CMAJ OPEN

Research

Temporal trends in ankyloglossia and frenotomy in British Columbia, Canada, 2004–2013: a population-based study

K.S. Joseph MD PhD, Brooke Kinniburgh MPH, Amy Metcalfe PhD, Neda Razaz MPH, Yasser Sabr MD MHSc, Sarka Lisonkova MD PhD

Abstract

Background: Routine surveillance of congenital anomalies has shown recent increases in ankyloglossia (tongue-tie) in British Columbia, Canada. We examined the temporal trends in ankyloglossia and its surgical treatment (frenotomy).

Methods: We conducted a population-based cohort study involving all live births in British Columbia from Apr. 1, 2004, to Mar. 31, 2014, with data obtained from the province's Perinatal Data Registry. Spatiotemporal trends in ankyloglossia and frenotomy, and associations with maternal and infant characteristics, were quantified using logistic regression analysis.

Results: There were 459 445 live births and 3022 cases of ankyloglossia between 2004 and 2013. The population incidence of ankyloglossia increased by 70% (rate ratio 1.70, 95% confidence interval [CI] 1.44–2.01), from 5.0 per 1000 live births in 2004 to 8.4 per 1000 in 2013. During the same period, the population rate of frenotomy increased by 89% (95% CI 52%–134%), from 2.8 per 1000 live births in 2004 to 5.3 per 1000 in 2013. The 2 regional health authorities with the lowest population rates of frenotomy (1.5 and 1.8 per 1000 live births) had the lowest rates of ankyloglossia and the lowest rates of frenotomy among cases with ankyloglossia, whereas the 2 regional health authorities with the highest population rates of frenotomy (5.2 and 5.3 per 1000 live births) had high rates of ankyloglossia and the highest rates of frenotomy among cases of ankyloglossia. Nulliparity, multiple birth, male infant sex, birth weight and year were independently associated with ankyloglossia.

Interpretation: Large temporal increases and substantial spatial variations in ankyloglossia and frenotomy rates were observed that may indicate a diagnostic suspicion bias and increasing use of a potentially unnecessary surgical procedure among infants.

Public health surveillance of congenital anomalies has a long tradition based in part on the lessons learned following experience with thalidomide and phocomelia,[1,2] and diethylstilbestrol and vaginal adenocarcinoma.[3] Outbreaks of congenital rubella syndrome have also served to reinforce the case for routine surveillance of congenital anomalies.[4] In Canada, there has been an increased focus on such surveillance over the previous decade, with the Public Health Agency of Canada providing regular reports on neural tube and congenital heart defects, Down syndrome, orofacial clefts, limb deficiency defects and gastroschisis.[5]

Recent surveillance activities carried out as part of routine monitoring of population perinatal health in the province of British Columbia, Canada,[6] led to the identification of a temporal increase in ankyloglossia (tongue-tie). The condition, characterized by an unusually short, thick or tight band of tissue (lingual frenulum) that limits the range of movement of the tongue, may interfere with feeding and speech. The unexpected increase in ankyloglossia in the province was initially categorized as an incidental finding and dismissed as being of no clinical or public health importance. However, clinicians and hospital staff attributed the rise to increased ascertainment owing to the Baby-Friendly Hospital Initiative of the World Health Organization (WHO) and UNICEF and to support for breastfeeding.[7] Increased focus on breastfeeding initiation before hospital discharge may have led to heightened diagnostic suspicion of ankyloglossia and a potential increase in surgical correction of the condition (frenotomy). We carried out a population-based study to examine the temporal trends and epidemiologic correlates in ankyloglossia.

Methods

We conducted a population-based cohort study involving all live births in British Columbia, Canada, using data obtained from the province's Perinatal Data Registry. This database contains detailed information on all mothers and babies in the

Competing interests: None declared.

This article has been peer reviewed.

Correspondence to: K.S. Joseph, kjoseph@cw.bc.ca

***CMAJ Open* 2016. DOI:10.9778/cmajo.20150063**

province that is collated by trained medical record abstractors using standardized forms and coding rules. Data quality is continually assessed by means of logic and consistency checks, and information in the database has been validated[8,9] and used extensively for health planning and research.[10]

We restricted the study period to fiscal years from Apr. 1, 2004, to Mar. 31, 2014 (hereafter referred to as years 2004 to 2013), when diagnoses and procedures among mothers and babies were consistently coded with the enhanced Canadian version of the 10th revision of the International Statistical Classification of Diseases and Related Health Problems (ICD-10-CA) and the Canadian Classification of Health Interventions (CCI), respectively.

Appropriate diagnostic and procedure codes were used to identify cases of ankyloglossia (ICD-10-CA code Q381) and tongue release (CCI code 1.FJ.72 for lingual frenectomy/frenotomy/frenulectomy or glossotomy for tongue-tie). We first examined rates of ankyloglossia by year, maternal characteristics (parity, body mass index [BMI] and plurality) and infant characteristics (sex, birth weight and gestational age). Rates of ankyloglossia were also quantified by length of hospital stay. The frequency of ankyloglossia and frenotomy was examined as a function of the rate of breast milk feeding and by regional health authority. The frequency of frenotomy among cases of ankyloglossia was examined by year to ascertain whether surgery was being increasingly used to treat the condition, and by regional health authority to ascertain potential spatial variation in surgery rates.

