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Published in:
Danish Medical Journal

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Publication date:
2022

Document Version
Publisher's PDF, also known as Version of record

[Link to publication from Aalborg University](#)

Citation for published version (APA):

Siggaard, L. D., Tingsgaard, P., Lüscher, M., Holm, J. P., Nielsen, J. K., & Berrett, T. Q. (2022). Parent-reported infant and maternal symptom relief following frenotomy in infants with tongue-tie. *Danish Medical Journal*, 69(5), [A12210934].

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Original Article

Dan Med J 2022;69(5):A12210934

Parent-reported infant and maternal symptom relief following frenotomy in infants with tongue-tie

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Dan Med J 2022;69(5):A12210934

ABSTRACT

Introduction. A tightening of the lingual frenulum may cause breastfeeding difficulties. Surgical release of the restricted frenulum is accomplished by a frenotomy. Between 2015 and 2019, frenotomy procedures in Danish primary healthcare doubled. Causality has not previously been established. The primary aim of this study was to investigate infant/maternal symptom relief and parent satisfaction following frenotomy and discuss potential causes for the increasing frenotomy frequency in Danish infants.

Methods. Between April 2019 and April 2020, 230 breastfed infants < 12 months had a frenotomy performed in three private ENT clinics. Parents of 163 infants participated in a phone interview.

Results. A moderate to high degree of symptom relief was reported in 138 (85%) infants and 127 (78%) mothers. If more than one preoperative symptom was reported, post-operative infant/maternal symptom relief increased significantly, and maternal symptom alleviation occurred more quickly. Also, infant and maternal symptom relief increased significantly when “infant breastfeeding difficulty” or “maternal nibble/breast pain during breastfeeding” was reported preoperatively. Most parents (95%) would have a frenotomy performed on their child again under similar circumstances.

Conclusions. Most parents reported a moderate to high degree of infant and maternal symptom relief following frenotomy. Parent satisfaction was compelling. A uniform assessment tool may sharpen diagnostic criteria and eventually stabilise the frenotomy frequency in Danish infants.

Funding. none.

Trial registration. not relevant.

Breastfeeding is among the most influential health-promoting activities for infants and mothers owing to its disease-preventing effects [1]. Breastfeeding has been shown to promote both short- and long-term health in infants by reducing the risk of infectious diseases, obesity and diabetes while stimulating cognition and neurological development [2]. Furthermore, breastfeeding reduces the risk of breast cancer, uterus cancer and type 2 diabetes in mothers [2].

When breastfeeding difficulties occur, their causality must therefore be established quickly and efficiently. One cause may be associated with the lingual frenulum – a dynamic, layered structure formed by oral mucosa and

the underlying floor of mouth fascia – which mobilises into a midline fold with tongue elevation and/or retraction [3]. A tightening of the lingual frenulum may potentially cause limited tongue movement and thereby possibly lead to breastfeeding difficulties. The condition is often labelled as ankyloglossia and is casually referred to as tongue-tie (TT). To relieve the infant and mother of the problematic consequences of TT, a frenotomy is usually performed. The procedure is accomplished by stretching the restricted frenulum by lifting of the tongue towards the palate and cutting through the white fascia-like tissue with surgical scissors.



Infant boy (eight days old) with severe ankyloglossia (tongue-tie and breastfed babies assessment score = 0) undergoing lingual frenotomy. Foto: © Jonas Peter Holm, private ENT specialist.

A recent Danish review on treatment of infants with TT reported an 11% prevalence of TT in infants internationally among whom breastfeeding difficulties occurred in 18% [4]. Another recent study reported a significant increase in lingual frenotomy among children treated in Danish hospitals between 1996 and 2015 [5].

A similar trend has been observed in primary healthcare according to data from the Danish National Health Service Registry [2]. Private ear, nose and throat (ENT) specialists performed 5,986 frenotomies in 2019 compared with 2,929 in 2015: a doubling in the course of a five-year period, affecting approximately 10% of Danish infants in 2019. Moreover, data from the Danish Regions regarding reimbursement applications show that an increasing number of parents seek surgical treatment for their child outside of Denmark. The reasons explaining the frenotomy procedure increase in Danish infants have not previously been investigated.

Several studies have investigated the effect of frenotomy in children suffering from TT [6-10] and a significant reduction in maternal breast pain during breastfeeding has been reported shortly after frenotomy. However, evidence of the effect on the child's ability to breastfeed remains sparse [2, 11, 12].

