Population Demographic Indicators Associated With Incidence of Pyloric Stenosis

Teresa To, PhD; Anne Wajja, MD, MSc; Paul W. Wales, MD, MSc; Jacob C. Langer, MD

Objectives: To calculate incidence rates of pyloric stenosis (estimated by the rate of pyloromyotomy) among infants in Ontario and determine their association with population sociodemographic indicators.

Methods: Pyloromyotomy rates were calculated from hospital discharge data from 1993 through 2000. Four-year data (1993-1996 and 1997-2000) were combined to ensure the stability of the rates. Small-area variations in pyloromyotomy rates and correlations between sociodemographic indicators were studied.

Results: Approximately 84.0% of the patients were male infants (younger than 1 year). The sex-adjusted pyloromyotomy rates were 1.57 and 1.86 per 1000 with a 3.4-fold and 3.0-fold regional variation in 1993-1996 and 1997-2000, respectively. Urban areas consistently had the lowest pyloromyotomy rate (1.04 and 1.11 per 1000 in Metropolitan Toronto), but the highest rates were from more rural areas (3.30 and 3.38 per 1000 in Quinte, Kingston, Rideau). After adjusting for socioeconomic status and availability of surgeons in the region, living in a rural area remained a significant factor associated with a higher incidence of pyloromyotomy. The risk of pyloromyotomy for an infant who lives in a region with more than two thirds of its area classified as rural was 1.79 (95% confidence interval, 1.23-2.61; P<.005).

Conclusions: The observed changes in incidence and a higher rate among male infants are consistent with results from previous comparative studies conducted in North America and Sweden. The rural/urban differences suggest that environmental influences related to living in these areas may have a role in the etiology of pyloric stenosis. Further research is needed to evaluate these differences.

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Pyloric Stenosis, also known as infantile hypertrophic pyloric stenosis (IHPS), is a relatively common condition affecting infants (younger than 1 year). Several predisposing risk factors have been associated with the condition, but its etiology remains largely unknown. Although nonoperative approaches to management have been attempted and reported over the past 50 years,\textsuperscript{1,6} the use of medical management remains controversial and is still not widely accepted in Europe and North America.\textsuperscript{7} Pyloromyotomy is therefore largely the standard treatment of IHPS,\textsuperscript{8} particularly in North America. Because it is highly unlikely that any confirmed cases of IHPS are treated by medical means in North America, pyloromyotomy is a good proxy measure for incidence of pyloric stenosis.

Previous epidemiological studies in IHPS have shown different trends in the 1970s, 1980s, and 1990s,\textsuperscript{9-17} but few have documented the association of geographic variations and population sociodemographic indicators with IHPS incidence rates. In the Greater Glasgow area, Sule et al\textsuperscript{11} showed an increase in the annual incidence of IHPS from 1980 to 1988 but not thereafter. Few studies have documented the association of geographic variations with incidence of pyloric stenosis. In Sweden, Hedbäck et al\textsuperscript{12} reported dramatic geographic differences in IHPS. Similar to the results of the study in Glasgow, the results from these authors indicate a substantial decline in incidence of IHPS from 1987 to 1996. Furthermore, the incidence of IHPS in south Sweden was 3 times greater than in the north.

The objective of this article is 3-fold: to calculate incidence rates of IHPS (estimated by the rate of pyloromyotomy) among infants in Ontario, to determine whether IHPS is associated with population sociodemographic indicators and the availability of surgeons, and to measure geographic variations of the incidence of IHPS.