The precision of rate estimates was quantified using exact binomial 95% confidence intervals (CIs), and the significance of temporal trends in rates was evaluated using a χ^2 test for linear trend in proportions. Associations between year, maternal characteristics and infant characteristics, and ankyloglossia were quantified using proportion type rate ratios with 95% CIs. Logistic regression was used to obtain adjusted odds ratios expressing the association between year, maternal characteristics and infant characteristics, and ankyloglossia. Because the frequency of ankyloglossia was low, odds ratios obtained from logistic models were interpreted as rate ratios. All analyses were carried out using SAS version 9.3 (SAS Institute Inc.).

The study was approved by the Research Ethics Board of the University of British Columbia.

Results

There were 459 445 live births and 3022 cases of ankyloglossia in the province during the study period, for a birth prevalence of 6.6 cases per 1000 live births (95% CI 6.3–6.8). The rate of ankyloglossia increased from 5.0 (95% CI 4.3–5.7) per 1000 live births in 2004 to 6.4 (95% CI 5.7–7.2) per 1000 in 2008, and to 8.4 (95% CI 7.6–9.3) per 1000 in 2013 (p for linear trend < 0.001; Figure 1A). Rates of ankyloglossia varied significantly between the 5 regional health authorities; the lowest and highest rates were 3.8 per 1000 and 8.2 per 1000 live births, respectively (odds ratio for highest v. lowest 2.15, 95% CI 1.92–2.40).

The frequency of ankylogossia decreased with maternal age; infants of women aged 40 years or more were 0.81 (95% CI 0.66–1.00) times as likely to have ankyloglossia as infants of mothers aged 20–24 years (Table 1). The declining trend in ankyloglossia with increasing maternal age was not statistically significant (p for linear trend 0.20). Infants of nulliparous women and women with a BMI of 30 or greater had relatively high rates of ankyloglossia; the p value for the linear trend in ankyloglossia rates with increasing BMI was 0.06. Male infant sex was associated with a higher rate of ankyloglossia (rate ratio 1.72, 95% CI 1.60–1.85), whereas preterm infants had relatively lower rates compared with term and post-term infants (p for linear trend < 0.001). Birth weight was also associated with ankyloglossia: compared with infants with a birth weight of 3000–3499 g, those weighing less than 3000 g had relatively low rates and infants with birth weights of 4000 g or more had relatively high rates. There was a significant linear trend in ankyloglossia rates with increasing birth weight ($p < 0.001$).

Table 2 shows the association between the length of hospital stay for the infant during the birth admission and rates of ankyloglossia. Rates of ankyloglossia increased steadily from 3.6 per 1000 live births among infants discharged within 24 hours after birth to 9.3 per 1000 among infants discharged 96–167 hours after birth. Infants whose hospital stay was 168 hours or longer had an ankyloglossia rate of 4.7 per 1000 live births.

Table 3 presents the results of the logistic regression analyses carried out among term infants. Significant determinants of ankyloglossia included nulliparity (v. multiparity; adjusted rate ratio [aRR] 1.47, 95% CI 1.36–1.59), BMI of 30 or greater (v. BMI 18–24; aRR 1.14, 95% CI 1.00–1.30), multiple birth (v. singleton; aRR 0.68, 95% CI 0.63–0.74), male infant sex (v. female; aRR1.74, 95% CI 1.61–1.89), and birth weight of 4000–4499 g and of 4500 g or more (v. 3000–3499 g; aRR 1.26, 95% CI 1.12–1.42, and aRR 1.39, 95% CI 1.11–1.76, respectively). Adjusted rates of ankyloglossia were also significantly higher in the years 2008 to 2013 compared with 2004 (aRR 1.70, 95% CI 1.43–2.02).

There were 1765 frenotomy procedures carried out in the study population, for a frenotomy rate of 3.8 (95% CI 3.7–4.0) per 1000 live births. The rate of frenotomy increased by 89% (95% CI 52%–134%), from 2.8 per 1000 live births in 2004 to 5.3 per 1000 live births in 2013. The association between maternal and infant characteristics and frenotomy among term infants (Table 4) was similar in direction and magnitude to the association between maternal and infant characteristics and ankyloglossia (Table 3).

The temporal increase in frenotomy rates paralleled the temporal increase in ankyloglossia rates (Figure 1A), and annual rates of ankyloglossia and frenotomy were closely correlated (correlation coefficient 0.98, $p < 0.001$). Rates of frenotomy varied between the different health authorities, with the lowest and highest rates being 1.8 per 1000 live births and 5.3 per 1000 (odds ratio for highest v. lowest 2.99, 95% CI 2.49–3.58). Both ankyloglossia and frenotomy rates increased as rates of breast milk feeding increased from 93% to 95% (Figure 1B); annual rates of ankyloglossia and frenotomy were closely correlated with breast milk feeding (correlation coefficient 0.93, $p < 0.001$ for ankyloglossia; correlation coefficient 0.92, $p < 0.001$ for frenotomy).