Therefore, the aim of this retrospective cohort study was to investigate infant and maternal symptom relief and parent satisfaction following frenotomy and, based on these results, to discuss possible causes of the increasing

frenotomy frequency in Danish infants.

METHODS

In three Danish private ENT clinics located in Randers, Aarhus and Slagelse, 230 breastfed infants under the age of one year had a frenotomy performed between 1 April 2019 and 1 April 2020. A frenotomy was performed based on symptoms caused by dysfunctional breastfeeding and an individual, objective ENT specialist assessment of the lingual frenulum. Informed consent of the procedure was obtained from the parents. Information regarding known complications such as bleeding, pain and recurrence of symptoms was given preoperatively. The mother, if present, was advised to breast- or bottle-feed immediately after the procedure. The parents were advised to contact the clinic directly in case of post-operative bleeding or signs of infection. All parents were contacted by phone by clinic staff members during April 2020 and invited to participate in a multiple-choice structured phone interview. See Figure 1 for a translated version of the phone interview. The outcome measure “symptom relief” was defined as overall post-operative relief of one or all reported preoperative symptoms.

FIGURE 1 Questionnaire regarding your child's tongue-tie surgery. A translated version of the applied Danish structured phone interview.

Questionnaire regarding your child's tongue-tie surgery	
1) Birthweight: _____ g	
2) Which of the following symptoms did you or your child suffer from prior to the surgery? (mark as many as you like)	
2.1 Infant breastfeeding difficulty	
2.2 Insufficient infant weight gain	
2.3 Infant abdominal pain/flatulence	
2.4 Maternal nibble/breast pain during breastfeeding	
2.5 Other symptoms not further specified	
3) Who raised the suspicion of tongue-tie?	
3.1 Parents	
3.2 Midwife	
3.3 Nurse/Infant healthcare nurse	
3.4 Doctor	
3.5 Someone else	
4) Do you have any other children, who have had a frenotomy performed?	
4.1 Yes	
4.2 No	
5) Did/does your child suffer from other illnesses/conditions?	
5.1 Yes	
If yes, which?	
5.2 No	
6) How satisfied were you with the preoperative information level?	
6.1 Very satisfied	
6.2 Satisfied	
6.3 Not satisfied	
7) Has your child had a frenotomy performed more than once?	
7.1 Yes	
7.2 No	
7.3 Do not recall	
8) Did your child bleed from the mouth after the procedure?	
8.1 Yes	
8.2 No	
8.3 Do not recall	
9) Did you give your child pain medicine after the procedure?	
9.1 Yes	
9.2 No	
9.3 Do not recall	
10) Did your child receive antibiotics after the procedure?	
10.1 Yes	
10.2 No	
10.3 Do not recall	
11) To your recollection, what was the level of discomfort for your child after the procedure?	
1 _____ 2 _____ 3 _____ 4 _____ 5 _____	
Score 1-5 where 1 = little or no discomfort 5 = severe discomfort	
12) How would you rate the effect of the procedure on your child?	
1 _____ 2 _____ 3 _____ 4 _____ 5 _____ Not relevant _____	
Score 1-5 where 1 = no effect at all 5 = almost complete or complete symptom relief Check "Not relevant" if your child did not have any pre-operative symptoms.	
13) If you scored > 1 in Question 12: To your recollection when did the symptom relief set in?	
13.1 Immediately	
13.2a After a few days	
13.2b After one week	
13.2c Later than one week	
13.2d Do not recall	
13.3 Not relevant	
14) How old was your child when you stopped breastfeeding?	
14.1 Approximately: _____ Years _____ Months	
14.2 Still breastfeeding	
14.3 Do not recall	
15) With what you know now, would you choose to have the procedure performed on your child again under similar circumstances?	
15.1 Yes	
15.2 No	
15.3 Unsure	
16) Have you heard of a so-called "deep cut" of the tongue?	
16.1 Yes, I have (if yes see below)	
Yes	
No	
16.1a Have you considered, if your child would benefit from this type of procedure?	
16.1b Did you discuss this type of procedure with the ENT specialist at the preoperative consultation?	
16.1c Have you had this type of procedure performed on your child subsequently?	
16.2 No, I have not	
16.3 Do not recall	
17) Who answered this questionnaire?	
17.1 Mother	
17.2 Father	
17.3 Both parents	
17.4 Someone else	
18) May we contact you again in case of further questions regarding your child's procedure?	
18.1 Yes	
18.2 No	

Notification to the Health Research Ethics Committee for Southern Denmark was not needed (case no. 1-10-72-1-20).