Methods

Patient Data

Computerized data on hospital discharges from the Canadian Institute for Health Informa-
transferred to another hospital for pyloromyotomy. Detailed that the diagnosis was made at 1 hospital but the patient was present to reduce the possibility of duplicates in the event criteria required both the diagnosis code and procedure code to classification of Diseases, Ninth Revision, Clinical Modification (ICD-9) code 750.5 and a hospital admission for pyloromyotomy (International Classification of Diseases, Ninth Revision, Clinical Modification code 43.3) in fiscal years 1993-2000. The inclusion criteria required both the diagnosis code and procedure code to be present to reduce the possibility of duplicates in the event that the diagnosis was made at 1 hospital but the patient was transferred to another hospital for pyloromyotomy. Detailed description of this study population is available elsewhere.10

METHOD OF ANALYSIS

Because only infants younger than 12 months were included in this study, age standardization was not necessary; however, we used the method of direct standardization in calculating the sex-adjusted rates.21 The 1996 Canadian census population was used as the standard population in the direct standardization. All rates were calculated per 1000 children. We used the 1-df $\chi^2$ test to determine whether the rate of an area was statistically different from a standard or referent area.22,23 Discharge rates were reported by the patient’s district health council (DHC) of residence as determined by the residence codes. District health councils are local health planning and advisory organizations that report to the Ontario Ministry of Health and Long-Term Care (Toronto). A DHC was classified as rural based on the percent of its total population residing in non-CMA or non-CA regions. Geographic rates were adjusted for sex and were reported for 4-year intervals to ensure the stability of rates.

SMALL-AREA VARIATION ANALYSIS

We calculated 3 commonly used statistics to measure variation between DHCs in Ontario. The extremal quotient is the ratio of the highest to lowest rate. The coefficient of variation, which takes into account the population sizes, divides the standard deviation of the rates by the average rate, and the systematic component of variation measures the relative systematic component of variation in rates between regions by subtracting the random component of variance from the total variance.24-26 These methods have been widely used by health services researchers in characterizing small-area variation.27-30

REgressions

The logistic regression model was used to model the association or risk of high admissions for pyloromyotomy. The outcome variable used was the pyloromyotomy rate in each of the 16 DHCs. Independent factors considered in the regression model included sociodemographic variables such as household income, education, percent of the population that was English speaking, and percent of the residing area defined as rural. We used univariate regression to test the statistical significance of risk factors for pyloromyotomy. Inclusion of covariates in the final multivariable regression model was based in part on patterns of correlation, statistical significance, and evidence of confounding.

INCIDENCE TREND

A total of 1918 pyloromyotomies were performed during fiscal years 1993-2000 in Ontario. Approximately 84.0% of this patient population was male with a 4:1 male-female ratio. The overall rate of IHPS (measured by rate of pyloromyotomy) decreased from 1.81 per 1000 in 1993 to 1.48 per 1000 in 1996 (18.2% decline) and then increased to 1.98 per 1000 in 2000 (27.7% increase) (Figure). However, the overall trend of incidence of pyloromyotomy was not statistically significant ($P = .18$).

GEOGRAPHIC VARIATIONS WITHIN ONTARIO

Table 1 shows the sex-adjusted distribution of IHPS by DHCs in Ontario. The overall sex-adjusted rate of pyloromyotomy in fiscal years 1993-1996 and 1997-2000 were 1.57 and 1.86 per 1000, respectively. The detailed DHC-specific data showed that in both time periods, the Quinte, Kingston, Rideau DHC had the highest pyloromyotomy rate (3.30 and 3.38 per 1000 in 1993-1997 and 1996-2000). Metropolitan Toronto had one of the lowest rates in both time periods (1.04 and 1.11 per 1000 in 1993-
1996 and 1997-2000) representing a 3-fold high-low ratio. The overall variation among the DHCs in Ontario was moderately large, as summarized by the statistics in Table 1. Although most DHCs in Northern Ontario had a higher percentage of rural living, certain DHCs in Southern Ontario were also more than 50% rural, indicating neither specific nor significant geographical clustering of rural areas. Table 1 shows DHCs sorted in ascending order from highest to lowest percentage of rural living. The rates of pyloromyotomy in the Quinte, Kingston, Rideau DHC and the Metropolitan Toronto DHC were significantly different from the provincewide rate. The P values indicate the statistical difference in pyloromyotomy rates between each DHC and the provincewide rate based on the 1-df $\chi^2$ test. Four cases were excluded from this part of the analysis due to a lack of valid residence codes.