The rate of frenotomy among ankyloglossia cases was 58.0%; this rate increased nonsignificantly and in a nonlinear fashion from 57.1% in 2004 to 63.5% in 2013 (11% increase, 95% CI 3% decrease to 28% increase; Figure 1C). The 2 regional health authorities with the lowest population rates of frenotomy (1.5 and 1.8 per 1000 live births) had the lowest rates of ankyloglossia (3.8 and 4.6 per 1000 live births) and the lowest rates of frenotomy among cases of ankyloglossia (33.3%

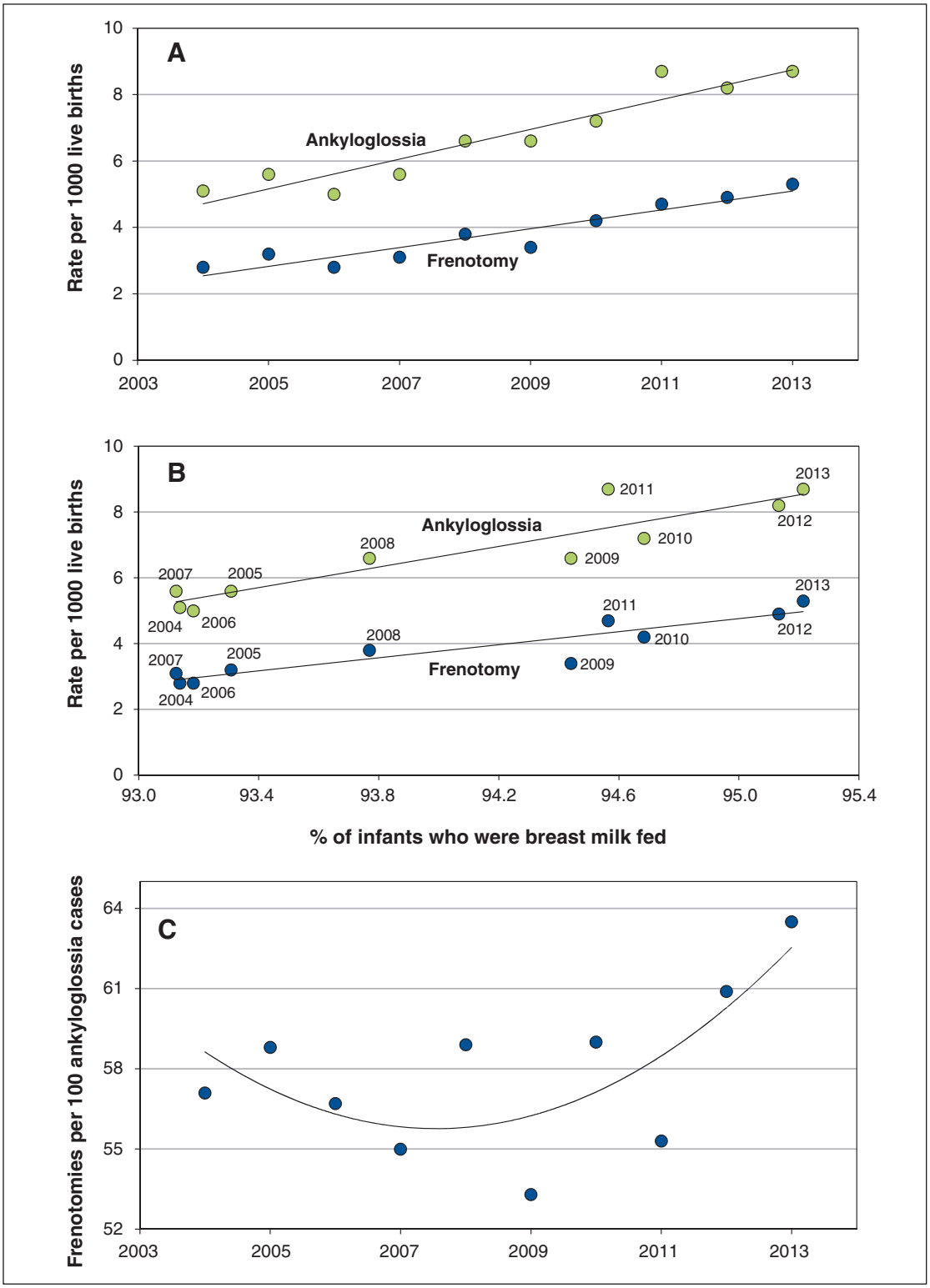

Figure 1: Rates of ankyloglossia and frenotomy by year (A) and by proportion of infants who were breast milk fed (B), and temporal trends in frenotomy among ankyloglossia cases (C).

Table 1: Frequency of ankyloglossia by maternal and infant characteristics among all live births, British Columbia, Canada, 2004–2013

