Changes in the incidence and surgical treatment of ankyloglossia in Canada

K.S. Joseph MD, PhD
Professor
Department of Obstetrics and Gynaecology
School of Population and Public Health
University of British Columbia
Need to examine temporal trends in congenital anomalies

1. Public health surveillance obligation
   a) Identify unexpected increases in anomalies
   b) Evaluate effects of interventions (e.g., folic acid)
   c) Keep the public informed (dispel myths)

2. Data is collected routinely
What temporal trends do these curves show?
Thalidomide sales and CAs (phocomelia)

Graph showing the relation between the malformations of the thalidomide type and the sales of thalidomide (figures for Germany excluding Hamburg).

- - - Thalidomide sales (January 1961 = 100)
- - - 845 abnormalities of the thalidomide type (October 1961 = 100)

Hennina Sjostrom and Robert Nilsson, Thalidomide and the Power of the Drug Companies 1972
Canadian experience with thalidomide
1961-62 (115 cases)

Fig. 1.—Cases of congenital malformations associated with maternal thalidomide intake by month of birth—Canada, 1961-1962.
Thalidomide – the Canadian experience

Late 1959: Available as "samples"
April 1961: Licensed for prescription use
Dec 1961: Withdrawn from West Germany & UK
March 1962: Withdrawn from Canada
May 1962: Still available in some pharmacies

Over 100 cases in Canada

Not licensed in the US, thanks to Dr. Frances Kelsey
Methods

British Columbia’s Perinatal Data Registry
(maintained by Perinatal Services BC)

Information on all births in BC including home births

Data on all live births from April 2004 to March 2013

Ankyloglossia: ICD-10 CA code Q381

Frenotomy: CCI code 1.FJ.72
Results

Temporal trends in some congenital anomalies
All expected patterns except:
Increase in ankyloglossia (tongue tie)

Definition: A minor congenital anomaly characterized by a short, thickened or abnormally tight lingual frenulum, which may result in varying degrees of decreased tongue movement.

Incidental finding
459,445 live births

3,022 cases of ankyloglossia

Birth prevalence of 6.6 cases per 1000 live births (95% CI 6.3–6.8).

1,765 frenotomy procedures

Rate of 3.8 (95% CI 3.7–4.0) per 1000 live births
Ankyloglossia and frenotomy in BC
Frenotomy rates among cases of ankyloglossia

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Risk factors for ankyloglossia

<table>
<thead>
<tr>
<th>Determinant</th>
<th>No. of term and post-term live births</th>
<th>No. of cases</th>
<th>Rate per 1000 live births</th>
<th>Rate ratio (95% CI)</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
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<tr>
<td>Age, yr</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>&lt; 20</td>
<td>11 504</td>
<td>92</td>
<td>8.0 (6.5–9.8)</td>
<td>1.07 (0.85–1.34)</td>
</tr>
<tr>
<td>20–24 (ref)</td>
<td>52 289</td>
<td>391</td>
<td>7.5 (6.8–8.3)</td>
<td>1.00 (–)</td>
</tr>
<tr>
<td>25–29</td>
<td>108 508</td>
<td>742</td>
<td>6.8 (6.4–7.3)</td>
<td>0.91 (0.81–1.03)</td>
</tr>
<tr>
<td>30–34</td>
<td>129 939</td>
<td>930</td>
<td>7.2 (6.7–7.6)</td>
<td>0.96 (0.85–1.08)</td>
</tr>
<tr>
<td>35–39</td>
<td>71 894</td>
<td>495</td>
<td>6.9 (6.3–7.5)</td>
<td>0.92 (0.81–1.05)</td>
</tr>
<tr>
<td>≥ 40</td>
<td>15 968</td>
<td>104</td>
<td>6.5 (5.3–7.9)</td>
<td>0.87 (0.70–1.08)</td>
</tr>
<tr>
<td>Parity</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>0</td>
<td>180 970</td>
<td>1 519</td>
<td>8.4 (8.0–8.8)</td>
<td>1.42 (1.32–1.54)</td>
</tr>
<tr>
<td>≥ 1 (ref)</td>
<td>209 121</td>
<td>1 235</td>
<td>5.9 (5.6–6.2)</td>
<td>1.00 (–)</td>
</tr>
<tr>
<td>Body mass index</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>&lt; 18</td>
<td>16 473</td>
<td>106</td>
<td>6.4 (5.3–7.8)</td>
<td>0.92 (0.75–1.12)</td>
</tr>
<tr>
<td>18–24 (ref)</td>
<td>167 813</td>
<td>1 174</td>
<td>7.0 (6.6–7.4)</td>
<td>1.00 (–)</td>
</tr>
<tr>
<td>25–29</td>
<td>56 208</td>
<td>398</td>
<td>7.1 (6.4–7.8)</td>
<td>1.01 (0.90–1.13)</td>
</tr>
<tr>
<td>≥ 30</td>
<td>33 730</td>
<td>270</td>
<td>8.0 (7.1–9.0)</td>
<td>1.15 (1.00–1.31)</td>
</tr>
</tbody>
</table>