The Biostatistical Advisory Service at Aarhus University Hospital assisted with statistical analyses. χ^2 -test was used for inter-group comparison, and a two-sided p-value < 0.05 was considered statistically significant.

Trial registration: not relevant.

RESULTS

Parents of 163 children consented to participate in the phone interview, corresponding to a 71% response rate. There were 103 (63%) boys in the responder group. The infants in the responder and the non-responder group were similar in terms of gender distribution ($p = 0.94$) and age (ratio of medians 1.14; 95% confidence interval: 0.8-1.6, $p = 0.44$).

In the responder group, 22 infants (14%) had a frenotomy performed more than once in the study period. A total of 35 infants (21%) had comorbidity such as premature birth, reflux syndrome, icterus, congenital heart condition or asthmatic bronchitis. Furthermore, 27 infants (17%) had siblings who had previously needed a frenotomy and 79 infants (48%) were still breastfed at the time of data collection. Questionnaires were answered as follows: mothers answered 141 (87%), fathers 15 (9%), both parents six (4%) and one other (1%).

Parent reports regarding preoperative symptoms, origin of TT suspicion and parent satisfaction with the preoperative information level and overall procedure are presented in **Table 1**. No complications during the procedure were reported according to the patient files nor reported by the parents.

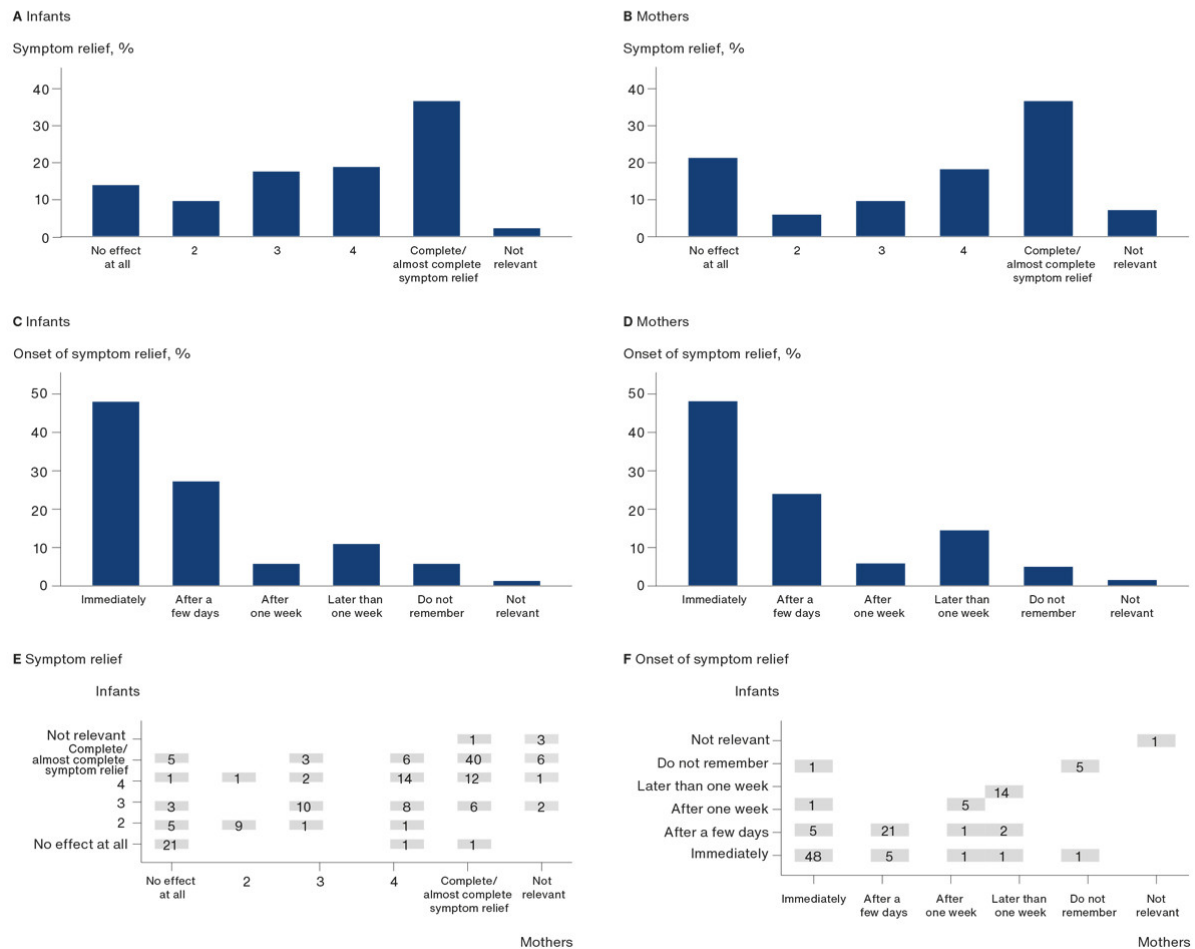
TABLE 1 Parent reports regarding preoperative symptoms prior to surgery, origin of tongue-tie suspicion, post-operative complications and degree of parent satisfaction (N = 163).

