Problems in Using Birth Certificate Files in the Capture-Recapture Model to Estimate the Completeness of Case Ascertainment in a Population-Based Birth Defects Registry in New York State

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BACKGROUND: The limitations and underlying assumptions of the capture-recapture methods have hindered their application in epidemiological settings, especially in evaluating the completeness of birth defects registries. This study explored the possibility of using birth certificates as the secondary data source in a simple two-source capture-recapture model to estimate the completeness of case ascertainment of the Congenital Malformations Registry (CMR) for selected major birth defects. METHODS: The CMR and the birth certificates were used as the primary and secondary sources, respectively. Children who were born in 1996–2001 and had selected major birth defects were identified from the two sources. The accuracy of the diagnoses was examined by comparing the individual birth defect categories of the children from the two sources. RESULTS: Discrepancies in birth defect categories in the two data sources and false positives in the birth certificates were the major problems encountered in estimating the completeness of the CMR using the simple two-source capture-recapture method. The estimated completeness for selected major birth defects was only about 71%. Stratified analyses resulted in relatively high estimated completeness for oral clefts (90%) and Down syndrome (88%). CONCLUSIONS: Although the birth certificate data was not a good source for estimating the completeness of case ascertainment of the CMR using capture-recapture methods, the analyses provided reasonable estimates for some conditions that were relatively easy to identify and diagnose at birth, such as oral clefts and Down syndrome.


Key words: birth defects; congenital malformations registry; capture-recapture; completeness; hospital discharge data; birth certificates

INTRODUCTION

Evaluating the completeness of registration of birth defect cases has been an especially important concern and a priority activity for birth defects registries. The completeness of a registry, that is, the ability to identify and register all new cases diagnosed within a population, is essential to produce accurate statistics and conduct valid studies on birth defects in a population. In the past decades, a number of studies were conducted to assess the completeness of birth defects registries’ data (Boyed et al., 2005; Czeizel, 1997; Honein and Paulozzi, 1999; Larsen et al., 2003; Wang et al., 2001; Wen et al., 2000; Berghold et al., 2001; Cronk et al., 2003; Knox et al., 1984). Interestingly, only a few of these studies used the capture-recapture methods (Honein and Paulozzi, 1999; Berghold et al., 2001).

Capture-recapture methods, originally developed to estimate the size of a closed animal population (Cormack, 1968), have been used increasingly in epidemiological studies to assess the completeness of cancer registries (McClish and Penberthy, 2004; Silcocks and Robinson, 2004;...
were available. Sources that collect birth defect information independently diagnose at birth. The birth certificate files were used as the estimate the completeness of case ascertainment for selected the possibility of using birth certificates as the secondary the population. The objective of this study was to explore an unbiased estimate of the number of birth defects in important and necessary to explore, develop, and validate all CMR reports (Wang et al., 2005).

ing from hospital discharge audits comprised about 21% of defects in New York State; new reports result-

CMR, included major congenital malformations in the was constructed for identifying cases in this study. This birth certificate files. Moreover, some of the birth defect codes indicated on the birth certificates were not specific enough for classification. Thus, a list of selected major birth defects, which were in both sources and were relatively easy to identify and correctly diagnose at birth, was constructed for identifying cases in this study. This list, which accounted for about 13% of all cases in the CMR, included major congenital malformations in the central nervous, digestive, and musculoskeletal systems, oral clefts, and chromosomal anomalies. The selected major birth defects were then grouped into 11 categories. Children (not the defects) with one or more of these defects were counted because not all major malformations of a newborn were available in the birth certificate files.

Birth defects such as congenital anomalies of the cardio-

MATERIALS AND METHODS

Data Sources

CMR database. Hospitals and physicians are required to report to the CMR all children 2 years of age or younger who were born or reside in New York State and were diagnosed with major birth defects. Annually, the CMR receives birth defect reports for more than 10,000 children of New York State residents, which comprise about 4% of live births. CMR case ascertainment consists of: (1) mandatory reporting from hospitals and physicians; and (2) supplementary hospital audits by the CMR staff using SPARCS hospital discharge files (Wang et al., 2005).

