Paediatrics & Child Health, 2017, 382–386
doi: 10.1093/pch/pxx112
Original Article
Advance Access publication 25 September 2017

OXFORD

Original Article

Changes in the incidence and surgical treatment of ankyloglossia in Canada

Michelle Lisonek[1,2], Shiliang Liu MB PhD[3], Susie Dzakpasu PhD[3], Aideen M. Moore MD FRCPC MHSc[4,5], K. S. Joseph MD PhD[1,2,6,7]; for the Canadian Perinatal Surveillance System (Public Health Agency of Canada)

[1]Department of Obstetrics and Gynaecology, University of British Columbia, Vancouver, British Columbia; [2]Children's and Women's Hospital and Health Centre of British Columbia, Vancouver, British Columbia; [3]Maternal, Child and Youth Health, Surveillance and Epidemiology Division, Centre for Chronic Disease Prevention, Public Health Agency of Canada, Ottawa, Ontario; [4]The Hospital for Sick Children, Toronto, Ontario; [5]Division of Neonatology, University of Toronto, Toronto, Ontario; [6]School of Population and Public Health, University of British Columbia, Vancouver, British Columbia; [7]The BC Children's Hospital Research Institute, Vancouver, British Columbia

Correspondence: K. S. Joseph, Room C403, Department of Obstetrics and Gynaecology, Women's Hospital of British Columbia, 4500 Oak Street, Vancouver, British Columbia V6H 3N1. Telephone 604-875-2000 ext 4811, fax 604-875-2987, e-mail: kjoseph@cw.bc.ca

Abstract

Background: Recent reports show increases in rates of ankyloglossia and frenotomy in British Columbia. We carried out a study to determine temporal trends and regional variations in ankyloglossia and frenotomy in Canada.

Methods: The study included all hospital-based live births in Canada (excluding Quebec) between April 2002 and March 2015, with information obtained from the Canadian Institute for Health Information. Information on ankyloglossia and frenotomy was obtained from records of hospital admission for childbirth. Temporal trends and provincial/territorial variations were quantified using rate ratios (RR) and 95% confidence intervals (CI).

Results: Ankyloglossia rates increased from 6.86 in 2002 to 22.6 per 1000 live births in 2014 (P for trend < 0.001), while frenotomy rates increased from 3.76 in 2002 to 14.7 per 1000 live births in 2014 (P for trend < 0.001). Frenotomy rates among infants with ankyloglossia increased from 54.7% in 2002 to 63.9% in 2014 (RR: 1.18, 95% CI: 1.13–1.24). Compared with British Columbia, rates of ankyloglossia were over three-fold higher in Saskatchewan (RR: 3.40, 95% CI: 3.16–3.67), Alberta (RR: 3.50, 95% CI: 3.29–3.72) and the Yukon (RR: 3.62, 95% CI: 2.67–4.92), while rates of frenotomy were three- to four-fold higher in the Yukon (RR: 3.41, 95% CI: 2.28–5.10), Alberta (RR: 4.01, 95% CI: 3.71–4.33) and Saskatchewan (RR: 4.12, 95% CI: 3.76–4.52).

Conclusion: A desire to increase rates of breast feeding initiation and absence of standardized criteria for the diagnosis of ankyloglossia have resulted in runaway rates of frenotomy for newborn infants in some parts of Canada.

Keywords: *Ankyloglossia; Canada; Epidemiology; Frenotomy.*

Ankyloglossia (tongue-tie), a minor congenital anomaly characterized by a short, thickened or abnormally tight lingual frenulum, may result in varying degrees of decreased tongue movement. Recent increases in support for breast feeding initiation and the Baby Friendly Hospital Initiative (1) appear to have increased the diagnosis of ankyloglossia (2). Identifying ankyloglossia is problematic as there are no universally accepted criteria for diagnosis. The Canadian Paediatric Society's 2011 position statement (reiterated in 2014) stated that frenotomy for ankyloglossia could not be recommended among newborns (3). In 2015, the statement was modified; although frenotomy was not recommended for all cases of ankyloglossia, the potential for benefit in cases with significant breast feeding difficulties was recognized (4).

