



## ORIGINAL ARTICLE

# Complications following frenotomy for ankyloglossia: A 24-month prospective New Zealand Paediatric Surveillance Unit study

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**Aim:** To investigate the incidence and characteristics of complications arising from frenotomy for ankyloglossia (tongue-tie) in New Zealand.

**Methods:** Prospective surveillance among hospital-based paediatricians of complications arising from frenotomy for ankyloglossia to children <1 year old was conducted by the New Zealand Paediatric Surveillance Unit for 24 months, from August 2016 to July 2018, inclusive.

**Results:** A total of 16 cases of complications arising from frenotomy were reported. The overall average annual incidence rate was 13.9/100 000. Geographic variation was noted with a peak of 85.6/100 000 in one region. Complications reported: poor feeding (44%), respiratory events (25%), pain (19%), bleeding (19%) and weight loss (19%). Three children (19%) also had delayed diagnosis of an underlying medical condition initially overlooked in favour of treating their ankyloglossia, this has not previously been reported. The majority (75%) of cases required admission to hospital. Treatments given included supplementary feeds (44%), surgical intervention (25%), breastfeeding support (19%), analgesia (13%) and blood products (13%). A total of 25% of children had one or more frenotomies; 50% were treated for two or more of: 'anterior' ankyloglossia, 'posterior' ankyloglossia or 'lip tie'; 50% had their frenotomies performed out of the hospital. Dentists were the most common performing practitioner (31%).

**Conclusions:** Frenotomy rates in New Zealand are unknown. Poor feeding, pain, bleeding, weight loss and delayed diagnosis of an alternative underlying medical condition are important complications that require hospital assessment and admission. Practitioners and parents/families need to be aware of these possibilities. Centralised guidelines with access to specialist second opinions should be developed.

**Key words:** ankyloglossia; breastfeeding; frenotomy; lingual frenulum; neonatology.

## What is already known on this topic

- 1 Tongue-tie (ankyloglossia) can interfere with successful breastfeeding in infants, which may be improved with frenotomy.
- 2 Frenotomy has an approximately 1% rate of minor complications.
- 3 Internationally, ankyloglossia diagnosis and treatment has increased leading to concerns of potential overtreatment.

## What this paper adds

- 1 Complications following frenotomy are occurring that require assessment and admission by hospital-based Paediatricians in New Zealand, with a significant geographical variation.
- 2 Undue focus on a potential tongue-tie as a cause for poor feeding may lead to alternative diagnoses being missed.
- 3 Respiratory complications such as apnoea are occurring following frenotomy in community settings that may not be equipped to deal with these.

Tongue-tie (ankyloglossia) is a condition characterised by the lingual frenulum being short, thick or tight, causing limitation in the range of tongue movement, which may interfere with feeding and later speech.<sup>1</sup> There is uncertainty regarding the correlation between the appearance of the lingual frenulum and functionally significant limitation of tongue movement, as many babies who

are diagnosed with ankyloglossia are able to breastfeed without difficulties.<sup>2-5</sup> International incidence rates of ankyloglossia are reported to range between 0.1 and 10.7%,<sup>3,4</sup> consistent with heterogeneity in the literature describing diagnosis and treatment.<sup>6</sup>

For infants with ankyloglossia experiencing difficulties with breastfeeding, division of the lingual frenulum (frenotomy) can improve feeding, particularly when maternal pain with breastfeeding is present.<sup>7</sup> Frenotomy is performed by a range of practitioners including midwives, doctors and dentists, while traditionally it has been performed using scissors, there is a trend for the use of a laser.

Ankyloglossia is sometimes divided into 'anterior' and 'posterior' depending on the location of frenulum attachment on the

