 2 studies have been published, reinforcing the increasing publication trend. Most articles were published in medicine journals, followed by nursing and dentistry, as listed in Table 2.

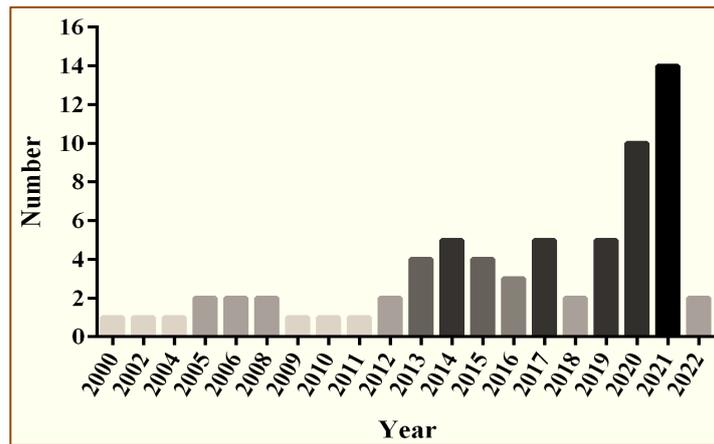


Figure 1. Distribution of publications per year.

Table 2. Distribution of articles by publication area.

Subjective Area of Publication	N (%)
Medicine	64 (94.1)
Nursing	13 (19.1)
Dentistry	9 (13.2)
Arts and Humanity	5 (7.3)
Speech Therapy	2 (2.9)
Environmental Science	2 (2.9)

Among the journals that are published most often are medical journals, with Breastfeeding Medicine (n=7) being the one with the highest number of publications, followed by the International Journal of Pediatric Otorhinolaryngology (n=6) (Table 3). Many journals only have a single publication (45.9%). Regarding the number of citations, the journal Pediatrics has the highest number (n=412).

Table 3. Top journals according to the number of publications.

Magazine	N (%)
Breastfeeding Medicine	7 (10.3)
International Journal of Pediatric Otorhinolaryngology	6 (8.8)
CODAS	5 (7.3)
Journal of Human Lactation	4 (5.9)
Pediatrics	3 (4.4)
Clinical Oral Investigation	2 (2.9)
European Journal of Pediatric Dentistry	2 (2.9)
International Journal of Environmental Research and Public Health	2 (2.9)
Journal of Pediatric Surgery	2 (2.9)
Laryngoscope	2 (2.9)
Otolaryngology Head and Neck Surgery United States	2 (2.9)
Other journals	31 (45.9)

The United States had the highest number of publications (n=21), followed by Brazil (n=9) and the United Kingdom (n=9) (Table 4).

Table 4. Distribution of publications by country.

Country of Publication	N (%)
United States	22 (32.3)
Brazil	9 (13.2)
United Kingdom	9 (13.2)

Canada	5 (7.3)
Israel	5 (7.3)
Spain	5 (7.3)
Australia	4 (5.9)
Thailand	4 (5.9)
Austria	1 (1.4)
Croatia	1 (1.4)
Germany	1 (1.4)
Ireland	1 (1.4)
Lithuania	1 (1.4)
Netherlands	1 (1.4)
Peru	1 (1.4)
New Zealand	1 (1.4)
Italy	1 (1.4)
Switzerland	1 (1.4)
Turkey	1 (1.4)

Turkey was the country that first published on the topic, but countries like Australia, Brazil, Lithuania, and Peru have a more recent trend of publications. The predominant language of publications was English, with 97% (n=66), followed by Portuguese with 5.9% (n=4), Spanish with 2.9% (n=2) and Bosnian with 1.4% (n=1); the articles in Bosnian and Portuguese are also in English.

Figure 2 shows us the word cloud containing the co-occurrence authors' keywords related to the themes presented one or more times. It is possible to observe the grouping of words into twenty related clusters.