Characteristic	No. of live births	Ankyloglossia		
		No. of cases	Rate per 1000 live births (95% CI)	Rate ratio (95% CI)
Maternal age, yr				
< 20	14 073	98	7.0 (5.7–8.5)	1.01 (0.81–1.25)
20–24 (ref)	61 794	428	6.9 (6.3–7.6)	1.00 (–)
25–29	126 550	811	6.4 (6.0–6.9)	0.93 (0.82–1.04)
30–34	151 380	1 016	6.7 (6.3–7.1)	0.97 (0.87–1.08)
35–39	85 717	557	6.5 (6.0–7.1)	0.94 (0.83–1.06)
≥ 40	19 927	112	5.6 (4.6–6.8)	0.81 (0.66–1.00)
Parity				
Nulliparous	214 690	1 656	7.7 (7.3–8.1)	1.38 (1.29–1.48)
Multiparous (ref)	244 728	1 366	5.6 (5.3–5.9)	1.00 (–)
Body mass index				
< 18	19 449	120	6.2 (5.1–7.4)	0.94 (0.78–1.13)
18–24 (ref)	194 100	1 276	6.6 (6.2–6.9)	1.00 (–)
25–29	66 173	439	6.6 (6.0–7.3)	1.01 (0.91–1.12)
≥ 30	40 720	302	7.4 (6.6–8.3)	1.13 (1.00–1.28)
Missing data	139 003	885	6.4 (6.0–6.8)	0.97 (0.89–1.05)
Plurality				
Singleton (ref)	443 374	2 956	6.7 (6.4–6.9)	1.00 (–)
Multiple	16 071	66	4.1 (3.2–5.2)	0.62 (0.49–0.79)
Gestational age,* wk				
28–36	39 573	209	5.3 (4.6–6.0)	0.75 (0.65–0.86)
37–43 (ref)	390 194	2 754	7.1 (6.8–7.3)	1.00 (–)
Missing data	27 832	55	1.9 (1.5–2.6)	0.28 (0.21–0.37)
Infant sex				
Male	237 230	1 957	8.2 (7.9–8.6)	1.72 (1.60–1.85)
Female (ref)	222 208	1 065	4.8 (4.5–5.1)	1.00 (–)
Birth weight, g				
< 1500	4 711	10	2.1 (1.0–3.9)	0.33 (0.18–0.62)
1500–1999	7 003	17	2.4 (1.4–3.9)	0.38 (0.24–0.61)
2000–2499	18 469	66	3.6 (2.8–4.5)	0.57 (0.45–0.72)
2500–2999	69 407	406	5.8 (5.3–6.4)	0.92 (0.85–1.00)
3000–3499 (ref)	163 673	1 075	6.6 (6.2–7.0)	1.00 (–)
3500–3999	137 549	967	7.0 (6.6–7.5)	1.04 (0.99–1.09)
4000–4499	46 636	393	8.4 (7.6–9.3)	1.23 (1.13–1.34)
≥ 4500	8 845	83	9.4 (7.5–11.6)	1.48 (1.19–1.84)
Missing data	3 152	5	1.6 (0.5–3.7)	0.25 (0.10–0.60)
Year				
2004 (ref)	42 802	212	5.0 (4.3–5.7)	1.00 (–)
2005	43 147	233	5.4 (4.7–6.1)	1.09 (0.90–1.31)
2006	44 208	215	4.9 (4.2–5.6)	0.98 (0.81–1.19)
2007	46 617	258	5.5 (4.9–6.3)	1.12 (0.93–1.34)
2008	47 491	304	6.4 (5.7–7.2)	1.29 (1.09–1.54)
2009	47 762	306	6.4 (5.7–7.2)	1.29 (1.09–1.54)
2010	46 592	327	7.0 (6.3–7.8)	1.41 (1.19–1.68)
2011	47 025	396	8.4 (7.6–9.3)	1.70 (1.44–2.01)
2012	47 320	379	8.0 (7.2–8.8)	1.62 (1.37–1.91)
2013	46 481	392	8.4 (7.6–9.3)	1.70 (1.44–2.01)
Overall	459 445	3 022	6.6 (6.3–6.8)	–

Note: CI = confidence interval, ref = reference category.
*Results for live births < 28 weeks' gestation not shown because of small number of cases (< 5).

and 43.3%). The 2 health authorities with the highest population rates of frenotomy (5.2 and 5.3 per 1000 live births) had high rates of ankyloglossia (7.3 and 8.2 per 1000 live births) and the highest rates of frenotomy among cases of ankyloglossia (63.6% and 72.1%).

Interpretation

Our study showed that rates of ankyloglossia and frenotomy in British Columbia increased significantly from 2004 to 2013. Nulliparity, obesity, male infant sex and macrosomia were positively associated with ankyloglossia, whereas preterm birth, and twin and higher-order plurality were protective factors. There was a monotonic increase in ankyloglossia rates associated with increasing duration of hospital stay from less than 24 hours to 96–167 hours. Maternal and infant characteristics associated with frenotomy were similar to those associated with ankyloglossia, and there was a two- to threefold variation in regional ankyloglossia and frenotomy rates.

The rate of ankyloglossia in our study (6.6 per 1000 live births) is substantially lower than estimates from previous studies that have typically reported birth prevalence rates of 4% to 10% (range 0.02%–11%).[11–16] This underestimation is not surprising, because the anomaly is usually asymptomatic and there is no consensus on the definition and diagnostic criteria for ankyloglossia. Diagnosis of ankyloglossia and other minor congenital anomalies during routine hospital admissions for childbirth is typically sporadic and symptom prompted (i.e., it is especially likely when problems arise with infant feeding). With the recent increased emphasis on breastfeeding initiation before hospital discharge because of the Baby Friendly Hospital Initiative, it is not surprising that ankyloglossia is being diagnosed with increasing frequency.

