Akyloglossia: Where congenital anomaly surveillance meets the baby friendly initiative

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Abstract

Background: Routine surveillance of congenital anomalies shows recent increases in ankyloglossia (tongue-tie). We examined the clinical and epidemiologic correlates of ankyloglossia and its surgical treatment (frenotomy) since several pediatric societies have stated that frenotomy among newborns cannot be recommended based on current evidence.
Methods: We carried out a population-based study of all live births in British Columbia, Canada, from 2004 to 2013 with data obtained from the British Columbia Perinatal Data Registry. Temporal trends in ankyloglossia and frenotomy, and associations with maternal and infant characteristics, were quantified using logistic regression.

Results: Rates of ankyloglossia rates increased by 70% (95% confidence interval [CI] 44-101%) from 5.0 in 2004 to 8.4 per 1,000 live births in 2013. Frenotomy rates increased from 2.8 in 2004 to 5.3 per 1,000 live births in 2013 (89% increase, 95% CI 52-134%). The regional health authorities with the lowest frenotomy rates (1.5 and 1.8 per 1,000 live births) had the lowest rates of ankyloglossia (3.8 and 4.6 per 1000 live births) and frenotomy given ankylogolssia (33.3% and 43.3%); the health authorities with the highest rates of frenotomy (5.2 and 5.3 per 1,000 live births) had high rates of ankyloglossia (7.3 and 8.2 per 1,000 live births) and the highest rates of frenotomy given ankyloglossia (63.6% and 72.1%). Nulliparity, multiple birth, male infant sex and birth weight were independently associated with ankyloglossia.

Interpretation: Spatio-temporal variations in ankyloglossia and frenotomy rates highlight the possibility of a diagnostic suspicion bias and increasing use of a potentially unnecessary surgery for infants.

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Introduction

Public health surveillance for congenital anomalies has a long tradition based partly on the lessons learnt following experience with thalidomide and phocomelia [1,2], and diethylstilbesterol and vaginal adenocarcinoma [3]. Outbreaks of congenital rubella syndrome have also served to reinforce the case for routine congenital anomaly surveillance [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 British Columbia, Canada [6], led to the identification of a temporal increase in ankyloglossia (tongue tie). This condition, characterized by an unusual band of tissue (lingual frenulum) that limits the range of movement of the tongue, can potentially interfere with feeding and speech. The unexpected increase in ankyloglossia in British Columbia 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 in ankyloglossia to increased ascertainment because of the Baby Friendly Hospital Initiative and support for breast feeding [7]. Increased focus on breast feeding initiation before hospital discharge may have led to heightened diagnostic suspicion of ankyloglossia and a potential increase in surgical correction of the condition (frenotomy). This issue is important because the Canadian Paediatric Society [8,9] (and some but not all pediatric societies [10-15]) have stated that frenotomy among newborns cannot be recommended based on current evidence. We therefore carried out a population-based study to examine the clinical and epidemiologic correlates of ankyloglossia.

Methods

The study included all live births in British Columbia, Canada, between April 1, 2004 and March 31, 2014, with data obtained from the British Columbia Perinatal Data Registry. This

population-based database contains detailed information on all mothers and babies in the province. Information in the database is collated by trained medical record abstractors using standardized forms and coding rules. Data quality is continually assessed by logic and consistency checks, and information in the database has been validated [16,17] and used extensively for health planning and research [18]. We restricted the study to the fiscal years from April 2004 to March 2014 (hereafter referred to as fiscal years 2004 to 2013) when diagnoses and procedures among mothers and babies were consistently coded with International Classification of Diseases version 10 (ICD10-CA) and the Canadian Classification of Health Interventions (CCI), respectively.

Appropriate diagnosis and procedure codes were used to identify cases of ankyloglossia (code Q381) and frenotomy (code 1FJ72 for frenectomy, lingual; frenotomy, lingual; frenulectomy, lingual and glossotomy, for tongue tie). We first examined rates of ankyloglossia by year, maternal characteristics (namely, parity, body mass index and plurality) and infant characteristics (namely 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 if surgery was being increasingly used to treat the condition and by regional health authority in order to ascertain potential spatial variation in surgery rates.