We previously carried out a population-based study of all live births in British Columbia, Canada, from 2004 to 2013 to quantify temporal trends and regional variations in rates of ankyloglossia and frenotomy (2). Our study showed that rates of ankyloglossia increased by 70% (95% confidence interval [CI]: 44–101%) from 5.0 per 1000 live births in 2004 to 8.4 per 1000 live births in 2013, while frenotomy rates increased from 2.8 per 1000 live births in 2004 to

5.3 per 1000 live births in 2013 (89% increase, 95% CI: 52–134%). There were marked regional differences in ankyloglossia and frenotomy rates; ankyloglossia rates varied over two-fold between the five regional health authorities, while frenotomy rates varied three-fold. Nulliparity, multiple birth, male infant sex, birth weight and year were independently associated with ankyloglossia (2).

Health behaviours during pregnancy and childbirth differ across Canada, and significant variations in rates of obstetric intervention exist between British Columbia and other Canadian provinces and territories. We, therefore, carried out a study to examine temporal trends and spatial variations in ankyloglossia and frenotomy in Canada.

METHODS

The study included all live born infants in Canada with information obtained from the Discharge Abstract Database of the Canadian Institute for Health Information between April 2002 and March 2015 (hereafter referred to as fiscal years 2002–2014). This database includes all birth hospitalizations (98% of births) in Canada (excluding Quebec). Information in the database is collated by trained medical record abstractors using standardized definitions and has been previously validated (5,6).

We identified diagnoses of ankyloglossia and frenotomy procedures during the childbirth admission. Diagnoses in the database were coded using International Classification of Diseases (ICD-10 CA) codes, and procedures were coded using the Canadian Classification of Interventions. Ankyloglossia (Q381) and frenotomy (1FJ72) were identified using appropriate codes. We first quantified rates of ankyloglossia and frenotomy in Canada (excluding Quebec) by year among all live births with the precision of the rate estimates expressed using exact 95% CI. The statistical significance of temporal trends in ankyloglossia and frenotomy was assessed using a chi-square test for linear trend. Spatial differences in rates of ankyloglossia and frenotomy in the provinces and territories were quantified using rate ratios (RR) and 95% CIs. Live births in the province of British Columbia, where we have previously documented two- to three-fold differences in rates of ankyloglossia and frenotomy between regional health authorities (2) were used as the reference group for these calculations. Temporal trends in ankyloglossia rates were also examined in each of the provinces and territories. Three-year moving averages were used to provide stability to rate estimates in provinces/territories with small numbers of cases. Finally, we also assessed temporal changes in rates of frenotomy among infants diagnosed with ankyloglossia.

The study was carried out as part of the Public Health Agency of Canada's Canadian Perinatal Surveillance System using anonymized data (for which ethics approval is not required).

RESULTS

The study population included 3,611,986 live births and 40,457 cases of ankyloglossia yielding a birth prevalence of 11.2 per 1000 live births. There were 24,975 cases of frenotomy (incidence rate 6.91 per 1000 live births). Ankyloglossia rates increased from 6.86 per 1000 live births in 2002 to 22.6 per 1000 live births in 2014 (P value for trend < 0.001), while frenotomy rates increased from 3.76 per 1,000 in 2002 to 14.7 per 1,000 live births in 2014 (P value for trend < 0.001; Table 1 and Figure 1). Frenotomy rates among infants with ankyloglossia increased from 54.7% in 2002 to 63.9% in 2014.