Birth certificate files. The birth certificate files are maintained in the Vital Records Bureau of the NYSDOH, which annually records more than 255,000 live births in the State of New York. If a baby is diagnosed with birth defects at the time of birth, the birth certificate should indicate these malformations. One or more birth defects from a list of 27 conditions could be recorded on a newborn infant’s birth certificate (New York State Department of Health, 2001).

Birth Defects Selected for the Study

Not all major birth defect categories were recorded in the birth certificate files. Moreover, some of the birth defect codes indicated on the birth certificates were not specific enough for classification. Thus, a list of selected major birth defects, which were in both sources and were relatively easy to identify and correctly diagnose at birth, was constructed for identifying cases in this study. This list, which accounted for about 13% of all cases in the CMR, included major congenital malformations in the central nervous, digestive, and musculoskeletal systems, oral clefts, and chromosomal anomalies. The selected major birth defects were then grouped into 11 categories. Children (not the defects) with one or more of these defects were counted because not all major malformations of a newborn were available in the birth certificate files.

Data Matching

Matching cases in the CMR to the birth certificate files has been a routine procedure to obtain various birth variables including parents’ demographic information, potential risk factors, and birth certificate number. The identifying variables such as the hospital’s Permanent Facility Identifier (PFI), both infant’s and mother’s name, date of birth, medical record number, and mother’s social security number and residential information are used as matching variables. Extensive matching with multiple matching variables results in more than 95% of all CMR cases and 99.5% of CMR cases of New York State residents matched to the birth records.
For this study, a dataset containing information about children who were born in 1996–2001 to New York State residents and had selected major birth defects noted on their birth certificates was abstracted from the birth certificate files. This dataset, used as the secondary source in the capture-recapture analysis, was linked to the primary source, the CMR records of children who had the same selected major birth defects and were born to New York State residents, by birth year and birth certificate number. The matched cases from the linkage were identified as the cases captured by both sources. The accuracy of the diag-

Table 1

Simple Two-Source Capture-Recapture Model for Estimating the Total Number of Birth Defects Cases

<table>
<thead>
<tr>
<th>Cases ascertained by secondary source</th>
<th>Yes</th>
<th>No</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>Cases ascertained by primary source</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Yes</td>
<td>A</td>
<td>B</td>
<td>n₁ = A + B</td>
</tr>
<tr>
<td>No</td>
<td>C</td>
<td>D</td>
<td>n₂ = A + C</td>
</tr>
<tr>
<td>Total</td>
<td>n   = A + B + C + D</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

A: Cases captured by both sources.
B: Cases captured only by the primary source.
C: Cases captured only by the secondary source.
D: Cases missed by both sources, estimate based on the assumption that the probability of ascertainment from both sources is equal, that is, \( A \times D = B \times C \). Thus, \( D = (B \times C)/A = (n₁ - A)/A \).

\( n₁ \): The total number of cases captured by the primary source.
\( n₂ \): The total number of cases captured by the secondary source.
\( n \): The estimated total number of cases in a population, \( n = A + B + C + D = (n₁ \times n₂)/A \).

For this study, a dataset containing information about children who were born in 1996–2001 to New York State residents and had selected major birth defects noted on their birth certificates was abstracted from the birth certificate files. This dataset, used as the secondary source in the capture-recapture analysis, was linked to the primary source, the CMR records of children who had the same selected major birth defects and were born to New York State residents, by birth year and birth certificate number. The matched cases from the linkage were identified as the cases captured by both sources. The accuracy of the diag-

Table 2

Results from the Simple Two-Source Capture–Recapture Analysis Using Birth Certificate Files as the Secondary Source

<table>
<thead>
<tr>
<th>Cases ascertained by primary source: CMR</th>
<th>Yes</th>
<th>No</th>
</tr>
</thead>
<tbody>
<tr>
<td>Yes</td>
<td>2,824</td>
<td>4,855</td>
</tr>
<tr>
<td>No</td>
<td>1,178</td>
<td>2,025</td>
</tr>
<tr>
<td>Total</td>
<td>4,002</td>
<td>10,882</td>
</tr>
</tbody>
</table>

Results are from birth years 1996–2001.
*Estimated completeness of the data source.