Our study showed that maternal nulliparity and obesity and infant male sex and macrosomia were positively associated with ankyloglossia, whereas preterm births and births of twins or triplets were negatively associated with ankyloglossia. However, the true association between these factors and ankyloglossia may have been somewhat distorted in our study because of the symptom-prompted nature of the diagnosis. The strength of the association between the risk/protective factors in our study was likely influenced by associations between the above-mentioned factors and breastfeeding difficulties.

There has been a long-standing controversy over the optimal management of infants with ankyloglossia and breastfeeding problems, and this is reflected in contemporary national and international guidelines. The Canadian Paediatric Society's 2011 and 2014 position statements on ankyloglossia state that frenotomy cannot be recommended based on the available evidence.[17,18] The Dutch and Japanese pediatric societies[19–21] also do not endorse frenotomy, whereas recommendations from the United Kingdom's National Institute for Health and Care Excellence mention both conservative management and early surgical division for cases of ankyloglossia associated with breastfeeding problems.[22] UNICEF's Baby Friendly Initiative[23] and the American Academy of Pediatrics[24] recommend early frenotomy for symptomatic ankyloglossia and describe it as a simple, safe and efficacious procedure. This lack of consensus on the treatment of ankyloglossia arises because of the quality of the evidence on the efficacy of frenotomy. Although several randomized trials have assessed the efficacy of frenotomy for ankyloglossia-associated breastfeeding difficulties,[25–29] they were all relatively small studies, with methodologic problems related to inclusion criteria, lack of blinding and subjectively defined outcomes.

The observed increase in the diagnosis of ankyloglossia appears to be a consequence of increased surveillance secondary to the focus on breastfeeding initiation and the Baby Friendly Hospital Initiative. Population rates of frenotomy in British Columbia exhibited a substantial spatial variation by regional health authority, as did rates of frenotomy among cases of ankyloglossia. This is concerning insofar as it reflects arbitrariness with regard to the diagnosis of ankyloglossia and in the use of a potentially unnecessary surgical procedure among newborns. The controversy with regard to the use of frenotomy has been framed as a conflict between lactation nurses, breastfeeding support groups and mothers who have experienced difficulties in breastfeeding versus pediatricians

Table 2: Frequency of ankyloglossia by length of infant's hospital stay among all live births, British Columbia, Canada, 2004–2013

		Ankyloglossia		
Length of stay, h	No. of live births	No. of cases	Rate per 1000 live births (95% CI)	Rate ratio (95% CI)
< 24	47 703	174	3.6 (3.1–4.2)	0.62 (0.53–0.72)
24–47 (ref)	182 578	1 079	5.9 (5.6–6.3)	1.00 (–)
48–71	104 509	850	8.1 (7.6–8.7)	1.38 (1.26–1.51)
72–95	59 906	546	9.1 (8.4–9.9)	1.54 (1.39–1.71)
96–167	28 940	268	9.3 (8.2–10.4)	1.57 (1.37–1.79)
≥ 168	13 404	63	4.7 (3.6–6.0)	0.80 (0.62–1.02)
Missing	22 405	42	1.9 (1.4–2.5)	0.32 (0.23–0.43)
Total	459 445	3 022	6.6 (6.3–6.8)	–

Note: CI = confidence interval, ref = reference category.

Table 3: Logistic regression analysis showing association between maternal and infant determinants and ankyloglossia among term and post-term (37–43 wk) live births, British Columbia, Canada, 2004–2013