Table 2 shows the spatial variation in the diagnosis of ankyloglossia in the most recent years of the study period (2012–2014). The highest rates of ankyloglossia in this period were observed in Saskatchewan (32.4 per 1000 live births), Alberta (33.3 per 1000 live births) and the Yukon (34.6 per 1000 live births), while the lowest rates were seen in Nunavut (7.5 per 1000 live births), British Columbia (9.5 per 1000 live births), Newfoundland and Labrador (11.1 per 1000 live births) and Manitoba (11.7 per 1000 live births). Compared with British Columbia, rates of ankyloglossia were over three-fold higher in Saskatchewan (RR: 3.40, 95% CI: 3.16–3.67), Alberta (RR: 3.50, 95% CI: 3.29–3.72) and the Yukon (RR: 3.62, 95% CI: 2.67–4.92). In 2014, the highest birth prevalence of ankyloglossia was observed in the Yukon (40 per 1000 live births, Alberta (43.0 per 1000 live births) and Nova Scotia (57.4 per 1000 live births).

The highest rates of frenotomy in 2012–2014 occurred in the Yukon (20.2 per 1000 live births), Alberta (23.8 per 1000 live births) and Saskatchewan (24.5 per 1000 live births), while the lowest rates were observed in Newfoundland and Labrador (1.1 per 1000 live births), Nunavut (<4.2 per 1000 live births) and British Columbia (5.9 per 1000 live births). Compared with British Columbia, rates of frenotomy were three- to four-fold higher in the Yukon (RR: 3.41, 95% CI: 2.28–5.10), Alberta (RR: 4.01, 95% CI: 3.71–4.33) and Saskatchewan (RR: 4.12, 95% CI: 3.76–4.52). In 2014, the highest rate of frenotomy was observed in Saskatchewan (25.1 per 1000 live births), Alberta (30.4 per 1000 live births) and Nova Scotia (42.1 per 1000 live births).

Table 1. Numbers and rates of ankyloglossia and frenotomy among live births, Canada (excluding Quebec), 2002–2014

Year	Live births	Ankyloglossia				Frenotomy			
		Number	Rate/1000 live births	Rate ratio	95% confidence interval	Number	Rate/1000 live births	Rate ratio	95% confidence interval
2002	238,176	1634	6.86	1.00	—	895	3.76	1.00	—
2003	251,784	1896	7.53	1.10	1.03–1.17	1108	4.40	1.17	1.07–1.28
2004	265,699	2040	7.68	1.12	1.05–1.19	1230	4.63	1.23	1.13–1.34
2005	269,248	2129	7.91	1.15	1.08–1.23	1272	4.72	1.26	1.15–1.37
2006	277,335	2092	7.54	1.10	1.03–1.17	1310	4.72	1.26	1.15–1.37
2007	288,133	2472	8.58	1.25	1.18–1.33	1487	5.16	1.37	1.26–1.49
2008	289,336	2599	8.98	1.31	1.23–1.39	1532	5.29	1.41	1.30–1.53
2009	291,488	2920	10.0	1.46	1.37–1.55	1713	5.88	1.56	1.44–1.70
2010	286,126	3222	11.3	1.64	1.55–1.74	1969	6.88	1.83	1.69–1.98
2011	287,917	3768	13.1	1.91	1.80–2.02	2344	8.14	2.17	2.01–2.34
2012	289,394	4135	14.3	2.08	1.97–2.20	2640	9.12	2.43	2.25–2.62
2013	287,269	4994	17.4	2.53	2.40–2.68	3222	11.2	2.98	2.77–3.21
2014	290,081	6556	22.6	3.29	3.12–3.48	4253	14.7	3.90	3.63–4.19
Total	3,611,986	40,457	11.2	—	—	24,975	6.91	—	—