### Table 2. Risk of Pyloromyotomy Adjusted for Confounders by Logistic Regression

<table>
<thead>
<tr>
<th>Covariate</th>
<th>Unadjusted Odds Ratio (95% Confidence Interval)</th>
<th>Significant Variable Adjusted Odds Ratio (95% Confidence Interval)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Rural living</td>
<td>1.54 (1.09-2.17)*</td>
<td>1.79 (1.23-2.61)†</td>
</tr>
<tr>
<td>Home language</td>
<td>1.02 (1.01-1.03)†</td>
<td>1.01 (0.99-1.02)</td>
</tr>
<tr>
<td>English</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Low family income</td>
<td>1.01 (0.99-1.02)</td>
<td>1.01 (0.99-1.03)</td>
</tr>
<tr>
<td>Availability of surgeon</td>
<td>0.97 (0.95-0.99)†</td>
<td>0.97 (0.94-1.00)</td>
</tr>
<tr>
<td>in the region</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Below high school education</td>
<td>0.97 (0.91-1.03)†</td>
<td>0.94 (0.88-0.99)*</td>
</tr>
<tr>
<td>University education‡</td>
<td>0.97 (0.95-0.99)*</td>
<td>0.97 (0.94-0.99)</td>
</tr>
</tbody>
</table>

*P<.05.  †P<.005.  ‡The reference category is high school education.

### SOCIODEMOGRAPHIC INDICATORS

The association between pyloromyotomy and sociodemographic variables was modeled by logistic regression (Table 2). All available sociodemographic variables were included in the model initially and removed by a backward selection process. There was a negative association between low education and the risk of pyloromyotomy. After adjusting for socioeconomic status (measured by income and education), living in a rural area remained a significant factor associated with pyloromyotomy. The risk of pyloromyotomy for an infant who lived in a DHC with more than two thirds of its area classified as rural was 1.79 (95% confidence interval, 1.23-2.61; P<.005).
Table 3. Summary of Population-Based Studies Examining Trends of Infantile Hypertrophic Pyloric Stenosis

<table>
<thead>
<tr>
<th>Location</th>
<th>Source</th>
<th>Data Collection Period</th>
<th>Infantile Hypertrophic Pyloric Stenosis, No.</th>
<th>Incidence Rate per 1000</th>
<th>Trend</th>
<th>Geographic Variation</th>
</tr>
</thead>
<tbody>
<tr>
<td>Europe and the Commonwealth</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Northern Ireland (Belfast)</td>
<td>Dodge27</td>
<td>1957-1969</td>
<td>521</td>
<td>3.1 to 2.2</td>
<td>Declining (P&lt;0.05)</td>
<td>NA</td>
</tr>
<tr>
<td>United Kingdom (Wales)</td>
<td>Webb et al96</td>
<td>1970-1979</td>
<td>115</td>
<td>1.2 to 3.6</td>
<td>Increasing (P&lt;0.001)</td>
<td>NA</td>
</tr>
<tr>
<td>United Kingdom (Central Scotland)</td>
<td>Knox et al15</td>
<td>1974-1980</td>
<td>1176</td>
<td>2.3 to 3.5</td>
<td>Increasing (P&lt;0.001)</td>
<td>NA</td>
</tr>
<tr>
<td>United Kingdom (Liverpool)</td>
<td>Tarn and Chan13</td>
<td>1976-1988</td>
<td>237</td>
<td>1.5 to 2.2</td>
<td>Increasing (P&lt;0.01)</td>
<td>NA</td>
</tr>
<tr>
<td>Sweden</td>
<td>Hedbäck et al12</td>
<td>1987-1996</td>
<td>2157</td>
<td>2.7 to 0.85</td>
<td>Declining South is 3×north</td>
<td>NA</td>
</tr>
<tr>
<td>Western Australia</td>
<td>Hitchcock et al14</td>
<td>1971-1984</td>
<td>602</td>
<td>1.4 to 2.9</td>
<td>Increasing</td>
<td>NA</td>
</tr>
<tr>
<td>North America</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>United States (New York State)</td>
<td>Applegate and Druschel19</td>
<td>1983-1990</td>
<td>3742</td>
<td>2.4 to 1.7</td>
<td>Declining (P&lt;0.001)</td>
<td>NA</td>
</tr>
<tr>
<td>Canada (Saskatchewan)</td>
<td>Habbick and To10</td>
<td>1970-1985</td>
<td>813</td>
<td>3.6 to 2.5</td>
<td>Declining</td>
<td>NA</td>
</tr>
<tr>
<td>Canada (Ontario)</td>
<td>To et al (current study)</td>
<td>1995-2000</td>
<td>1918</td>
<td>1.8 to 1.5</td>
<td>Declining (1993-1996)</td>
<td>Rural is 2×urban</td>
</tr>
</tbody>
</table>