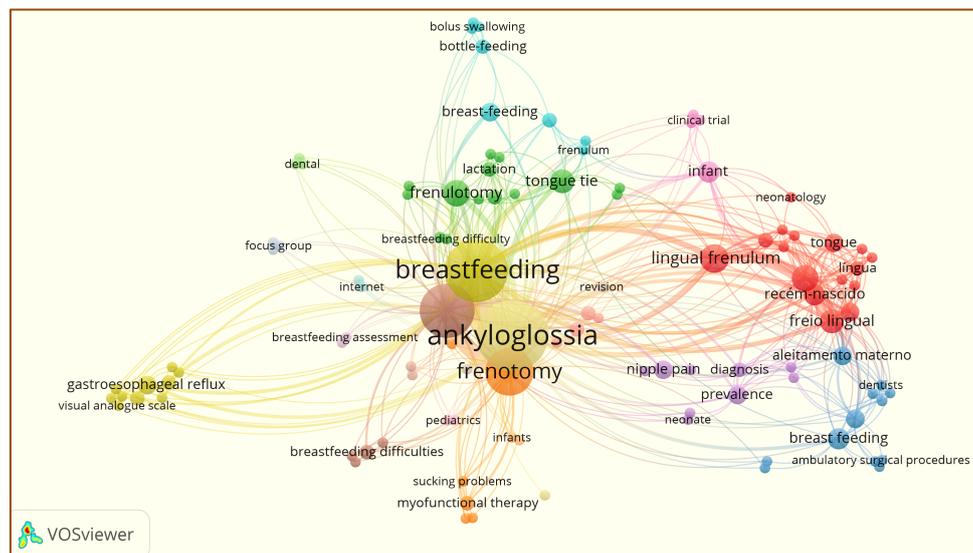


Figure 2. Word cloud containing terms related to the topic repeated twice or more.

The author with the highest number of publications was Botze and Dollbert (n=3), from Israel, who are co-authors with one publication in 2006 and two in 2014, and Ghaheri and Mace (n=3), from the United States, who are co-authors with publications in 2017, 2018 and 2021 (n=3).

Figure 3 shows the map containing documents related to bibliographic coupling. It is possible to observe that previously published manuscripts, such as Messner et al. [28], are more commonly cited by recently published studies. The lines illustrate these documents' strength and connection, demonstrating a common theme.

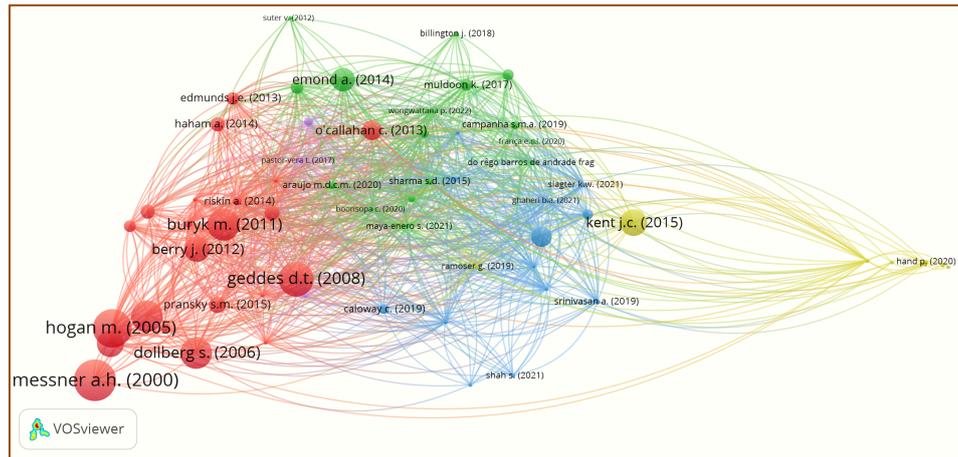


Figure 3. Map containing the authors and their networks.

Discussion

The present study shows that the publications related to ankyloglossia and breastfeeding began in 2000 in Turkey, where a study was carried out to assess the clinical relevance of ankyloglossia in breastfeeding. In this study, there was no difference in the duration of breastfeeding between mothers who breastfed babies with and without ankyloglossia [28]. Although they did not appear among the authors that publish the most on this topic, they published a recent article to establish a clinical consensus on ankyloglossia in children, which concluded that babies with ankyloglossia would not necessarily present difficulties with breastfeeding and the diagnosis of ankyloglossia itself should not be indicative of surgery in the lingual frenulum [15].