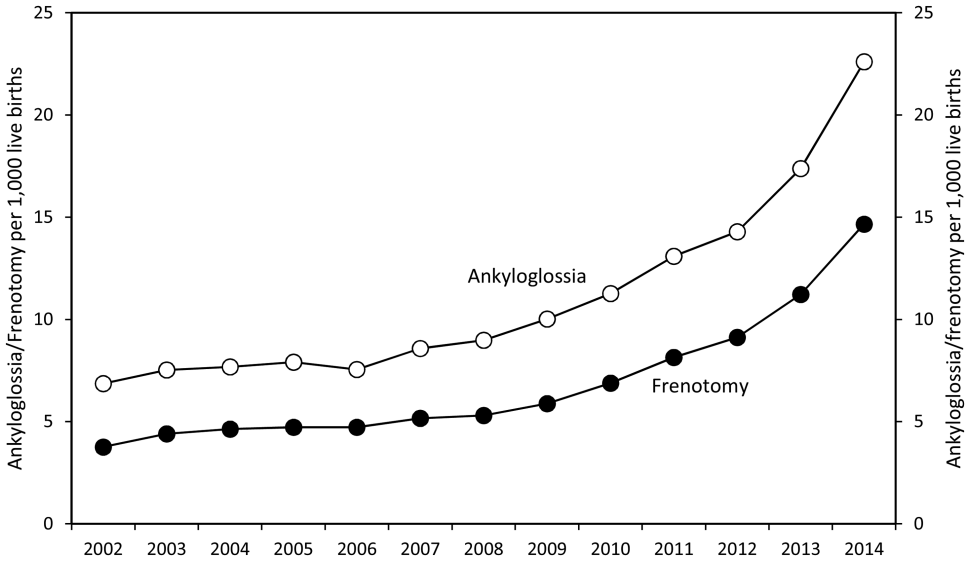

Figure 1. Rates of ankyloglossia and frenotomy, Canada (excluding Quebec), 2002–2014

Table 2. Numbers and rates of ankyloglossia and frenotomy among live births, Canada, provinces and territories, 2012–2014

Year	Live births	Number	Rate	Rate ratio	95% confidence interval
Ankyloglossia					
Newfoundland and Labrador	13,550	150	11.1	1.16	0.98–1.37
Prince Edward Island	4036	67	16.6	1.74	1.36–2.22
Nova Scotia	25,627	668	26.1	2.73	2.49–3.00
New Brunswick	20,939	262	12.5	1.31	1.15–1.50
Ontario	416,609	5914	14.2	1.49	1.40–1.58
Manitoba	49,759	580	11.7	1.22	1.11–1.35
Saskatchewan	45,470	1475	32.4	3.40	3.16–3.67
Alberta	158,066	5270	33.3	3.50	3.29–3.72
British Columbia (reference)	128,033	1221	9.54	1.00	—
Northwest Territories	2273	28	12.3	1.29	0.89–1.87
Nunavut	1196	9	7.53	0.79	0.41–1.52
Yukon	1186	41	34.6	3.62	2.67–4.92
Canada	866,744	15,685	18.1	—	—
Frenotomy					
Newfoundland and Labrador	13,550	15	1.11	0.19	0.11–0.31
Prince Edward Island	4036	47	11.7	1.96	1.46–2.63
Nova Scotia	25,627	403	15.7	2.65	2.35–2.99
New Brunswick	20,939	173	8.26	1.39	1.18–1.64
Ontario	416,609	3447	8.27	1.39	1.18–1.64
Manitoba	49,759	353	7.09	1.20	1.05–1.36
Saskatchewan	45,470	1112	24.5	4.12	3.76–4.52
Alberta	158,066	3762	23.8	4.01	3.71–4.33
British Columbia (reference)	128,033	760	5.94	1.00	—
Northwest Territories	2273	16	7.04	1.19	0.72–1.94
Nunavut	1196	<5*	<4.18*	—*	—*
Yukon	1186	24	20.2	3.41	2.28–5.10
Canada (excluding Quebec)	866,744	10,117	11.7	—	—

*Suppressed because of small cell counts.