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### Risk factors for ankyloglossia (cont.)

<table>
<thead>
<tr>
<th>Determinant</th>
<th>No. of term and post-term live births</th>
<th>No. of cases</th>
<th>Rate per 1000 live births</th>
<th>Rate ratio (95% CI)</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td>Crude</td>
</tr>
<tr>
<td>Plurality</td>
<td></td>
<td></td>
<td></td>
<td>Adjusted*</td>
</tr>
<tr>
<td>Singleton (ref)</td>
<td>385,104</td>
<td>2,727</td>
<td>7.1 (6.8–7.4)</td>
<td>1.00 (–)</td>
</tr>
<tr>
<td>Multiple</td>
<td>4,995</td>
<td>27</td>
<td>5.4 (3.6–7.9)</td>
<td>0.76 (0.52–1.11)</td>
</tr>
<tr>
<td>Infant sex</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Female (ref)</td>
<td>190,993</td>
<td>966</td>
<td>5.1 (4.7–5.4)</td>
<td>1.00 (–)</td>
</tr>
<tr>
<td>Male</td>
<td>199,106</td>
<td>1,788</td>
<td>9.0 (8.6–9.4)</td>
<td>1.78 (1.65–1.93)</td>
</tr>
<tr>
<td>Birth weight, † g</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>2000–2499</td>
<td>5,315</td>
<td>31</td>
<td>5.8 (4.0–8.3)</td>
<td>0.88 (0.61–1.25)</td>
</tr>
<tr>
<td>2500–2999</td>
<td>50,987</td>
<td>311</td>
<td>6.1 (5.4–6.8)</td>
<td>0.92 (0.81–1.04)</td>
</tr>
<tr>
<td>3000–3499 (ref)</td>
<td>149,808</td>
<td>996</td>
<td>6.6 (6.2–7.1)</td>
<td>1.00 (–)</td>
</tr>
<tr>
<td>3500–3999</td>
<td>131,133</td>
<td>945</td>
<td>7.2 (6.8–7.7)</td>
<td>1.08 (0.99–1.19)</td>
</tr>
<tr>
<td>4000–4499</td>
<td>44,505</td>
<td>389</td>
<td>8.7 (7.9–9.6)</td>
<td>1.32 (1.17–1.48)</td>
</tr>
<tr>
<td>≥ 4500</td>
<td>8,127</td>
<td>80</td>
<td>9.8 (7.8–12.2)</td>
<td>1.49 (1.18–1.87)</td>
</tr>
</tbody>
</table>

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Correlation between breast milk feeding rates and ankyloglossia/frenotomy rates
Regional variation in ankyloglossia and frenotomy rates

Vancouver Island: 7.4
Fraser: 8.2
Northern: 7.3
Interior: 4.6
Vancouver Coastal: 3.8

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What’s the situation in Canada?

Methods

Data source: Discharge Abstract Database of the Canadian Institute for Health Information

All hospital live births (excluding Quebec)
April 2002 to March 2015
Ankyloglossia: ICD-10 CA code Q381
Frenotomy: CCI code 1.FJ.72
Results

Changes in the incidence and surgical treatment of ankyloglossia in Canada

Michelle Lisonek, Shiliang Liu MB, PhD, Susie Dzakpasu, Aideen M Moore MD, FRCPC, MHSc, K.S. Joseph MD, PhD for the Canadian Perinatal Surveillance System (Public Health Agency of Canada)

3,611,986 live births
40,457 cases of ankyloglossia yielding a Birth prevalence of 11.2 per 1,000 live births
24,975 cases of frenotomoty
Rate 6.91 per 1,000 live births.