Determinant	No. of term and post-term live births	No. of cases	Rate per 1000 live births	Rate ratio (95% CI)	
				Crude	Adjusted*
Age, yr					
< 20	11 504	92	8.0 (6.5–9.8)	1.07 (0.85–1.34)	0.98 (0.78–1.23)
20–24 (ref)	52 289	391	7.5 (6.8–8.3)	1.00 (–)	1.00 (–)
25–29	108 508	742	6.8 (6.4–7.3)	0.91 (0.81–1.03)	0.93 (0.83–1.06)
30–34	129 939	930	7.2 (6.7–7.6)	0.96 (0.85–1.08)	1.01 (0.90–1.14)
35–39	71 894	495	6.9 (6.3–7.5)	0.92 (0.81–1.05)	1.01 (0.88–1.16)
≥ 40	15 968	104	6.5 (5.3–7.9)	0.87 (0.70–1.08)	0.96 (0.78–1.20)
Parity					
0	180 970	1 519	8.4 (8.0–8.8)	1.42 (1.32–1.54)	1.47 (1.36–1.59)
≥ 1 (ref)	209 121	1 235	5.9 (5.6–6.2)	1.00 (–)	1.00 (–)
Body mass index					
< 18	16 473	106	6.4 (5.3–7.8)	0.92 (0.75–1.12)	0.94 (0.77–1.14)
18–24 (ref)	167 813	1 174	7.0 (6.6–7.4)	1.00 (–)	1.00 (–)
25–29	56 208	398	7.1 (6.4–7.8)	1.01 (0.90–1.13)	1.01 (0.90–1.14)
≥ 30	33 730	270	8.0 (7.1–9.0)	1.15 (1.00–1.31)	1.14 (1.00–1.30)
Missing data	115 881	806	7.0 (6.5–7.5)	0.99 (0.91–1.09)	1.06 (0.97–1.16)
Plurality					
Singleton (ref)	385 104	2 727	7.1 (6.8–7.4)	1.00 (–)	1.00 (–)
Multiple	4 995	27	5.4 (3.6–7.9)	0.76 (0.52–1.11)	0.68 (0.63–0.74)
Infant sex					
Female (ref)	190 993	966	5.1 (4.7–5.4)	1.00 (–)	1.00 (–)
Male	199 106	1 788	9.0 (8.6–9.4)	1.78 (1.65–1.93)	1.74 (1.61–1.89)
Birth weight,† g					
2000–2499	5 315	31	5.8 (4.0–8.3)	0.88 (0.61–1.25)	0.90 (0.63–1.29)
2500–2999	50 987	311	6.1 (5.4–6.8)	0.92 (0.81–1.04)	0.94 (0.82–1.06)
3000–3499 (ref)	149 808	996	6.6 (6.2–7.1)	1.00 (–)	1.00 (–)
3500–3999	131 133	945	7.2 (6.8–7.7)	1.08 (0.99–1.19)	1.06 (0.97–1.16)
4000–4499	44 505	389	8.7 (7.9–9.6)	1.32 (1.17–1.48)	1.26 (1.12–1.42)
≥ 4500	8 127	80	9.8 (7.8–12.2)	1.49 (1.18–1.87)	1.39 (1.11–1.76)
Year					
2004 (ref)	36 593	199	5.4 (4.7–6.2)	1.00 (–)	1.00 (–)
2005	36 791	218	5.9 (5.2–6.8)	1.09 (0.90–1.32)	1.09 (0.90–1.32)
2006	37 802	202	5.3 (4.6–6.1)	0.98 (0.81–1.20)	0.98 (0.80–1.19)
2007	39 743	232	5.8 (5.1–6.6)	1.07 (0.89–1.30)	1.07 (0.88–1.29)
2008	40 267	281	7.0 (6.2–7.8)	1.29 (1.07–1.54)	1.28 (1.06–1.53)
2009	40 367	282	7.0 (6.2–7.8)	1.29 (1.07–1.54)	1.28 (1.07–1.54)
2010	39 610	279	7.0 (6.2–7.9)	1.30 (1.08–1.56)	1.29 (1.08–1.55)
2011	39 827	366	9.2 (8.3–10.2)	1.70 (1.43–2.02)	1.70 (1.43–2.02)
2012	39 936	335	8.4 (7.5–9.3)	1.55 (1.30–1.84)	1.55 (1.30–1.85)
2013	39 169	360	9.2 (8.3–10.2)	1.70 (1.43–2.02)	1.70 (1.43–2.02)

Note: CI - confidence interval, ref = reference category.
*Adjusted for all other factors in the table.
†Results for live births < 2000 g not shown because of small number of cases (< 5).

Table 4: Logistic regression analysis showing association between maternal and infant determinants and frenotomy among term and post-term (37–43 wk) live births, British Columbia, Canada, 2004–2013

Determinant	No. of term and post-term live births	No. undergoing frenotomy	Rate per 1000 live births	Rate ratio (95% CI)	
				Crude	Adjusted*
Age, yr					
< 20	11 504	61	5.3 (4.1–6.8)	1.23 (0.92–1.65)	1.12 (0.84–1.49)
20–24 (ref)	52 289	225	4.3 (3.8–4.9)	1.00 (–)	1.00 (–)
25–29	108 508	447	4.1 (3.7–4.5)	0.96 (0.82–1.12)	0.98 (0.83–1.15)
30–34	129 939	546	4.2 (3.9–4.6)	0.98 (0.84–1.14)	1.04 (0.89–1.21)
35–39	71 894	281	3.9 (3.5–4.4)	0.91 (0.76–1.08)	1.01 (0.84–1.21)
≥ 40	15 968	50	3.1 (2.3–4.1)	0.73 (0.54–0.99)	0.82 (0.60–1.12)
Parity					
0	180 970	912	5.0 (4.7–5.4)	1.51 (1.37–1.67)	1.56 (1.40–1.72)
≥ 1 (ref)	209 121	698	3.3 (3.1–3.6)	1.00 (–)	1.00 (–)
Body mass index					
< 18	16 473	65	3.9 (3.0–5.0)	0.96 (0.74–1.24)	0.98 (0.76–1.27)
18–24 (ref)	167 813	691	4.1 (3.8–4.4)	1.00 (–)	1.00 (–)
25–29	56 208	231	4.1 (3.6–4.7)	1.00 (0.86–1.16)	0.99 (0.86–1.56)
≥ 30	33 730	166	4.9 (4.2–5.7)	1.20 (1.01–1.42)	1.18 (0.99–1.40)
Missing data	115 881	457	3.9 (3.6–4.3)	0.96 (0.85–1.08)	1.03 (0.91–1.16)
Plurality					
Singleton (ref)	385 104	1 596	4.1 (3.9–4.3)	1.00 (–)	1.00 (–)
Multiple	4 995	14	2.8 (1.5–4.7)	0.68 (0.40–1.14)	0.79 (0.46–1.36)
Infant sex					
Female (ref)	190 993	534	2.8 (2.6–3.0)	1.00 (–)	1.00 (–)
Male	199 106	1 076	5.4 (5.1–5.7)	1.93 (1.74–2.14)	1.87 (1.69–2.08)
Birth weight,† g					
2000–2499	5 315	17	3.2 (1.9–5.1)	0.84 (0.52–1.36)	0.87 (0.53–1.41)
2500–2999	50 987	163	3.2 (2.7–3.7)	0.84 (0.71–1.00)	0.86 (0.72–1.02)
3000–3499 (ref)	149 808	569	3.8 (3.5–4.1)	1.00 (–)	1.00 (–)
3500–3999	131 133	566	4.3 (4.0–4.7)	1.14 (1.01–1.28)	1.11 (0.99–1.25)
4000–4499	44 505	245	5.5 (4.8–6.2)	1.45 (1.25–1.68)	1.39 (1.19–1.62)
≥ 4500	8 127	49	6.0 (4.5–8.0)	1.59 (1.19–2.12)	1.49 (1.11–2.00)
Year					
2004 (ref)	36 593	116	3.2 (2.6–3.8)	1.00 (–)	1.00 (–)
2005	36 791	126	3.4 (2.9–4.1)	1.08 (0.84–1.39)	1.08 (0.84–1.39)
2006	37 802	112	3.0 (2.4–3.6)	0.93 (0.72–1.21)	0.93 (0.72–1.21)
2007	39 743	131	3.3 (2.8–3.9)	1.04 (0.81–1.33)	1.03 (0.80–1.33)
2008	40 267	164	4.1 (3.5–4.7)	1.28 (1.01–1.63)	1.28 (1.01–1.62)
2009	40 367	154	3.8 (3.2–4.5)	1.20 (0.95–1.53)	1.20 (0.94–1.53)
2010	39 610	165	4.2 (3.6–4.9)	1.31 (1.04–1.67)	1.31 (1.03–1.66)
2011	39 827	208	5.2 (4.5–6.0)	1.65 (1.31–2.07)	1.66 (1.32–2.08)
2012	39 936	204	5.1 (4.4–5.9)	1.61 (1.28–2.02)	1.62 (1.29–2.04)
2013	39 169	230	5.9 (5.1–6.7)	1.85 (1.48–2.31)	1.87 (1.49–2.33)