Abbreviation: NA, not applicable.

COMMENT

This study is the first population-based study in North America to document demographic and geographic variations with IHPS incidence rates. The Ontario provincial sex-adjusted rate of 1.86 per 1000 children is similar to the rates of 2.50 per 1000 live births reported by Habbick et al14 for Saskatchewan in 1978-1983 and the rate of 1.92 per 1000 live births reported by Hedbäck et al12 for Sweden in 1987-1996, but much lower than the rate of 3.60 per 1000 live births reported by Sule et al11 for the Greater Glasgow area in 1980-1996.

Previous epidemiological studies reported inconsistent findings showing both absence and presence of variations as well as upward and downward trends in IHPS. Although most of the population studies conducted in Europe showed an increasing trend,11,13,15,16 2 studies conducted in North America9,10 and 1 from Sweden12 showed a decline in IHPS incidence, which is similar to our study findings in the earlier time period (1993-1996). A declining trend was observed in Saskatchewan from 1970 to 1985,8 New York State from 1983 to 1990,10 and Sweden from 1987 to 1996.12 Studies conducted in early 1990 have consistently shown a decline in IHPS incidence rates. Sule et al11 in Scotland found an initial increase in early 1980s followed by a significant decline from 1988 to 1996. Our study using more recent data in Ontario showed an initial decline of IHPS incidence rates from 1.81 per 1000 in 1993 to 1.48 per 1000 in 1996 and then a 28% increase in trend from 1.55 per 1000 in 1997 to 1.98 per 1000 in 2000 (Table 3). Presently, no clear explanations can be given for the actual decline and subsequent increase from 1997. Albeit statistically significant, our results contribute to the current international "debate" and confirm findings from previous studies,9-17 which showed variations in IHPS incidence trend over time. It is imperative to continue to observe how the incidence of IHPS changes over the next 10 years. The time trends suggest that unknown environmental factors which change over time may have an impact in the IHPS incidence. Although not significant, the change in trend found in our study is unlikely to be artifactual. Canadian Institute for Health Information data used for this study was complete and reliable. The Canadian health care system is highly centralized, and all hospitals and community health centers in Ontario are mandated to submit data on hospital discharges to the Canadian Institute for Health Information. There has also been no change in reporting procedures during the study period. Therefore, the change in incidence cannot be attributed to changes in reporting.

The statistically significant negative relationship between pyloromyotomy and low education, an indicator of low socioeconomic status, is consistent with previous reports suggesting a strong bias toward higher social class.17 As well, the 4:1 male preponderance found in this study is comparable and consistent with previous studies.9-17

It is currently not possible to suggest a plausible explanation for the negative association between pyloromyotomy and the availability of surgeons in the area. Although a positive association would have suggested an artifactual increase possibly due to overdiagnosis, this was not found to be so. The negative relationship between the availability of surgeons and pyloromyotomy further supports the positive association of pyloromyotomy with rural living because it would be unlikely that pyloric stenosis would be overdiagnosed or treated in rural as opposed to urban areas.

Overall, the descriptive distributions of variations among DHCs in Table 1 correspond well with results from the regression analysis showing higher pyloromyotomy rates in areas that were more rural. Some of the variations could have been accounted for by other population parameters such as the sociodemographic variables that we included in the regression analysis.