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Background			
The majority of babies with tongue-tie are able to breastfeed without difficulty. If there are breastfeeding difficulties, a lactation consultant referral is recommended			
Assessment (by referring clinician)			
Discuss breastfeeding concerns			
Examine beneath the tongue:			
<ul style="list-style-type: none"> <li>• A frenulum attachment beneath the tongue is a variant of normal anatomy</li> <li>• Ankyloglossia (tongue-tie) is when the frenulum is abnormally short or dense and limits the range of tongue movement</li> </ul>			
Explain that a lactation consultant assessment is necessary before tongue-tie release surgery can be considered			
Management			
Provide reassurance that most babies with tongue-tie do not need surgery unless it is causing breastfeeding difficulties. There is no published evidence supporting a link between breastfeeding issues and lip tie			
If the tongue-tie is thought to be creating difficulty with breastfeeding:			
<ul style="list-style-type: none"> <li>• Refer to a lactation consultant for breastfeeding assessment, and Bristol tongue assessment tool (BTAT) Scoring</li> <li>• The lactation consultant will on-refer for tongue-tie release surgery, if indicated</li> <li>• Consideration for funded surgery requires a breastfeeding assessment by a lactation consultant, or midwife who has completed additional training</li> </ul>			
Frenotomy indications			
Frenotomy within the first 48 h was restricted to infants diagnosed with tongue-tie and severe feeding difficulties, following a specialist medical review			
Infants aged between 48 h and 8 weeks with breast-feeding difficulties needed an assessment by a lactation consultant (or midwife with additional training) and a BTAT score $\leq 4$ for frenotomy with a neonatologist			
An otorhinolaryngology surgeon would see infants older than 8 weeks following a breast-feeding assessment by a lactation consultant (or midwife with additional training) and a BTAT score $\leq 4$ , or infants with an anatomical abnormality			
Importance of education for hospital staff and community, with skilled breast-feeding support acknowledged and promoted			
BTAT			
Score	0	1	2
Appearance of tongue tip	Heart shaped	Slight cleft/Notched	Rounded
Attachment of frenulum to lower gum edge	Attached at top of gum ridge	Attached to inner aspect of gum	Attached to floor of mouth
Lift of tongue wide mouth wide (crying)	Minimal tongue lift	Edges only to mid-mouth	Full tongue lift to mid-mouth
Protrusion of tongue	Tip stays behind gum	Tip over gum	Tip can extend over lower lip
Total score of 0–3 indicates severe reduction of tongue function			

**Fig. 1** Example of successful pathway for tongue-tie release for babies with breast feeding difficulties (summary of canterbury District Health Board local pathway<sup>15</sup>).

ventral surface of the tongue. Some clinicians also consider the labial frenulum as having a role in limiting upper lip mobility and contributing to breastfeeding difficulties, with a ‘lip tie’ sometimes being divided concurrently with the lingual frenulum. The anatomy of the lingual frenulum has recently been described in detail for the first time; being a structure formed by the dynamic elevation of a midline fold in the floor of mouth fascia, and not simply a band of tissue.<sup>8</sup>

Internationally it has been noted that the prevalence of diagnosis and treatment of ankyloglossia has increased, with concerns raised of potential overtreatment.<sup>1,9,10</sup> Frenotomy has been described as ‘pain and risk-free’,<sup>3</sup> but there is now awareness that infants experience pain and stress, with prolonged pain and subsequent oral aversion having been reported.<sup>3</sup> Additionally, there is the potential that a focus on ankyloglossia may cause undue delay in diagnosis and management of other potential causes of poor feeding, such as an underlying medical condition.<sup>4</sup> Complications have generally been published as case reports, including one series of severe bleeding.<sup>11</sup> Two systematic reviews of frenotomy both found an overall rate of 1% for mild complications, with minor bleeding the most frequent.<sup>12,13</sup> No serious complications were

reported, but the total numbers of infants included was small, and the studies of limited quality. No such studies have been performed in New Zealand.

Frenotomy rates in New Zealand are unknown; however, a recent audit<sup>14</sup> showed that a single general practitioner in Dunedin treated 12% (414) of babies (3504) in the Otago/Southern region that year, most of whom were not enrolled in his practice. Canterbury, another New Zealand region, has reported a process of developing an evidence-based local pathway (Fig. 1) for referral, assessment and treatment of ankyloglossia with the aim of limiting unnecessary surgery, reducing their frenotomy rate from 11.5 to 3.6% over a 2-year period.<sup>15</sup> They used a Bristol tongue-tie assessment tool score  $\leq 4$  (score 0–3 indicates severe reduction of tongue function) performed in a timely fashion by a specifically trained professional as the threshold for obtaining funded frenotomy, with skilled breastfeeding assessment and support.

A paucity of prospective studies examining moderate to severe complications following frenotomy led to this study. We aimed to determine rates of moderate to severe complications of tongue-tie procedures presenting to hospital-based paediatricians in New Zealand, and describe this population.

## Methods

The New Zealand Paediatric Surveillance Unit (NZPSU) undertakes surveillance of uncommon childhood conditions through a monthly electronic reporting card sent to New Zealand specialist paediatricians. NZPSU send reporting cards to 88% of all paediatricians registered with the New Zealand Medical Council asking them to state if they have seen one of the conditions of interest, over the study period their response rate was 89%. Using NZPSU from August 2016 until July 2018 inclusive, cases of frenotomy complications were requested prospectively.