	n (%)
<i>Preoperative symptoms^a</i>	
Infant breastfeeding difficulty	110 (67)
Insufficient infant weight gain	33 (20)
Infant abdominal pain/flatulence	41 (25)
Maternal nibble/breast pain during breastfeeding	66 (40)
Other symptoms	67 (41)
<i>Origin of tongue-tie suspicion^a</i>	
Parents	62 (38)
Midwife	16 (10)
Infant healthcare nurse	112 (69)
Doctor	9 (6)
Someone else	9 (6)
<i>Parent satisfaction</i>	
Satisfaction with the preoperative information level:	
Very satisfied	106 (65)
Satisfied	44 (27)
Dissatisfied	13 (8)
Parents who would have frenotomy performed on their child again under similar circumstances:	
Yes	152 (93)
No	8 (5)
Unsure	3 (2)

a) Multiple answers allowed.

In **Figure 2**, histograms illustrate the distribution of parent-reported infant (A) and maternal (B) symptom relief and onset of symptom relief for infants (C) and mothers (D) with symptom relief scores ≥ 2 . In Table E and F, symptom relief and onset of symptom relief scores for infants and mothers, respectively, are plotted against each other. In both cases, the responses are centred around a diagonal line, which implies an equal distribution of scores between infants and mothers ($p < 0.001$).

FIGURE 2 Parent-reported infant and maternal symptom relief (1 = “No effect at all” and 5 = “Complete/almost complete symptom relief”) and onset of infant and maternal symptom relief (reported only for infants/mothers with symptom relief score ≥ 2) following frenotomy are illustrated in A-D. In tables E and F, parent-reported symptom relief and onset of symptom relief scores for infants and mothers, respectively, are plotted against each other.



For further analysis, interview responses were divided into two sets of subgroups comparing interview responses performed less and more than three months after frenotomy, respectively, and interview responses with one or more than one preoperative symptom, respectively. The first analysis adjusting for procedure-to-interview delay showed no significant difference in the distribution of responses between subgroups regarding infant ($p = 0.431$) and maternal ($p = 0.954$) symptom relief or onset of symptom relief in infants ($p = 0.161$) and mothers ($p = 0.206$) after frenotomy. The second analysis regarding number of preoperative symptoms showed a significant relief of symptoms (score 4-5) in both infants ($p = 0.004$) and mothers ($p = 0.001$) when more than one preoperative symptom was reported. Also, maternal symptom alleviation occurred more promptly following frenotomy when more than one preoperative symptom was reported ($p = 0.044$).

Furthermore, parent-reported symptom relief increased significantly in infants when “infant breastfeeding difficulty” was reported preoperatively ($p = 0.005$) and in mothers when “maternal nibble/breast pain during breastfeeding” was reported preoperatively ($p < 0.001$).

DISCUSSION

The main purpose of the study was to investigate infant and maternal symptom relief after frenotomy. Results show that most parents reported a moderate to high degree of infant and/or maternal symptom relief (score 2-5)

after frenotomy. Specific preoperative symptoms such as “infant breastfeeding difficulty” or “maternal nibble/breast pain during breastfeeding” were significantly associated with increased parent-reported symptom relief after frenotomy. This was also the case when more than one preoperative symptom was reported. Approximately half of the infants and mothers with a moderate or high post-operative parent-reported symptom relief (score 2-5) experienced symptom alleviation either instantly or a few days after the procedure. This is in line with previous findings as presented in a Cochrane review from 2017 [11] in which positive immediate effects on infants and mothers following TT surgery were compared in five randomised trials. The review found that maternal nibble pain had been affirmed to decrease significantly following frenotomy, which is a finding also reported in other studies [13, 14]. However, the review also concluded that no consistent improvement in the infants’ ability to breastfeed had been reported, why the gains described following frenotomy were modest. The beneficial effect of frenotomy is therefore a subject for continued discussion.