Table 3

Comparison of the Birth Defects of the Children Captured by Both Source

<table>
<thead>
<tr>
<th>Agreement of the birth defects</th>
<th>Completely agree*</th>
<th>Partially agree†</th>
<th>Completely disagree‡</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Total n</td>
<td>%</td>
<td>n</td>
</tr>
<tr>
<td>Birth year</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>1996</td>
<td>487</td>
<td>411</td>
<td>84.4</td>
</tr>
<tr>
<td>1997</td>
<td>489</td>
<td>412</td>
<td>84.3</td>
</tr>
<tr>
<td>1998</td>
<td>488</td>
<td>417</td>
<td>85.5</td>
</tr>
<tr>
<td>1999</td>
<td>475</td>
<td>400</td>
<td>84.2</td>
</tr>
<tr>
<td>2000</td>
<td>434</td>
<td>359</td>
<td>82.7</td>
</tr>
<tr>
<td>2001</td>
<td>451</td>
<td>408</td>
<td>90.5</td>
</tr>
<tr>
<td>Hospital location</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Upstate New York(^ sensual )</td>
<td>2,054</td>
<td>1,767</td>
<td>86.0</td>
</tr>
<tr>
<td>New York City</td>
<td>770</td>
<td>640</td>
<td>83.1</td>
</tr>
<tr>
<td>Childern with birth defects in CMR</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Multiple</td>
<td>204</td>
<td>56</td>
<td>27.5</td>
</tr>
<tr>
<td>Single</td>
<td>2,620</td>
<td>2,351</td>
<td>89.7</td>
</tr>
<tr>
<td>Malformation categories in CMR</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Neural tube defects</td>
<td>227</td>
<td>132</td>
<td>58.1</td>
</tr>
<tr>
<td>Encephalus</td>
<td>37</td>
<td>18</td>
<td>48.6</td>
</tr>
<tr>
<td>Hydrocephalus</td>
<td>242</td>
<td>192</td>
<td>79.3</td>
</tr>
<tr>
<td>Oral clefts</td>
<td>968</td>
<td>908</td>
<td>93.8</td>
</tr>
<tr>
<td>Rectal atresia/stenosis</td>
<td>131</td>
<td>97</td>
<td>74.0</td>
</tr>
<tr>
<td>Tracheoesophageal fistula/oesophageal atresia</td>
<td>84</td>
<td>71</td>
<td>84.5</td>
</tr>
<tr>
<td>Limb reduction</td>
<td>52</td>
<td>47</td>
<td>90.4</td>
</tr>
<tr>
<td>Diaphragmatic hernia</td>
<td>104</td>
<td>94</td>
<td>90.4</td>
</tr>
<tr>
<td>Omphalocele/gastrochisis</td>
<td>213</td>
<td>209</td>
<td>98.1</td>
</tr>
<tr>
<td>Down syndrome</td>
<td>596</td>
<td>504</td>
<td>84.6</td>
</tr>
<tr>
<td>Other chromosomal anomalies</td>
<td>170</td>
<td>135</td>
<td>79.4</td>
</tr>
<tr>
<td>Total</td>
<td>2,824</td>
<td>2,407</td>
<td>85.2</td>
</tr>
</tbody>
</table>

Sources were CMR and birth certificates, birth years: 1996–2001.
*All defects agree.
†One or more but not all defects agree.
‡None of the defects agree.
\(^ sensual \) New York State excluding New York City.
\(^ sensual \) These 120 children had a single birth defect in the CMR but were identified as having multiple birth defects in the birth certificate files.

WANG ET AL.