The precision of rate estimates was quantified using exact binomial 95% confidence intervals (CI) and the significance of temporal trends in rates was evaluated using a chi-square test for a linear trend in proportions. Associations between year, maternal characteristics and infant characteristics and ankyloglossia were quantified using proportion type rate ratios with 95% confidence intervals. Logistic regression was used to obtain adjusted odds ratios expressing the association between year, maternal characteristics and infant characteristics and ankyloglossia. Since the frequency of ankyloglossia was low, odds ratio obtained from logistic models were interpreted as rate ratios. All analyses were carried out using SAS version 9.3 (SAS Institute Inc., Cary, NC, USA). The study was approved by the ethics review committee of the University of British Columbia (H13-00688).

Results

There were 459,445 live births and 3,022 cases of ankyloglossia in British Columbia between 2004 and 2013, yielding a birth prevalence of 6.6 per 1,000 live births (95% CI 6.3-6.8). The rate of ankyloglossia increased from 5.0 (95% CI 4.3-5.7) in 2004 to 6.4 (95% CI 5.7-7.2) in 2008, and to 8.4 (95% CI 7.6-9.3) per 1,000 live births in 2013 (P for linear trend <0.0001; 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 highest vs lowest 2.15, 95% CI 1.92-2.40).

The frequency of ankylogossia decreased with maternal age; infants of women \geq 40 years of age were 0.81 (95% CI 0.66-1.00) times as likely to have ankyloglossia as compared with infants of mothers aged 20-24 years (Table 1). However, 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 \geq 30 kg/m² had relatively high rates of ankyloglossia;

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the increase in ankyloglossia rates with increasing BMI was of borderline significance (P for linear trend 0.06). Male infant sex was associated with a higher rate of ankyloglossia (rate ratio 1.72, 95% CI 1.60-1.85), while preterm infants had relatively lower rates compared with term and post-term infants (P for linear trend <0.0001). Birth weight was also associated with ankyloglossia; infants weighing <3,000 g had relatively low rates, while infants with birth weights \geq 4,000 g had relatively high rates compared with infants with a birth weight between 3,000 and 3,499 g. There was a significant linear trend in ankyloglossia rates with increasing birth weight (P <0.0001).

Table 2 shows the relationship 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 1,000 live births among infants discharged within 24 hours after birth to 5.9, 8.1, 9.1 and 9.3 per 1,000 live births among infants with a length of stay of 24-47 hours, 48-71 hours, 72-95 hours and 96-167 hours, respectively. Infants admitted in hospital for 168 hours or more had an ankyloglossia rate of 4.7 per 1,000 live births.

Table 3 presents the results of the logistic regression analyses carried out among term infants; significant determinants of ankyloglossia included nulliparity (vs multiparity; adjusted rate ratio [aRR] 1.47, 95% CI 1.36-1.59), BMI \geq 30 kg/m² (vs BMI 18-24; aRR 1.14, 95% CI 1.00-1.30), multiple birth vs singleton; aRR 0.68, 95% CI 0.63-0.74), male infant sex (vs female; aRR1.74, 95% CI 1.61-1.89), birth weight 4000-4499 g and \geq 4500 g (vs birth weight 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 2013 vs 2004 1.70, 95% CI 1.43-2.02).

There were 1,765 frenotomy procedures carried out among study infants, yielding 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 was similar in direction and magnitude to the association between maternal and infant characteristics and ankyloglossia (Table 4).

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 value <0.001). Rates of frenotomy varied between the different health authorities with the lowest and highest rates being 1.8 per 1,000 and 5.3 per 1,000 live births (odds ratio for frenotomy: highest vs 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 value <0.0001 for ankyloglossia and correlation coefficient 0.92, P value <0.0001 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 to 28%; Figure 1C). The 2 regional health authorities with the lowest frenotomy rates (1.5 and 1.8 per 1,000 live births) had the lowest rates of ankyloglossia (3.8 and 4.6 per 1000 live births) and frenotomy given ankylogolssia (33.3% and 43.3%). On the other hand, the 2 health authorities with the highest rates of frenotomy (5.2 and 5.3 per 1,000 live births) had high rates of ankyloglossia (7.3 and 8.2 per 1,000 live births) and the highest rates of frenotomy given ankyloglossia (63.6% and 72.1%).