The rural Quinte, Kingston, Rideau DHC had the highest pyloromyotomy rate, and it is unlikely that this highest observed rate is a result of random chance or an outlier based on the size of the child population and the
number of pyloromyotomies performed in this DHC. On the other hand, it is possible that because of the small population and low number of pyloromyotomies performed in the more rural DHC of Grey, Bruce, Huron, Perth (the DHC with the lowest pyloromyotomy rate), when compared with the total, its rate could be unstable or unreliable, meaning it could potentially make this DHC a statistical outlier. However, when both these DHCs were removed from the analysis (the lowest rate of 0.95 per 1000 in the Grey, Bruce, Huron, Perth DHC and the highest rate of 3.30 per 1000 in the Quinte, Kingston, Rideau DHC), the variations remained at almost a 3-fold level and the correlation between percent rurality and pyloromyotomy rates remained unchanged. It is also important to note that the distributions of pyloromyotomy rates by DHC and rurality over the 2 time periods (1993-1996 and 1997-2000) were consistent, which indicated confidence that the observed pyloromyotomy rates could not be attributable to a random chance observation. Furthermore, the pyloromyotomy rate for the Quinte, Kingston, Rideau DHC was statistically significantly different from the overall provincial rate; therefore, this observation is solid because the probability that the difference observed is due to error or random chance is less than 5% (P<.05).

The 3-fold geographical variation in rate of pyloromyotomy between rural and urban DHCs in our study is comparable with the 3-fold geographical variation between the southern and northern Sweden rates reported by Hedbäck et al. Although climate is the only obvious difference between north and south Sweden, the main difference between the DHCs with high rates and low rates in our study is rural vs urban living. This suggests that environmental influences related to urban or rural living may have a role in this disorder. Applegate and Druschel identified similar geographic differences in their study, with much higher rates of IHPS in less urban areas of New York State than in New York City.

Some of the rural/urban differences observed in our study could be related to other environmental factors previously found to be related to IHPS, such as breastfeeding and bottle-feeding practices. Data limitations did not allow for detailed evaluation of the role of these factors. However, data from the 1994 National Longitudinal Survey of Children and Youth Cycle suggested that the initiation of breastfeeding at birth in Ontario has increased over the years from 64% in the 1970s to over 80% in the early 1990s. Unfortunately, this data did not provide specific rates of breastfeeding in rural and urban areas.

Although the rural/urban differences in New York State were partly attributed to underreporting, other sociodemographic indicators such as ethnicity and race were suggested. Further research into the effect of changing ethnic distribution should also be considered. Previous studies have shown that the risk of IHPS varies with race; it is highest among white people. Although ethnic origin is not recorded in our data, it is known that the majority of immigrants to Ontario live in the more urban areas of Ontario such as metropolitan Toronto. We examined the relationship between the risk of pyloromyotomy and home language, and it was not found to be statistically significant. However, it is important to note that home language may not reflect the underlying ethnic distribution accurately. It is possible that the changing ethnic distribution may contribute to the rural/urban differences related to IHPS. Ethnic changes and cultural practices may have an impact on some of the environmental influences thought to be related to IHPS, such as breastfeeding practices and birth order. An evaluation of the association of race and ethnicity with IHPS would help rule out or clarify the impact of ethnicity on the rural/urban differences observed in our study.

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Disclaimer: This analysis was based on anonymized data on hospital discharges submitted to the Canadian Institute for Health Information (Ottawa, Ontario) from 1993 to 2000. Other data sources for sociodemographic variables included the 1996 Canada Census population data from Statistics Canada (Ottawa) and the Canadian National Physician Database available through the Institute for Clinical Evaluative Science (Toronto, Ontario). All computations were prepared by the Population Health Sciences Program, the Hospital for Sick Children Research Institute (Toronto), and the Institute for Clinical Evaluative Sciences. The responsibility for the use and interpretation of this data is entirely that of the authors. The opinions expressed do not represent the views of the Canadian Institute for Health Information, the Institute for Clinical Evaluative Sciences, or Statistics Canada. Acknowledgment: We are grateful to Minh Duong Hua for providing programming and statistical support.

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