Case definition included children under 1 year of age, with any complication attributable to, or arising from, any form of frenotomy for ankyloglossia, irrespective of whether this was the main reason for presenting to the paediatrician.

Following notification, a unique link to an online questionnaire was sent to the reporting paediatrician. Anonymous information requested included demographic data, details of the complication(s) and their management, the frenotomy itself, any association with delayed diagnosis of a different condition, and the clinician's perspectives of the case. Analysis of the case was included if the questionnaire was completed by the reporting paediatrician.

Data is presented using mean or medians (range) as appropriate. Statistical analysis was performed using the Poisson distribution to calculate 95% confidence intervals. Population data obtained from the Statistics New Zealand 2013 Census<sup>16</sup> was used to estimate incidence rates, presented as average annual rates over the study period.

Ethical approval for this research was granted by the Lower South Regional Ethics Committee, of the New Zealand Ministry of Health.

## Results

During the study period, there were 23 notifications of complications relating to frenotomy on children under 1 year old from 17 paediatricians. For these notifications, 16 (70%) completed online questionnaires were received and 7 (30%) were not.

The annualised incidence rate of frenotomy complications for children under 1 year old presenting to hospital-based paediatricians in New Zealand was 13.9/100 000 (95% confidence interval 7.96–22.6), with a high estimate of 20/100 000 if non-responders were included.<sup>15</sup> Rates of complications by region are

presented in Table 1. The highest incidence was in Otago/Southland at 85.6/100 000 (95% confidence interval 31.4–186), no cases were reported from Canterbury and Nelson-Marlborough/Westland. The  $\chi^2$  test suggests the regional differences are significant ( $P < 0.001$ ) but the numbers are too small for this to be a reliable test.

## Demographic details of cases

Table 2 summarises demographic data. In total, 63% of the infants were male, with 14 (88%) cases identified as European ethnicity. Mean age at complication was 45 days (range 6–206), mean age at procedure 32 days (range 1–79), mean time between procedure and presentation where known was 3 days (range 0–15), with 67% presenting on the same day. The apparent discrepancy is due to incomplete data in four cases for the age at frenotomy, with the older infants' ages less likely to be known. Six (38%) of babies were exclusively or fully breastfeeding, six (38%) had mixed feeding (breast and bottle) and four (25%) were fully bottle-fed.

Indications reported for the procedures included breastfeeding difficulties (13 cases, 81%), breast/nipple pain (3, 19%) and weight loss (3, 19%), for 2 (13%) it was unknown. 'Other' specific reasons reported in three infants (19%) were 'unsettledness and jaundice', 'fussy and windy' and 'slow weight gain'.

Diagnosis of ankyloglossia was made by a lactation consultant and/or midwife in 12 cases (75%), one by a speech and language therapist (SLT), one parent diagnosed, and for two the diagnosing practitioner was unknown.

Treatment procedures for ankyloglossia were carried out in private clinic setting for eight (50%) cases, three (19%) in a public hospital and one (6%) in a private operating theatre, with the location unknown in four (25%). Frenotomy was performed by: dentists five (31%); general practitioners, lactation consultants and otolaryngology surgeons performed two each (13%); one each by paediatrician (6%) and paediatric surgeon (6%). In three cases (19%), the practitioner was unknown.

In nine cases (56%), the technique for ankyloglossia treatment was unknown. 'Anterior' ankyloglossia was treated in eight cases (50%) and eight (50%) had a 'lip tie' divided. 'Posterior' ankyloglossia was treated in two (13%), with half (eight) of babies treated for two or more types. The most common combination being 'anterior' and 'lip tie' treated together in five infants

**Table 1** Incidence of tongue-tie complication in babies less than 1 year old in New Zealand by province

Combined provincial regions	n (%)	Population estimate, n†	Annual incidence rate per 100 000	95% CI
Auckland/Northland	3 (19)	22 821	6.6	1.36–19.2
Waikato/Bay of Plenty/Taranaki/Gisborne	5 (31)	11 538	21.7	7.04–50.6
Wellington/ Hawke's Bay/Wanganui	2 (13)	11 181	8.9	1.08–32.3
Canterbury	0 (0)	6570	—	0–94.7
Nelson-Marlborough/Westland	0 (0)	1947	—	0–28.1
Otago/Southland	6 (38)	3504	85.6	31.4–186
Total	16 (100)	57 561	13.9	7.96–22.6

†Population estimates provided by statistics New Zealand, from 2013 census data. CI, confidence interval.