Discussion

Our study showed that rates of ankyloglossia increased by 70% from 5.0 per 1,000 live births in 2004 to 8.4 per 1,000 live births in 2013. Over the same period, rates of frenotomy increased by 89% from 2.8 per 1,000 live births in 2004 to 5.3 per 1,000 live births in 2013. Nulliparity, obesity, male infant sex and macrosomia were positively associated with ankyloglossia, while preterm birth and twin and higher-order plurality were protective factors. There was a monotonic increase in ankyloglossia rates with duration of hospital stay from <24 hours to 96-167 hours. Maternal and infant characteristics associated with frenotomy were similar to those associated ankyloglossia and there was a 2- to 3-fold variation in regional ankyloglossia and frenotomy rates.

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 relationship 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 as such deficiencies are inevitable in large population-based databases.

The rate of ankyloglossia in our study was 6.6 per 1000 live births and this is significantly lower than estimates from previous studies that have typically yielded birth prevalence rates of 4% to 10% (range 0.02% to 11%) [19-24]. This underestimation of prevalence is not surprising as there is no consensus on the definition and diagnostic criteria for ankyloglossia and the anomaly is typically asymptomatic. Diagnosis of ankyloglossia and other minor congenital anomalies during routine hospitalization 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 breast feeding initiation before hospital discharge because of the Baby Friendly Hospital Initiative, it is not surprising that ankylogossia is being diagnosed with increasing frequency.

Our study showed that maternal nulliparity and obesity and infant's male sex, and macrosomia were positively associated, while preterm birth and multiple births were negatively associated with ankyloglossia. However, the true relationship 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 breast feeding difficulties.

There has been a longstanding controversy over the optimal management of infants diagnosed with ankylogossia and breast feeding 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 [8,9]. The Dutch and Japanese Pediatric Societies [10-12] also do not endorse frenotomy, while the United Kingdom's National Institute for Health and Care Excellence recommendations mention both conservative management and early surgical division for cases of ankyloglossia associated with breast feeding problems [13]. UNICEF's Baby Friendly Hospital Initiative [14] and the American Academy of Pediatrics [15] 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 been

carried out to assess the efficacy of frenotomy for ankyloglossia associated breast feeding difficulties [25-29], they are all relatively small studies, with methodological problems related to inclusion criteria, lack of blinding and subjectively defined outcomes.

Rates of frenotomy in British Columbia exhibited a substantial spatial variation by regional health authority, as did rates of frenotomy given 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 newborn infants. The controversy with regard to the use of frenotomy has been framed as a conflict between lactation nurses, breast feeding support groups and mothers who have experienced difficulties in breast feeding versus pediatricians who are focussed 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.

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

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

Maternal and infant	Number of	, <u>, e</u>				
characteristic	live births	Number	Rate/1,000 live	Rate ratio		
			births (95% CI)	(95% CI)		
Maternal age (years): <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 (-		
BMI (kg/m2): <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	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		
Gest age (weeks): <28*	1,846	<5	<2.7 (0.9-6.3)	<0.38 (0.16-0.92		
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	27,832	55	1.9 (1.5-2.6)	0.28 (0.21-0.37		
Birth weight (g): <1500	4,711	10	2.1 (1.0-3.9)	0.33 (0.18-0.62		
1500-1999	7,003	10	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	3,152	5	1.6 (0.5-3.7)	0.25 (0.10-0.60		
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 (-		
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		
2005	44,208	215	4.9 (4.2-5.6)	0.98 (0.81-1.19		
2000	46,617	213	5.5 (4.9-6.3)	1.12 (0.93-1.34		
2007 2008	40,017 47,491	238 304	6.4 (5.7-7.2)	1.12 (0.93-1.34		
2008 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		
Total	459,445	3,022	6.6 (6.3-6.8)	-		