Note: CI = confidence interval, ref = reference category.
*Adjusted for all other factors in the table.
†Results for live births < 2000 g not shown because of small number of cases (< 5).

who are focused on the evidence for the efficacy of frenotomy.[30] The latter position is also informed by a culture that has increasingly rejected minor surgical intervention (e.g., tonsillectomy, ear tubes) for babies and children with the understanding that most conditions improve spontaneously.

Strengths and limitations

The strengths of our study included its population-based provenance, the large study size and detailed information on maternal and infant characteristics, diagnoses and procedures. Study limitations included the sporadic and symptom-prompted nature of the diagnosis of ankyloglossia, which likely distorted the birth prevalence. The bias also likely distorted the association between risk/protective factors and ankyloglossia, albeit to a lesser extent. Some transcription errors and missing values with regard to maternal and infant characteristics were also likely, because such deficiencies are inevitable in large population-based databases. Rates of ankyloglossia and frenotomy were estimated among those with missing information and such subjects were included in the regression analysis as a separate category. Other limitations included a lack of detail in our data source regarding the type of anesthesia used for frenotomy and the cost of the procedure. We included frenotomies carried out only in the first few days after birth, since our information was restricted to the birth admission.

Conclusion

Our study showed a temporal increase in the rate of ankyloglossia and a corresponding increase in the frequency of frenotomy in British Columbia. The wide spatial variation evident in population rates of frenotomy suggests arbitrary differences in practice patterns with regard to such surgery for newborns. Better diagnostic criteria for ankyloglossia, clear indications for frenotomy, improved communication and more detailed clinical practice guidelines are necessary for ensuring that infants with breastfeeding problems due to ankyloglossia are treated appropriately. Surgical treatment for ankyloglossia should be available when indicated, and unnecessary surgery in infancy avoided.