Figure 2 shows temporal trends in rates of ankyloglossia between 2002 and 2014. Rates of ankyloglossia increased steadily in most provinces and territories except in Newfoundland and Labrador, Northwest Territories, Nunavut and Nova Scotia. In Newfoundland and Labrador, Northwest Territories and Nunavut, rates of ankyloglossia were stable until 2009–2010 after which rates increased sharply. In Nova Scotia, the rate increased slightly in 2013 and increased several-fold in 2014.

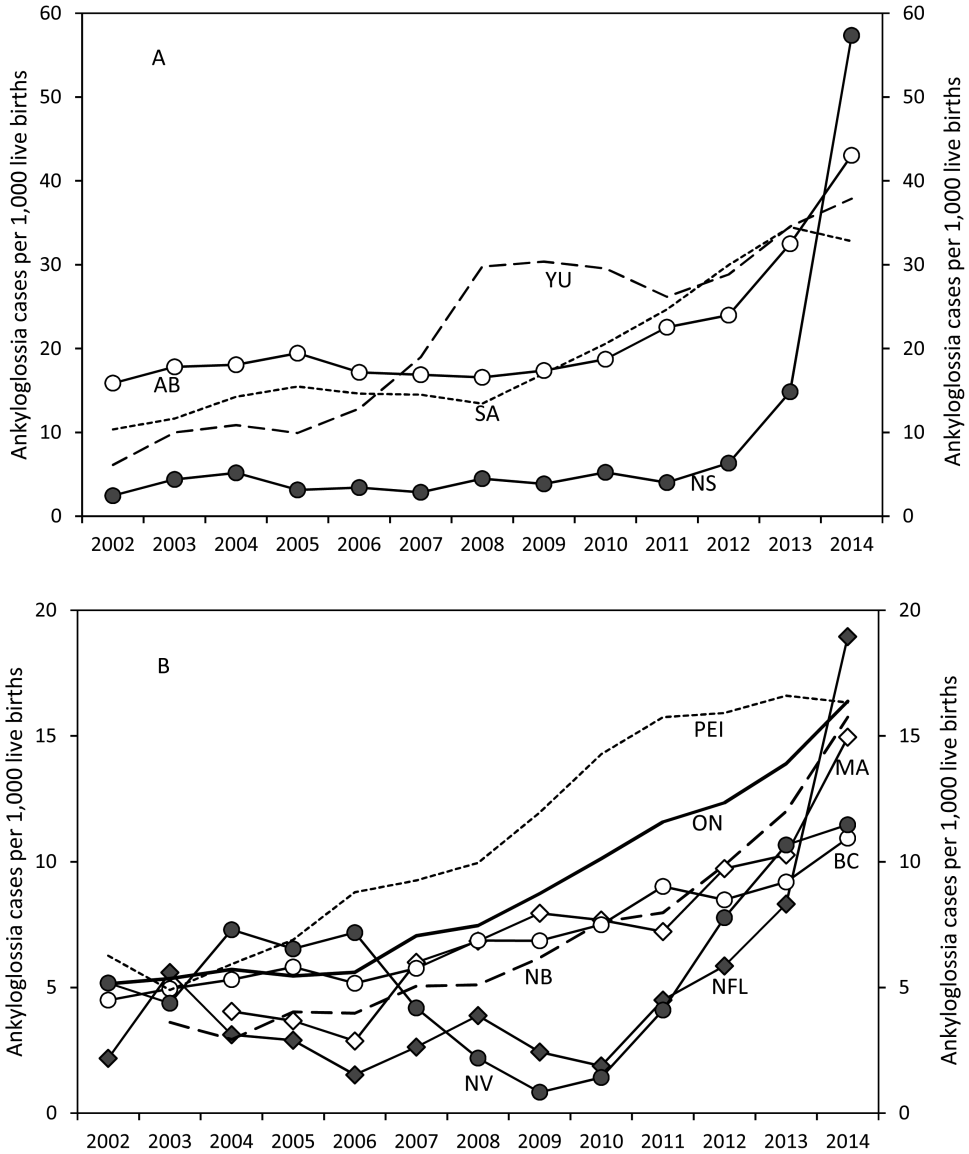

Figure 2. Temporal trends in ankyloglossia in the provinces and territories with relatively high rates of ankyloglossia in recent years (**A**) and in provinces and territories with relatively low rates (**B**), Canada (excluding Quebec), 2002–2014. *AB Alberta; BC British Columbia; MB Manitoba; NB New Brunswick; NFL Newfoundland and Labrador; NS Nova Scotia; NV Northwest Territories and Nunavut; ON Ontario; PEI Prince Edward Island; SA Saskatchewan; YU Yukon*. Rates for PEI and NWT-NU are 3-year moving averages.

DISCUSSION

Our study shows a rapid temporal increase in ankyloglossia and frenotomy rates in Canada from 2002 to 2014 and large spatial variation in rates. Ankyloglossia rates increased over three-fold between 2002 and 2014, while frenotomy rates increased almost four-fold over the same period. The increase in frenotomy rates occurred because of increases in the diagnosis of ankyloglossia and also because of increases in rates of frenotomy among infants diagnosed with ankyloglossia. Regional variations in rates of ankyloglossia and frenotomy were substantial, with rates in Saskatchewan, Alberta and the Yukon being three- to four-fold higher than rates in British Columbia. The temporal increase in frenotomy rates during the childbirth admission appears to be accelerating, with some provinces such as Alberta and Nova Scotia subjecting 3–4% of infants to this procedure in 2014.

The rate of ankyloglossia in our study was 11.2 per 1000 live births, and this is significantly lower than estimates from previous studies that have typically yielded birth prevalence rates of 4% (range 0.02–11%) (7–12). Diagnosis of ankyloglossia at the childbirth admission tends to be symptom prompted. The low rate in our study is not surprising given the typically asymptomatic nature of the condition. Wide variation in rates of ankyloglossia is also seen