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

Length of stay	Number of live – births	Ankyloglossia					
(hours)		Number	Rate	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)	-			

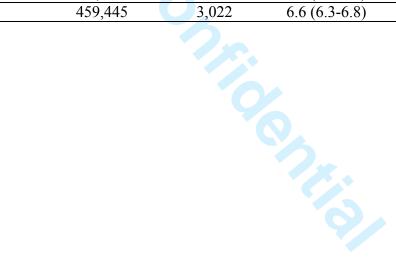


Table 3. Logistic regression analyses showing crude and adjusted rate ratios (aRR) with 95% confidence intervals (CI) expressing the association between maternal and infant determinants and ankyloglossia among term (3-43 weeks) live births, British Columbia, Canada, 2004 to 2013.

Determinants	Live	Ankyloglossia		RR	95% CI	aRR	95% CI
	births	Number	Rate				
Age <20 years	11,504	92	8.0	1.07	0.85-1.34	0.98	0.78-1.23
20-24 (ref.)	52,289	391	7.5	1.00	(-)	1.00	(-
25-29	108,508	742	6.8	0.91	0.81-1.03	0.93	0.83-1.0
30-34	129,939	930	7.2	0.96	0.85-1.08	1.01	0.90-1.14
35-39	71,894	495	6.9	0.92	0.81-1.05	1.01	0.88-1.1
≥ 40	15,968	104	6.5	0.87	0.70-1.08	0.96	0.78-1.2
Parity 0	180,970	1,519	8.4	1.42	1.32-1.54	1.47	1.36-1.5
≥ 1 (ref.)	209,121	1,235	5.9	1.00	(-)	1.00	(-
BMI (kg/m^2) : <18	16,473	106	6.4	0.92	0.75-1.12	0.94	0.77-1.1
18-24 (ref.)	167,813	1,174	7.0	1.00	(-)	1.00	(-
25-29	56,208	398	7.1	1.01	0.90-1.13	1.01	0.90-1.1
≥ 30	33,730	270	8.0	1.15	1.00-1.31	1.14	1.00-1.3
Missing	115,881	806	7.0	0.99	0.91-1.09	1.06	0.97-1.1
Plurality: Singleton (ref.)	385,104	2,727	7.1	1.00	(-)	1.00	(-
Multiple	4,995	27	5.4	0.76	0.52-1.11	0.68	0.63-0.7
Infant sex: Female (ref.)	190,993	966	5.1	1.00	(-)	1.00	(-
Male	199,106	1,788	9.0	1.78	1.65-1.93	1.74	1.61-1.8
Birth wt.*: 2000-2499 g	5,315	31	5.8	0.88	0.61-1.25	0.90	0.63-1.2
2500-2999	50,987	311	6.1	0.92	0.81-1.04	0.94	0.82-1.0
3000-3499 (ref.)	149,808	996	6.6	1.00	(-)	1.00	(-
3500-3999	131,133	945	7.2	1.08	0.99-1.19	1.06	0.97-1.1
4000-4499	44,505	389	8.7	1.32	1.17-1.48	1.26	1.12-1.4
≥4500	8,127	80	9.8	1.49	1.18-1.87	1.39	1.11-1.7
Year 2004 (ref.)	36,593	199	5.4	1.00	(-)	1.00	(-
2005	36,791	218	5.9	1.09	0.90-1.32	1.09	0.90-1.3
2006	37,802	202	5.3	0.98	0.81-1.20	0.98	0.80-1.1
2007	39,743	232	5.8	1.07	0.89-1.30	1.07	0.88-1.2
2008	40,267	281	7.0	1.29	1.07-1.54	1.28	1.06-1.5
2009	40,367	282	7.0	1.29	1.07-1.54	1.28	1.07-1.5
2010	39,610	279	7.0	1.30	1.08-1.56	1.29	1.08-1.5
2011	39,827	366	9.2	1.70	1.43-2.02	1.70	1.43-2.0
2012	39,936	335	8.4	1.55	1.30-1.84	1.55	1.30-1.8
2013	39,169	360	9.2	1.70	1.43-2.02	1.70	1.43-2.0

*Results for live births <2000 g not shown because of small numbers (<5 cases).