Two-Source Capture-Recapture Analysis

A simple two-source capture-recapture model was used for estimating the total number of cases (Brenner, 1994). The number of cases captured by both sources was determined by linking the two data sources. The number of cases missed by both sources was estimated based on the assumption that the probability of ascertainment from both sources was equal. The estimated completeness of the case ascertainment for the primary source can be calculated by dividing the number of cases captured by the total number of cases estimated from the model. The 95% CI of the estimated completeness was calculated using the normal distribution-based CI of the estimated total number of cases that occurred during the study period (Pollack et al., 1990).

RESULTS

Crude Analyses Using the Two-Source Capture-Recapture Model

Table 1 illustrates the simple two-source capture-recapture model for estimating the total number of selected major birth defects. The analysis was performed using the CMR and the birth certificate data as the primary and the secondary sources, respectively. The results are shown in Table 2. Among live births for the years of 1996 through 2001, 2,824 cases with selected major birth defects were captured by both sources. There were 1,178 cases that were captured by the birth certificate files but not by the CMR. According to the model, 2,025 cases were missed by both sources (the CMR and the birth certificate data). The analysis was performed using the CMR and the birth certificate data as the primary and the secondary sources, respectively. The results are shown in Table 2. Among live births for the years of 1996 through 2001, 2,824 cases with selected major birth defects were captured by both sources. There were 1,178 cases that were captured by the birth certificate files but not by the CMR. According to the model, 2,025 cases were missed by both sources.

Discrepancies of the Birth Defects in the Two Sources

To compare the birth defects of the children captured by both sources (the CMR and the birth certificates) and to examine factors that affect the discrepancies in birth defect categories defined in the two sources, analyses were performed by birth year, geographic location of the reporting hospitals, the number of birth defects (single or multiple), and birth defect category. The results are shown in Table 3. Out of 2,824 children captured by both sources, 85.2% had birth defect categories that completely agreed in both sources, 9.1% partially agreed (one or more but not all malformations agreed), and 5.7% completely disagreed. Children with a single birth defect had much higher per-
cent agreement (89.7%) in the birth defect category defined in the two sources compared to children with multiple defects (27.5%). Among the individual birth defect categories, defects that were visible, such as oral clefts, gastrochisis, and omphalocle combined were more likely to be diagnosed correctly on both sources. The overall agreement (completely and partially agreed) of the birth defect categories in the two sources did not change significantly over the study years, 1996–2001.

Stratified Analyses Using the Capture-Recapture Method

Stratified analyses were performed by the number of birth defects (single or multiple) and by birth defect categories for the children with a single birth defect, using the simple two-source capture-recapture model. The birth defect categories defined by the CMR were used for the children who were captured by both sources but had birth defect categories that disagreed in the two sources. The results are presented in Table 4. The estimated completeness of case ascertainment for children with multiple defects was significantly higher (88.7%, 95% CI 85.9–91.6%) than that for children with a single defect (69.5%, 95% CI 68.3–70.7%). Moreover, results from stratified analysis among children with a single birth defect by birth defect category showed that the estimates of completeness strongly depend on the specific birth defect. The highest estimates were 90.1 and 88.1% for oral clefts and Down syndrome, respectively, and the lowest estimate was 23.6% for encephalus. The estimate for omphalocle and gastrochisis was surprisingly low (52%), even though these defects are relatively easy to identify and correctly diagnose at birth.

DISCUSSION

Using birth certificate files as the comparison source in a simple two-source capture-recapture model, the estimated completeness of case ascertainment of the CMR for selected birth defects was about 71%. This relatively low estimate was most likely attributable to the false-positive reports in the birth certificate files (Olsen et al., 1996; Pollock et al., 1990). A previous study conducted by our CMR staff to determine whether birth certificates could be used to ascertain unreported cases to the CMR found that about 45% of the children with one or more birth defects noted on their birth certificates were normal, that is, there was no mention of a malformation in their medical records (Olsen et al., 1996).