even in the more rigorous research setting as there is no consensus on the definition and diagnostic criteria for ankyloglossia. The development of tools which provide an objective, clear and simple measure of the severity of a tongue-tie may help address problems of incorrect diagnosis. A new tool has been described in the recent literature (13), although external validation and performance in clinical practice is awaited.

The recent increase in emphasis on breast feeding initiation before hospital discharge because of the Baby Friendly Hospital Initiative (1), appears to have led to a surveillance and detection bias that is responsible for ankyloglossia being diagnosed with increasing frequency in early infancy. On the other hand, the increase in ankyloglossia and frenotomy rates could represent earlier diagnosis and treatment of this condition during the childbirth admission resulting from the enhanced focus on breast feeding initiation. However, the large regional differences in rates of ankyloglossia and frenotomy seen in our study are poorly correlated with regional patterns of breast feeding initiation and duration (14,15).

Recent increases in rates of ankyloglossia and frenotomy in Canada have occurred despite guidelines from the Canadian Paediatric Society discouraging routine surgery in newborn infants (3,4). However, there is no international consensus on the appropriateness of frenotomy among newborns; the Dutch and Japanese Pediatric Societies (16–18) do not endorse frenotomy, while the United Kingdom guidelines (19), UNICEF's Baby Friendly Hospital Initiative (20) and the American Academy of Pediatrics (21) recommend frenotomy for symptomatic ankyloglossia. There is a lack of good quality evidence on the efficacy of frenotomy for addressing problems with breast feeding. To-date, the randomized trials carried out to assess the efficacy of frenotomy for ankyloglossia-associated breast feeding difficulties (11,22–25) have all been relatively small studies plagued by methodological problems related to inclusion criteria, lack of blinding and subjectively defined outcomes. A recent systematic review concluded that 'frenotomy may be associated with mother-reported improvements in breastfeeding and potentially in nipple pain, but with small, short-term studies with inconsistent methodology, strength of the evidence is low to insufficient' (26).