Table 4. Logistic regression analyses showing crude and adjusted rate ratios (aRR) with 95% confidence intervals (CI) expressing the association between maternal and infant determinants and frenotomy among term (37-43 weeks) live births, British Columbia, Canada, 2004 to 2013.

Determinants	Live	Frenotomy		RR	95% CI	aRR	95% CI
	births	Number	Rate	-			
Age <20 years	11,504	92	8.0	1.86	1.46-2.37	1.12	0.84-1.49
20-24 (ref.)	52,289	225	4.3	1.00	(-)	1.00	(-)
25-29	108,508	447	4.1	0.96	0.82-1.12	0.98	0.83-1.15
30-34	129,939	546	4.2	0.98	0.84-1.14	1.04	0.89-1.21
35-39	71,894	281	3.9	0.91	0.76-1.08	1.01	0.84-1.21
≥ 40	15,968	50	3.1	0.73	0.54-0.99	0.82	0.60-1.12
Parity 0	180,970	912	5.0	1.51	1.37-1.67	1.56	1.40-1.72
≥ 1 (ref.)	209,121	698	3.3	1.00	(-)	1.00	(-)
BMI (kg/m^2) : <18	16,473	65	3.9	0.96	0.74-1.24	0.98	0.76-1.27
18-24 (ref.)	167,813	691	4.1	1.00	(-)	1.00	(-)
25-29	56,208	231	4.1	1.00	0.86-1.16	0.99	0.86-1.56
≥ 30	33,730	166	4.9	1.20	1.01-1.42	1.18	0.99-1.40
Missing	115,881	457	3.9	0.96	0.85-1.08	1.03	0.91-1.16
Plurality: Singleton (ref.)	385,104	1,596	7.1	1.00	(-)	1.00	(-)
Multiple	4,995	14	2.8	0.68	0.40-1.14	0.79	0.46-1.36
Infant sex: Female (ref.)	190,993	534	2.8	1.00	(-)	1.00	(-)
Male	199,106	1,076	5.4	1.93	1.74-2.14	1.87	1.69-2.08
Birth wt.: 2000-2499 g	5,315	17	3.2	0.84	0.52-1.36	0.87	0.53-1.41
2500-2999	50,987	163	3.2	0.84	0.71-1.00	0.86	0.72-1.02
3000-3499 (ref.)	149,808	569	3.8	1.00	(-)	1.00	(-)
3500-3999	131,133	566	4.3	1.14	1.01-1.28	1.11	0.99-1.25
4000-4499	44,505	245	5.5	1.45	1.25-1.68	1.39	1.19-1.62
≥4500	8,127	49	6.0	1.59	1.19-2.12	1.49	1.11-2.00
Year 2004 (ref.)	36,593	116	3.2	1.00	(-)	1.00	(-)
2005	36,791	126	3.4	1.08	0.84-1.39	1.08	0.84-1.39
2006	37,802	112	3.0	0.93	0.72-1.21	0.93	0.72-1.21
2007	39,743	131	3.3	1.04	0.81-1.33	1.03	0.80-1.33
2008	40,267	164	4.1	1.28	1.01-1.63	1.28	1.01-1.62
2009	40,367	154	3.8	1.20	0.95-1.53	1.20	0.94-1.53
2010	39,610	165	4.2	1.31	1.04-1.67	1.31	1.03-1.66
2011	39,827	208	5.2	1.65	1.31-2.07	1.66	1.32-2.08
2012	39,936	204	5.1	1.61	1.28-2.02	1.62	1.29-2.04
2013	39,169	230	5.9	1.85	1.48-2.31	1.87	1.49-2.33

Results for live births <2000 g not shown because of small numbers (<5 cases).

Figure legend

Figure 1. Rates of ankyloglossia and frenotomy by year (panel A) and breast milk feeding (panel B), and temporal trends in frenotomy among ankyloglossia cases (panel C), British Columbia, Canada 2004 to 2013.