References

1. McBride WG. Thalidomide and congenital abnormalities. *Lancet* 1961;278:1358.
2. Lowry RB, Thunem NY, Anderson-Redick S. Alberta Congenital Anomalies Surveillance System. *CMAJ* 1989;141:1155-9.
3. Herbst AL, Ulfelder H, Poskanzer DC. Adenocarcinoma of the vagina. Association of maternal stilbestrol therapy with tumor appearance in young women. *N Engl J Med* 1971;284:878-81.
4. Elimination of rubella and congenital rubella syndrome — United States, 1969–2004. *MMWR Morb Mortal Wkly Rep* 2005;54:279-82.
5. *Congenital anomalies in Canada 2013: a perinatal health surveillance report*. Ottawa: Public Health Agency of Canada; 2013.
6. Joseph KS, Metcalfe A, Kinniburgh B, et al. Temporal trends in the birth prevalence of congenital anomalies in British Columbia, Canada, 2000 to 2011 [lecture]. Healthy Mothers and Healthy Babies: New Research and Best Practice Conference; 2014 Feb. 21–22; Vancouver.
7. *Baby-Friendly Hospital Initiative. Revised, updated and expanded for integrated care*. Geneva: World Health Organization and UNICEF; 2009.
8. Frosst G. Validity of pre-pregnancy body mass index (BMI) information derived from a population-based perinatal database [lecture]. Healthy Mothers and Healthy Babies: New Research and Best Practice Conference; 2014 Feb. 21–22; Vancouver.
9. Frosst G, Hutcheon J, Joseph KS, et al. Validating the British Columbia Perinatal Data Registry: a chart re-abstraction study. *BMC Pregnancy Childbirth* 2015;15:123.
10. Special reports: data and surveillance. Vancouver: Perinatal Services BC; 2015. Available: www.perinatalservicesbc.ca/health-professionals/data-surveillance (accessed 2016 Jan. 14).
11. Messner AH, Lalakea ML, Aby J, et al. Ankyloglossia: incidence and associated feeding difficulties. *Arch Otolaryngol Head Neck Surg* 2000;126:36-9.
12. Ballard JL, Auer CE, Khoury JC. Ankyloglossia: assessment, incidence, and effect of frenuloplasty on the breastfeeding dyad. *Pediatrics* 2002;110:e63.
13. Griffiths DM. Do tongue ties affect breastfeeding? *J Hum Lact* 2004;20:409-14.
14. Ricke LA, Baker N, Madlon-Kay D, et al. Newborn tongue-tie: prevalence and effect on breast-feeding. *J Am Board Fam Pract* 2005;18:1-7.
15. 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.
16. Segal LM, Stephenson R, Dawes M, et al. Prevalence, diagnosis, and treatment of ankyloglossia: methologic review. *Can Fam Physician* 2007;53:1027.
17. Rowan-Legg A; *Canadian Paediatric Society. Ankyloglossia and breastfeeding* [position statement]. Ottawa: Canadian Paediatic Society; 2005:209-13.
18. Rowan-Legg A. Ankyloglossia and breastfeeding. *Paediatr Child Health* 2011; 16:222.
19. Post ED, Rupert AW, Schulpen TW. Problematic breastfeeding due to a short frenulum [article in Dutch]. *Ned Tijdschr Geneeskd* 2010;154:A918.
20. Constantine AH, Williams C, Sutcliffe AG. A systematic review for ankyloglossia (tongue-tie) in breast fed infants. *Arch Dis Child* 2011;96(Suppl 1):A62-3.
21. Committee of Ethics, Japan Pediatric Society. Survey on operative treatment in ankyloglossia and its results. *Jpn Pediatr Soc* 2001;105:520-2.
22. *Division of ankyloglossia (tongue-tie) for breastfeeding*. NICE interventional procedure guidance [IPG149]. London (UK): National Institute for Health and Care Excellence; 2005. Available: www.nice.org.uk/guidance/ipg149 (accessed 2015 Mar. 31).
23. *What do mothers need to know about tongue-tie?* London (UK): UNICEF; Available: www.unicef.org.uk/BabyFriendly/Parents/Problems/Tongue-Tie/Division-of-tongue-tie/ (accessed 2015 Mar. 31).
24. *Breastfeeding: best for baby and mother*. Elk Grove Village (IL): American Academy of Pediatrics; 2004. Available: www2.aap.org/breastfeeding/files/pdf/BBM-8-27%20Newsletter.pdf (accessed 2015 Mar. 31).
25. Hogan M, Wescott C, Griffiths M. Randomised controlled trial of division of tongue-tie in infants with feeding problems. *J Paediatr Child Health* 2005;41:246.
26. Dollberg S, Botzer E, Grunis E, et al. Immediate nipple pain relief after frenotomy in breast-fed infants with ankyloglossia: a randomized, prospective study. *J Pediatr Surg* 2006;41:1598-600.
27. Buryk M, Bloom D, Shope T. Efficacy of neonatal release of ankyloglossia: a randomized trial. *Pediatrics* 2011;128:280-8.
28. Berry J, Griffiths M, Westcott C. A double-blind, randomized, controlled trial on tongue-tie division and immediate effect on breastfeeding. *Breastfeed Med* 2012;7:189-93.
29. 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.
30. Power RF, Murphy JF. Tongue-tie and frenotomy in infants with breastfeeding difficulties: achieving a balance. *Arch Dis Child* 2015;100:489-94.

Affiliations: Perinatal Services BC (Joseph, Kinniburgh), Provincial Health Services Authority; Department of Obstetrics and Gynaecology (Joseph, Razaz, Sabr, Lisonkova), University of British Columbia, and Children's and Women's Hospital and Health Centre of British Columbia; School of Population and Public Health (Joseph, Razaz, Lisonkova), University of British Columbia, Vancouver, BC; Department of Obstetrics and Gynecology (Metcalfe), University of Calgary, Calgary, Alta.; Department of Obstetrics and Gynaecology (Sabr), College of Medicine, King Saud University, Riyadh, Saudi Arabia

Contributors: K.S. Joseph conceptualized and designed the study and analyzed the data. Brooke Kinniburgh, Amy Metcalfe, Neda Razaz, Yasser Sabr and Sarka Lisonkova contributed to the data analyses. K.S. Joseph drafted the manuscript, and all of the authors revised it critically, gave final approval of the version to be published and agreed to act as guarantors of the work.

Acknowledgements: K.S. Joseph is supported by the Child and Family Research Institute and by a chair in maternal, fetal and infant health services research from the Canadian Institutes of Health Research (grant no. APR-126338).

Supplemental information: For reviewer comments and the original submission of this manuscript, please see www.cmajopen.ca/content/4/1/E33/suppl/DC1