The controversy with regard to the use of frenotomy has been framed as a conflict between lactation nurses, breast feeding support groups and mothers experiencing breast feeding difficulties versus paediatricians who question the efficacy of frenotomy (3,27–29). The culture among paediatricians increasingly rejects minor surgical intervention (e.g., tonsillectomy, ear tubes) for babies and children since most conditions improve spontaneously. A survey of lactation consultants showed that 30% believe ankyloglossia causes feeding problems occasionally, while 69% reported that it frequently or always causes feeding difficulties (30). In contrast, 90% of paediatricians reported that ankyloglossia rarely or never causes feeding difficulties. However, our study was not able to quantify the contribution of different health care providers to the increase in ankyloglossia diagnosis and treatment.

The strengths of our study include its population-based provenance and large size. Our data source collates information from all hospital-based deliveries in Canada (excluding Quebec) using standardized definitions and procedures for medical chart abstraction, and validation studies show that the data is accurate (5,6). The weaknesses of our study include the exclusion of some 2% of home births and the restriction of the study to the hospital admission for childbirth. Ankyloglossia diagnosed after hospital discharge and frenotomies carried out subsequent to the childbirth admission were not included in our rate estimates. Our estimates of ankyloglossia and frenotomy rates, therefore, represent underestimates of the prevalence in infancy since ankyloglossia diagnoses often occur following hospital discharge after childbirth and frenotomy procedures are increasingly performed by dentists. Data source constraints included a lack of information on ankyloglossia and frenotomy rates in New Brunswick in 2002 and in Manitoba in 2003 and 2004.

CONCLUSION

A desire to increase rates of breast feeding initiation, the absence of standardized criteria for the diagnosis of ankyloglossia and insufficient evidence favouring frenotomy in the newborn period have resulted in a contentious situation with runaway rates and huge regional variations in rates of ankyloglossia and frenotomy in Canada. High-quality randomized trials are required to resolve this problem involving potentially unnecessary surgery for newborn infants.

Acknowledgement

Dr Joseph's work is supported by an Investigator award from the BC Children's Hospital Research Institute and a Chair award from the Canadian Institutes of Health Research (APR-126338).