Regarding the country of publication, the United States appeared to have the highest number of publications, accounting for 32.3%. The United Kingdom and Brazil were the second most prolific, with 13.2%. The English language was the most predominant among the publications. This finding may be related to the fact that the United States holds most publications in the area, especially as English is the most spoken language in the world. In addition, this is the most widely accepted language for scientific communication [29], with around 92.64% of the content indexed in SCOPUS using English for scientific publication [30]. The present study showed that five groups interact with each other in publications on the subject. Networked work in this area can integrate different expertise, providing a satisfactory sample size and bringing external validity when the research subjects come from different centers. The present study demonstrated a recent trend of countries such as Australia, Brazil, Lithuania, and Peru publishing on the subject. In Brazil, specifically, there was a more significant trend of publications from 2015, a fact that is probably explained by the implementation of Federal Law 13,0002/14 on 06/20/2014 [11], where neonatal screening of the lingual frenulum became mandatory in maternity hospitals and Brazilian public and private hospitals based on the tongue-tie test [26]. However, the present bibliometric review demonstrates a limited number of published studies to support the Federal Law. The most recent technical-scientific report from the Ministry of Health of Brazil [12] indicates language assessment through the Bristol Protocol [7,8] since no gold standard test exists. Thus, the Ministry of Health of Brazil adopted this protocol as a validated, rapid, simple test, and the images help professionals with the diagnosis.

From 2013 onwards, an increase in the number of publications was observed, with 2021 being the year with the highest number of publications [31] and showing a preoccupation with the exponential increase in the number of lingual frenotomy surgeries in recent years; this increase in the number of surgeries may justify the growing number of studies on the subject, aiming to gather scientific evidence that supports or refutes this surgical practice.

The results of this bibliometric review show that Medicine appropriated the theme and constituted the area that most produced investigations related to the theme, with 94.1% of publications in the area. However, assessment of the lingual frenulum can be performed by other healthcare professionals such as dentists, speech therapists, and nurses [26]. Dentistry ranks third, with 13.2% of publications. Consequently, medical journals had a higher publication rate, with the journal “Breastfeeding Medicine” accounting for 10.3% of publications on the subject. The word cloud showed a grouping around seven themes, and, as expected, the term “frenulum” is the one that appears most frequently. Other related words appeared, such as nipple pain, lactation, breastfeeding difficulties, ambulatory surgical procedures, and neonatology. Unexpected words included bolus swallowing, myofunctional therapy, and gastroesophageal reflux.

When evaluating the types of studies among the 68 articles in this review, we saw a higher percentage of observational studies (72%), with clinical trials in the first ten years of publication and observational studies more recently. Although the evidence pyramid shows us superiority in randomized clinical trials, this superiority in observational studies will show us the difficulty in carrying out well-designed clinical trials on the subject [32]. Among the suggested challenges, the need for a multidisciplinary team and the team's training concerning the assessment scales of the lingual frenulum are mentioned. Another difficulty would be confounding factors such as attachment and milk flow, essential variables for the study. In addition, there is a need to assess actual outcomes, such as weight gain or exclusive breastfeeding duration, rather than surrogate outcomes, such as improved breastfeeding and pain relief.

Even with the growing number of studies on the association between ankyloglossia and breastfeeding, the consensus that ankyloglossia can negatively interfere with breastfeeding has not yet been established [1-4]. When evaluating the results of the 68 documents that comprised this review, it was noted that ankyloglossia can be associated with pain during breastfeeding. However, it is noteworthy that the present study was designed as a bibliometric review, so it does not aim to summarize the results and evaluate the methodological quality of the studies obtained. In this sense, our assessment was restricted to bibliometric data related to this field.

Conclusion

There is an increase in recent articles evaluating the correlation between ankyloglossia and breastfeeding, indicating a growing interest of researchers in this correlation.

Authors' Contributions

DNS	 https://orcid.org/0000-0002-9131-283X	Conceptualization, Methodology, Validation, Formal Analysis, Investigation, Data Curation, Writing - Original Draft, Writing - Review and Editing, Visualization and Funding Acquisition.
FBF	 https://orcid.org/0000-0002-4098-8637	Conceptualization, Methodology, Validation, Investigation, Resources, Data Curation, Writing - Original Draft, Writing - Review and Editing, Visualization, Supervision and Project Administration.
TKSF	 https://orcid.org/0000-0003-1340-9967	Conceptualization, Methodology, Validation, Formal Analysis, Investigation, Resources, Data Curation, Writing - Original Draft, Writing - Review and Editing, Visualization, Supervision and Project Administration.

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None.

Conflict of Interest

The authors declare no conflicts of interest.

Data Availability

The data used to support the findings of this study can be made available upon request to the corresponding author.

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