**Table 2** Demographic details of cases

	Mean (range)
Age at complication, days	45.3 (6–206)
Age at procedure, days	31.7 (1–79)
Gestation at birth, weeks	38.1 (30–41)
Birthweight, g	3366 (1190–3880)
Maternal age, years	29.8 (21–38)
Hospital admission, days	3.7 (1–14)
Ethnicity,† n (%)	
European	14 (88)
Maori	1 (6)
Asian	1 (6)

†Level 1 Ministry of Health ethnicity codes.

(31%). Four (25%) babies had undergone one or more previous frenotomy procedures.

## Complications

Table 3 summarises complications, given respondents were able to select more than one complication, totals add up to more than 100%. The most frequently reported presenting complications attributed to frenotomy included poor feeding (44%), apnoea, apparent life-threatening event or brief resolved unexplained episode, or other respiratory symptoms (25%), pain (19%), bleeding (19%) and weight loss (19%). Six (38%) of the 16 infants had immediately evident complications. Of those infants with weight loss, two lost more than 10% of their birthweight. Five reported 'other' complications which are listed in Table 3. Three (19%) infants additionally had reported the delayed diagnosis of another condition; these were a cardiac disorder, severe dehydration and weight loss, and failure to adequately establish feeding. One infant was born at 30 weeks' gestation and presented after their fourth frenotomy with scarring and poor-feeding. It was noted

**Table 3** Complication of tongue-tie procedure

Complication reported	n (%)†
Poor feeding	7 (44)
Apnoea, ALTE/BRUE, or other breathing difficulties	4 (25)
Pain	3 (19)
Bleeding	3 (19)
Weight loss‡	3 (19)
Pallor/Anaemia	2 (13)
Excess scarring	2 (13)
Other§	5 (31)

†Percentages add up to > 100% as more than one complication was present in some instances. ‡Weight loss greater than 10% of birthweight reported for 2 of 3 (67%). §'Other' complications reported (n = 1 each): unsettledness; peripheral cyanosis; greyish black stools; ulcer; severe hypernatraemia (171), hypothermia and 20% weight loss. Responders were able to select more than one option for some questions, totals may add up to more than 100%. ALTE, apparent life-threatening event; BRUE, brief, resolved, unexplained event.

**Table 4** Treatment for complication

Treatment given	n (%)†
Supplementary feeds	7 (44)
Surgical intervention	4 (25)
Breastfeeding support	3 (19)
Analgesia	2 (13)
Blood products	2 (13)
Antibiotics	1 (6)
Intravenous (IV) fluids	1 (6)
None or observation only	2 (13)
Other‡	2 (13)

†Percentages add up to >100% as more than one complication was present in some instances. ‡'Other' complications (n = 1 each): increased continuous positive airway pressure (CPAP) pressure; ferrous sulphate and vitamin C. Responders were able to select more than one option for some questions, totals may add up to more than 100%.

this infant was breast and bottle feeding well on discharge from the neonatal intensive care unit. After his frenotomies, he developed an oral aversion.

Treatments for complications are presented in Table 4. The majority (75%) of the cases required hospital admission with a mean length of stay of 3.7 days (range 1–14 days). For nine (56%), the complication of ankyloglossia treatment listed as the main reason for admission. One infant who presented with bleeding received both surgical intervention and blood products. Having bled for more than a day after frenotomy, the infant required treatment with intravenous vitamin K, packed red cell and fresh frozen plasma transfusions, and bipolar diathermy. Laboratory tests were consistent with a consumptive process and vitamin K deficiency bleeding, despite the infant having intramuscular vitamin K at birth.

Frequently clinical concerns were expressed by the notifying paediatrician. For example, one case was an inpatient on a children's ward receiving CPAP for airway obstruction when their frenotomy was performed without involvement of their paediatrician. Following frenotomy, the infant had worsening of their airway obstruction. Another clinician reported an infant presenting with severe weight loss (greater than 20%) and hypernatremia (171 mmol/L) due to feeding difficulties, with the family having been inappropriately reassured that the feeding problems would resolve post frenotomy. A different infant had delay in diagnosis of a large ventricular septal defect (VSD)/atrial septal defect (ASD) causing cardiac failure and associated poor weight gain, due to focus on ankyloglossia.

Other themes reported included a perceived financial conflict of interest (with frenotomy generating income) and concerns regarding lack of knowledge/care around the risks of bleeding and/or vitamin K deficiency in infants. Comments reported included families understandably regretting undergoing frenotomy that risks had been understated and that frenotomy had not improved the problems for which it had been recommended.