References

1. World Health Organization, UNICEF. Baby-Friendly Hospital Initiative: Revised, Updated and Expanded for Integrated Care. Geneva: World Health Organization and UNICEF, 2009.
2. Joseph KS, Kinniburgh B, Metcalfe A, Razaz N, Sabr Y, Lisonkova S. Temporal trends in ankyloglossia and frenotomy in British Columbia, Canada, 2004–2013: A population-based study. CMAJ Open 2016;4:E33–40.
3. Rowan-Legg A. Ankyloglossia and breastfeeding. Paediatr Child Health 2011;16:222.
4. Rowan-Legg A. Ankyloglossia and breastfeeding. Paediatr Child Health 2015;20:209–18.
5. Canadian Institute for Health Information. Data Quality Documentation, Discharge Abstract Database—Multi-Year Information. Standards and Data Submission. Ottawa: Canadian Institute for Health Information, 2012.
6. Joseph KS, Fahey J; Canadian Perinatal Surveillance System. Validation of perinatal data in the Discharge Abstract Database of the Canadian Institute for Health Information. Chronic Dis Can 2009;29:96–100.
7. Messner AH, Lalakea ML, Aby J, Macmahon J, Bair E. Ankyloglossia: Incidence and associated feeding difficulties. Arch Otolaryngol Head Neck Surg 2000;126:36–9.
8. Ballard JL, Auer CE, Khoury JC. Ankyloglossia: Assessment, incidence, and effect of frenuloplasty on the breastfeeding dyad. Pediatrics 2002;110:e63.
9. Griffiths DM. Do tongue ties affect breastfeeding? J Hum Lact 2004;20:409–14.
10. Ricke LA, Baker NJ, Madlon-Kay DJ, DeFor TA. Newborn tongue-tie: Prevalence and effect on breast-feeding. j Am Board Fam Pract 2005;18:1–7.
11. Hogan M, Westcott C, Griffiths M. Randomized, controlled trial of division of tongue-tie in infants with feeding problems. J Paediatr Child Health 2005;41:246–50.
12. Segal LM, Stephenson R, Dawes M, Feldman P. Prevalence, diagnosis, and treatment of ankyloglossia: Methodologic review. Can Fam Physician 2007;53:1027–33.
13. Ingram J, Johnson D, Copeland M, Churchill C, Taylor H, Emond A. The development of a tongue assessment tool to assist with tongue-tie identification. Arch Dis Child Fetal Neonatal Ed 2015;100:F344–8.
14. Public Health Agency of Canada. Canadian Perinatal Health Report, 2008 edn. Ottawa: Public Health Agency of Canada, 2008.
15. Public Health Agency of Canada. Perinatal Health Indicators for Canada 2013: A Report of the Canadian Perinatal Surveillance System. Ottawa: Public Health Agency of Canada, 2013.
16. Post ED, Rupert AW, Schulpen TW. Problematic breastfeeding due to a short frenulum. Ned Tijdschr Geneeskd 2010;154:A918.
17. Constantine AH, Williams C, Sutcliffe AG. A systematic review for ankyloglossia (tongue-tie) in breast fed infants. Arch Dis Child 2011;96(S1):A62–3.
18. Committee of Ethics, Japan Pediatric Society. Survey on operative treatment in ankyloglossia and its results. J Japan Pediatr Soc 2001;105:520–22.
19. Division of Ankyloglossia (tongue-tie) for Breastfeeding, National Institute for Health and Care Excellence. NICE Interventional Procedure Guidance [IPG149]. December 2005. <https://www.nice.org.uk/guidance/ipg149> (Accessed March 31, 2015).
20. World Health Organization, UNICEF. What Mothers Need to Know about Tongue-Tie. <http://www.unicef.org.uk/BabyFriendly /Parents/Problems/Tongue-Tie/Division-of-tongue-tie/> (Accessed March 31, 2015).
21. American Academy of Pediatrics. Breastfeeding: Best for Baby and Mother. <http://www2.aap.org/breastfeeding/files/pdf/BBM-8–27%20Newsletter.pdf> (Accessed March 31, 2015).
22. Dollberg S, Botzer E, Grunis E, Mimouni FB. Immediate nipple pain relief after frenotomy in breast-fed infants with ankyloglossia: A randomized, prospective study. J Pediatr Surg 2006;41:1598–600.
23. Buryk M, Bloom D, Shope T. Efficacy of neonatal release of ankyloglossia: A randomized trial. Pediatrics 2011;128:280–8.
24. Berry J, Griffiths M, Westcott C. A double-blind, randomized, controlled trial of tongue-tie division and its immediate effect on breastfeeding. Breastfeed Med 2012;7:189–93.
25. Emond A, Ingram J, Johson D, et al. Randomised controlled trial of early frenotomy in breastfed infants with mild-moderate tongue-tie. Arch Dis Child Fetal Neonatal Ed 2014;99:F189–95.
26. Francis DO, Krishnaswami S, McPheeters M. Treatment of ankyloglossia and breastfeeding outcomes: A systematic review. Pediatrics 2015;135:e1458–66.
27. Edmunds JE, Fulbrook P, Miles S. Understanding the experiences of mothers who are breastfeeding an infant with tongue-tie: A phenomenological study. J Hum Lact 2013;29:190–5.
28. Power RF, Murphy JF. Tongue-tie and frenotomy in infants with breastfeeding difficulties: Achieving a balance. Arch Dis Child 2015;100:489–94.
29. Naimer SA. To cut or not to cut? Approach to ankyloglossia. Can Fam Physician 2016;62:231–2.
30. Messner AH, Lalakea ML. Ankyloglossia: Controversies in management. Int J Pediatr Otorhinolaryngol 2000;54:123–31.