In order to evaluate the effect of false positives on the results of the capture-recapture analysis, we matched the cases that were found in the birth certificate files but not in the CMR to SPARCS hospital discharge data to verify the cases. When we assumed that the unmatched cases were false positives and removed them from the analysis, the estimated completeness was approximately 82% (data not shown). Although the use of SPARCS data to filter the false positives sacrifices the independence of data sources because the SPARCS data were used as a supplementary data source to the CMR, the finding demonstrates that the quality of the comparison data source is critical for evaluating the completeness of case ascertainment using the capture-recapture analysis. In order to identify false positive cases from the birth certificate files and ascertain unreported cases, we would need to request individual medical records from reporting hospitals. This would add an extra burden to the reporting hospitals and the CMR staff. The CMR staff has been actively seeking more practical and less expensive measures to identify unreported cases from other sources.

The current study found that although false positives in the birth certificate files lead to an overestimate of the total number of cases and thus, the completeness of the case ascertainment for the birth defects of interest was underestimated, they did not significantly affect the estimates for some birth defects that were relatively easy to identify and diagnose at birth, such as oral clefts and Down syndrome. The estimated completeness for oral clefts and Down syndrome was 90 and 88%, respectively. Our estimate for Down syndrome was consistent with that reported by Berghold et al. (2001) for the Styrian Malformation registry. They estimated that Down syndrome cases reported to the registry between 1985–1992 were 88% complete, using the two-source capture-recapture method allowing for time-varying dependency (Berghold et al., 2001).

The discrepancy in the case definition of data sources has been shown to be one of the major problems encountered in estimating the completeness of case ascertainment using the capture-recapture methods (Hook and Regal, 1999). The results from our study show that among children captured by both sources, about 6% had birth defects that totally failed to agree. In this study, hospitals reported birth defect cases to the CMR using a standard reporting card to provide ICD-9-CM codes and narratives, as well as the information about the child and parents. On the other hand, there were only 27 one-digit fields (with the value of 1 or 0) used for recording major anomalies (New York State Department of Health, 2001) in the birth certificate files and there were no narratives available, resulting in discrepancies in birth defect categories in the two data sources. Because we do not confirm all of the diagnosed cases reported to the CMR due to limited resources, the CMR staff has initiated an on-site auditing program to focus on improving the accuracy of diagnoses by visiting hospitals that have an unreasonably low number of case reports and/or insufficient diagnostic information for the reported cases.

There was no strong evidence of dependence between the two sources in the current study. Our study selected only children with major birth defects that were relatively easy to identify and diagnose correctly at birth, because the CMR receives case reports of children up to 2 years of age whereas the birth certificate files collect birth defects information at birth. This exclusion greatly reduced (if not eliminated) the chance of positive source dependency, that is, cases identified by one source (the CMR) having a higher chance of being included in another source (the birth certificates). It should also be noted that our estimate of completeness of case ascertainment for selected birth defects might not be generalized to all the birth defects in the CMR, because only a portion of birth defects in the CMR, which comprised 13% of all CMR cases, was included in the study.

In conclusion, discrepancies in birth defect categories defined in the two sources (the CMR and the birth certificates) and false positives in the birth certificate files were the major problems encountered in estimating the completeness of case ascertainment using the simple two-source capture-recapture method. False positives in the birth certificate files lead to the overestimation of the total number of
cases and thus, the underestimation of CMR’s completeness of case ascertainment for selected birth defects. Although the birth certificate data were not a good source for estimating the completeness of case ascertainment of the CMR using the capture-recapture methods, our results from the capture-recapture analyses provided reasonable estimates for some birth defects that are relatively easy to identify and correctly diagnose at birth, such as oral clefts and Down syndrome. The exploratory analysis and the findings of the current study should be helpful to the registries and researchers in the birth defects research community; our study has shown the importance of the quality of the data sources and has suggested that the two-source capture-recapture model should be used with caution in estimating the completeness of case ascertainment of birth defects if the quality of the comparison data source, such as the birth certificates, is in question.

REFERENCES