One case that highlights the potential complexity and importance of considering a broad differential for causes of feeding issues is that of a 4-week-old male who had an apnoea during

his procedure. The subsequent admission for this complication leads to a diagnosis of congenital heart disease and associated poor weight gain. A dentist had performed laser surgery to release lip and anterior tongue-ties, having been referred by a lactation consultant. No vitamin K had been given at birth, and the performing clinician was unaware of the risk of vitamin K deficiency bleeding.

## Discussion

This is the first study to prospectively report complications relating to ankyloglossia procedures in babies. We report an overall incidence rate of 13.9 complications seen by hospital-based specialist paediatricians per 100 000 infants less than 1 year old. Geographic variation is noted with a peak of 85.6/100 000 in Otago/Southland, and 0/100 000 in Canterbury. Notably, Canterbury had a centralised referral pathway (aiming to reduce unnecessary frenotomies) implemented 4 months after the start of our data collection.<sup>15</sup> National frenotomy rates are unknown, but for Otago/Southland this is estimated as at least 12 133/100 000 (based on the audit<sup>14</sup>), which would equate to an estimated moderate to severe complication rate for this region of approximately 1% for babies undergoing the procedure.

Complications following frenotomy reported commonly focussed on bleeding. In this study, where cases were reported by hospital-based paediatricians, common presentations were feeding difficulties, followed by respiratory events such as apnoea (or apparent life-threatening event/brief resolved unexplained episode), bleeding, and weight loss. Most bleeding post-frenotomy would be considered minor and self-resolving, therefore not warranting presentation to the hospital to be captured with our study methodology.

The majority of infants described had frenotomies performed in community settings, which may not be equipped to deal with a medical emergency such as apnoea. While this may have influenced reporting captured in this study (due to referral to the hospital more likely) these airway complications are important from a morbidity perspective. Of particular concern and not previously reported, are the three cases where potential for feeding difficulties to be due to another, serious underlying condition was overlooked, and the diagnosis missed or delayed.

Most of the post-frenotomy complications reported in this study were in patients who had ankyloglossia diagnosed and/or managed outside the public health sphere by non-paediatricians, emphasising the importance of communication between families and all health professionals providing care to a child. A sizeable minority of cases (25%) had repeat procedures, a rate similar to the audit from Otago/Southland (23%). If success has not occurred after the initial procedure, further specialist referral should be considered to avoid missing an alternative cause for feeding difficulties and/or repeated unnecessary surgery.

Strengths of this study include its prospective nature, utilising the established NZPSU network which reaches almost all New Zealand hospital-based paediatricians. There was a high NZPSU response rate over this time period (89%) with a high rate of completed questionnaires (70%) for our study allowing the researchers to obtain detailed information around the reported cases.

Limitations include that cases were reported by hospital-based paediatricians, thereby missing complications seen by other practitioners, along with the incomplete return of questionnaires for reported cases, giving an underestimate the true rate. Minor complications are likely to be managed outside of the hospital setting, and clinically significant complications are more likely to come to the attention of the practitioners (paediatricians) who completed the questionnaires. The denominator for frenotomy rates is unknown in this population, so the true complication rate is not established. Traditional surveillance unit data is also limited in its ability to follow-up cases, obtain objective data about the indication and efficacy of the procedure leading to the complication, and reporting clinicians' perspectives may be coloured by the complication seen. However, only three of the 16 total cases had symptoms that could possibly be pre-existing alone (weight loss, poor feeding and/or unsettledness) and no other complications (such as bleeding, pain, respiratory events) attributable to frenotomy.

## Conclusions

Whilst frenotomy rates in New Zealand are unknown, our data confirm that poor feeding, respiratory events, pain, bleeding, weight loss and delayed diagnosis of an alternative underlying medical condition are important complications that occurred following frenotomy and required hospital assessment and admission. Clinicians referring for or considering performing frenotomy should gauge the appropriateness of the procedure, and ideally use a recognised tongue-tie scoring criteria (i.e. Bristol tongue-tie assessment tool  $\leq 4$ ) to guide assessment. Practitioners conducting frenotomy need to be aware of the broad spectrum of potential complications that were encountered in this study. Parents/families need to be advised accordingly, and steps need to be taken to actively minimise the risk of these complications occurring. In the diagnosis and management of ankyloglossia, centralised guidelines with freely accessible breastfeeding support and timely access to specialist second opinions should be considered as the ideal model, as shown with the successful Canterbury approach.<sup>15</sup>

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