Lisonek et al. Paediatrics and Child Health (in press)
Ankyloglossia and frenotomy in Canada

Lisonek et al. Paediatrics and Child Health (in press)
Temporal increase

Ankyloglossia
2002: 6.9 per 1,000 live births
2014: 22.6 per 1,000 live births (P value <0.001)

Frenotomy
2002: 3.8 per 1,000 live births
2014: 14.7 per 1,000 live births (P value 0.001)

Frenotomy rates among infants with ankyloglossia
2002: 54.7%
2014: 63.9%

Lisonek et al. Paediatrics and Child Health (in press)
Ankyloglossia by selected province/territory

Lisonek et al. Paediatrics and Child Health (in press)
Ankyloglossia by selected province/territory

Lisonek et al. Paediatrics and Child Health (in press)
Regional variation in frenotomy

Lowest vs highest rates of frenotomy, 2012-14 (per 1000 live births)

<table>
<thead>
<tr>
<th>Region</th>
<th>Rate 2012-14 (per 1000 live births)</th>
</tr>
</thead>
<tbody>
<tr>
<td>NFL</td>
<td>1.1</td>
</tr>
<tr>
<td>NU</td>
<td>4.2</td>
</tr>
<tr>
<td>BC</td>
<td>5.9</td>
</tr>
<tr>
<td>YU</td>
<td>20.2</td>
</tr>
<tr>
<td>AB</td>
<td>23.8</td>
</tr>
<tr>
<td>SA</td>
<td>24.5</td>
</tr>
</tbody>
</table>

In 2014, the highest rates were in

<table>
<thead>
<tr>
<th>Region</th>
<th>Rate 2014 (per 1000 live births)</th>
</tr>
</thead>
<tbody>
<tr>
<td>SA</td>
<td>25.1</td>
</tr>
<tr>
<td>AB</td>
<td>30.4</td>
</tr>
<tr>
<td>NS</td>
<td>42.1</td>
</tr>
</tbody>
</table>

Lisonek et al. Paediatrics and Child Health (in press)
Discussion

Rapid temporal increase in rates of ankyloglossia and frenotomy in Canada 2002 to 2014

Large regional variation

Trends appear to be accelerating

3-4% frenotomy rate in AB and NS in 2014
Discussion

Ankyloglossia rate 11.2 per 1,000 live births

Rate in research studies  4% (range 0.02% to 11%)

Diagnosis of ankyloglossia at the childbirth admission symptom prompted

Low rate in our study not surprising: no consensus on the definition and diagnostic criteria

Better tools may help with incorrect diagnosis
Discussion

Reasons for increase in ankyloglossia:

- Emphasis on breast feeding initiation before hospital discharge because of the Baby Friendly Hospital Initiative. Surveillance and detection bias?
- OR
- Earlier diagnosis and treatment

However, diagnostic uncertainty

- large regional differences

suggest this is due to a surveillance bias
Status of frenotomy for ankyloglossia

Canadian Paediatric Society discourages routine surgery for ankyloglossia

2011 and 2014: Frenotomy for ankyloglossia not recommended for newborns

2015: Not recommended for all cases; potential for benefit in cases with significant breast feeding difficulties recognized

Dutch and Japanese Pediatric Societies do not endorse

UK, UNICEF’s BFHI and American Academy of Pediatrics recommend it for symptomatic ankyloglossia
Clinical equipoise a result of

Lack of good quality evidence on the efficacy of frenotomy for problems with breast feeding

Randomized trials to-date have all been
• relatively small studies
• plagued by methodological problems (e.g. lack of blinding, subjectively defined outcomes)

• Systematic review by Francis et al (Pediatrics 2015) concluded that "with small, short-term studies with inconsistent methodology, ….strength of the evidence is low to insufficient"
Origins of the controversy

Conflict between Lactation Nurses, breast feeding support groups and mothers facing breast feeding problems versus Pediatricians who question the efficacy of frenotomy

Surveys of Lactation Consultants show
30%: ankyloglossia causes feeding problems occasionally
69%: frequently or always causes feeding difficulties

Surveys of Pediatricians show
90%: ankyloglossia rarely/never causes feeding problems
In conclusion

1. Desire to increase breast feeding initiation rates
2. No standardized criteria for ankyloglossia diagnosis
3. Lack of evidence favouring frenotomy

Have led to 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
Acknowledgements

**BC study:** Brooke Kinniburgh, Amy Metcalfe, Neda Razaz, Yasser Sabr, Sarka Lisonkova

**Canadian Study:** Michelle Lisonek, Shiliang Liu, Susie Dzakpasu, Aideen Moore

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