Another aim was to evaluate overall parent satisfaction. Most of the parents (95%) would have frenotomy performed on their child again under similar circumstances, indicating a high level of parent satisfaction. This is also in line with findings from previous studies in which parents were willing to go to significant lengths to access the procedure [15].

Poor effect/complete absence of effect (score 1) was reported for 23 (14%) infants and 35 (21%) mothers following frenotomy. In the context of the inclining frenotomy frequency as reported by the Danish National Health Service Registry, this may imply some degree of overtreatment of this patient category.

The situation may partly be explained by the growing awareness of TT on social media and online sources, which boast an impressive list of potentially harmful effects of TT in infants such as troublesome breastfeeding, disturbed sleep, reflux, neck pain, colic, irritability, sensitivity, hick-ups, drooling, stomach-ache, respiratory difficulties, thrush, constipation, pickiness towards food, caries, halitosis, uneasiness, snoring, sleep apnoea, recessed chin, crooked teeth, dysregulated nerval system due to an affected vagus nerve and many others. Despite the lack of evidence, parents with breastfeeding difficulties may want to resolve a troublesome situation by releasing the TT, even though TT may in some cases not be the underlying cause. Therefore, the advocacy of potential harmful effects of TT on social media, albeit scientifically unsupported, may motivate parents to seek professional help and pursue surgical candidacy for their child.

The inefficiency of frenotomy in some infants and mothers as presented in our data may also be explained by unaligned treatment recommendation. Even though the surgical treatment decision relies on an ENT specialist assessment, disputing treatment strategy advice between other healthcare professionals such as midwives, nurses, breastfeeding consultants, osteopaths, physiotherapists and general practitioners may lead to parental confusion, frustration and essentially malaligned expectations. If Danish doctors find no indication for surgical treatment and/or advise against it, some parents may choose to travel outside Denmark to have the procedure performed elsewhere. Thus, the ENT treatment decision may be affected by some parents’ wish to have a frenotomy performed, which may potentially lead to overtreatment of this patient category.

TT diagnosis and treatment advice have been debated over the years among different groups of healthcare professionals and providers [16]. Some argue that TT resolves spontaneously or that affected children and mothers may learn to compensate for the decreased lingual mobility [17]. Others recommend a more progressive surgical treatment regime [18]. To coordinate a uniform TT assessment strategy across different groups of healthcare professionals, a new Danish national clinical guideline (NCG) on assessment and treatment of ankyloglossia [2] was published in June 2020. The guideline recommends the TT and Breastfed Babies assessment tool (TABBY) for evaluation of risk of TT in breastfeeding infants. International studies have shown that this tool may be administered by different groups of healthcare professionals [19]. This approach may sharpen the diagnostic criteria and expectedly clarify the treatment strategy for this group of patients in the

future. As TT assessment and treatment strategy between healthcare professionals align, the frenotomy frequency in Danish infants may eventually stabilise.

Potential study limitations must be considered. Risk of recall bias increases as latency between procedure performance and data registration increases. However, in the analysis adjusting for procedure-to-interview delay, no significant difference in parent reports were found regarding symptom relief and onset of symptom relief after frenotomy for either infants or mothers. Also, for infants with parent-reported onset of symptom relief later than one week after the procedure, the chance of spontaneous symptom relief not associated with the surgical procedure must be acknowledged. Another limitation is inclusion bias as cases of post-operative symptom relief may be overrepresented in the 71% responder group. Furthermore, as participants were recruited from a population of families that were already predisposed to the idea of surgical intervention, referral bias must be considered.

CONCLUSIONS

Most parents in the present study reported either a moderate or high post-operative infant and maternal symptom relief following frenotomy. Prompt symptom alleviation occurred in approximately half of these. Taking the quantity and the potential repercussions of the preoperative symptoms into account, frenotomy seems as a safe and effective treatment of infants with TT and breastfeeding difficulties.

In the context of an increasing frenotomy frequency in Danish infants over the past decade, the inefficiency of frenotomy in some infants and mothers as seen in our data may imply some degree of overtreatment of this patient category.

Use of a uniform assessment tool such as the TABBY in children with breastfeeding difficulties as presented in the 2020 NCG may sharpen diagnostic criteria, clarify treatment recommendation and eventually stabilise the frenotomy frequency in Danish infants.

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Accepted 17 March 2022

Conflicts of interest none. Disclosure forms provided by the authors are available with the article at ugeskriftet.dk/dmj

References can be found with the article at ugeskriftet.dk/dmj

Cite this as *Dan Med J* 2022;69(5):A12210934

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