The Influence of Misclassification Bias on the Reported Rates of Congenital Anomalies on the Birth Certificates for West Virginia—A Consequence of an Open-ended Query

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BACKGROUND: Passive surveillance for congenital anomalies using birth certificates are generally considered to have biased reporting, though the sources of those biases are not well-known nor controlled for. We have analyzed the congenital anomaly reporting data for 418,385 live births in West Virginia (1990–2009) from the 1989 US standard birth certificate and have newly identified a particular source of bias. **METHODS:** Congenital anomaly prevalence rates per 100 live births have been determined for both specified birth defects and for other congenital anomalies by county, by hospital, and by year. Extreme outliers were identified by *z* score. Text strings for ''other congenital anomaly'' reports recorded for 1998–2009 were assessed for information on congenital anomalies. **RESULTS:** While rates for specified birth defects reported in checked-box format showed little variation, rates for ''other congenital anomaly'' collected in open-ended format showed much variation. Nearly half of the ''other congenital anomaly'' reports were for neonatal conditions rather than for major structural congenital anomalies. This misclassification alone had elevated the state-wide congenital anomaly reporting rate from 1.1 to 1.8% of live births. Geographic clustering and a temporal bulge in congenital anomaly reports disappeared after misclassified data were removed. **CONCLUSIONS:** Data collected in checked-box format on specified birth defects showed consistent patterns over time and space, while data collected in open-ended format on ''other congenital anomalies'' showed an epidemiological pattern reflecting neonatal conditions rather than birth defects. The 2003 US standard birth certificate wisely limits data collection to specified birth defects using the checked-box format. *Birth Defects Research (Part A) 97:140–151, 2013.* © 2013 Wiley Periodicals, Inc.

Key words: birth defects; congenital anomalies; birth certificates; misclassification; neonatal conditions

INTRODUCTION

Birth defects occur with an overall prevalence of about 3% for live births and are the leading cause of death in children <1 year old in the US (Anderson et al., 1997; Hoyert et al., 2006). Each year, about 130,000 babies are born with birth defects, which cost over $2.6 billion annually for their care and treatment (NCBDDD, 2011). These usually have functional or cosmetic significance which may need monitoring or intervention. Alternative terms for birth defects are congenital malformation or congenital anomaly, indicating that they are conditions at birth which vary from the standard presentation and are primarily structural in nature. Major birth defects are

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conditions present at birth that result from structural changes in one or more parts of the body and can have a serious, adverse effect on health, development, or functional ability (CDC, 2011). Other definitions may extend beyond structural abnormalities that may have a serious adverse effect to include inborn errors of metabolism. Birth defect surveillance and monitoring programs are established for epidemiological purposes—to describe the birth prevalence rates of birth defects over time and area and to identify determinants of those distributions (Cordero, 1992)—and for planning, educational, and health care services (Lydberg and Edmunds, 1992). Such programs also serve as case finding for newborns needing assessment for medical or surgical interventions. Active surveillance programs may entail a professional specifically examining the newborn records to assure more complete and accurate ascertainment. Passive surveillance programs are frequently based on birth certificate information or hospital discharge information which exists as part of the ordinary activities of hospitals. Passive programs are more universal but are more likely to suffer from qualitative and quantitative issues of variability and inconsistency.

The categories of congenital anomalies on birth certificates, while generally understood, lack specificity and definition which leads to both false positive and false negative reports. Surveillance systems may include data on both specific birth defects selected for surveillance and on other congenital anomalies not specifically selected. This was the structure of the 1989 version of the standard US birth certificate. The subsequent 2003 version, which has yet to be universally adopted, is restricted to only certain specified birth defects. The section for congenital anomalies on the standard US birth certificate enables states to ascertain, track and study birth defects among their newborns. While all states may use the same birth certificate, the methodologies for data acquisition may vary across the states and within the states. Nonetheless, birth certificate data are used to estimate birth defect prevalences, changes in temporal and geographic patterns, and to examine the potential associations between birth defects and occupational or environmental exposures (Lamm, 1979; Piper et al., 1993; Olsen et al., 1996; Watkins et al., 1996; Buehler et al., 2000; Honein et al., 2001; Wyszynski and Wu, 2002; Gilbreath and Kass, 2006; Luquetti and Koifman, 2010).

The purpose of this study was to examine the birth defect reporting on the West Virginia birth certificate seeking apparent temporal or geographic patterns that might raise etiological hypotheses. We have further sought to understand what are the particular birth defects that may underlie the temporal and geographic patterns.

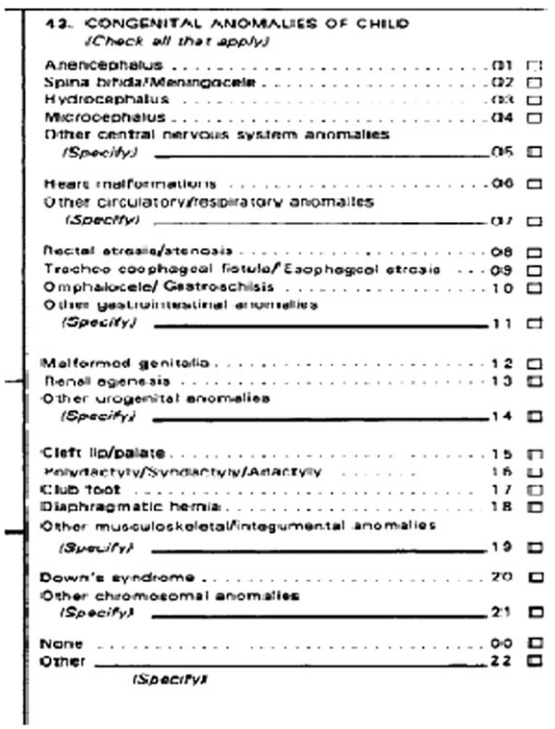
MATERIALS AND METHODS

We present the analysis of the congenital anomaly information from the birth certificates for residents of West Virginia (1990–2009) that was prepared for us as an electronic deidentified file in 2012 by the West Virginia Department of Health. The file included information on mothers, fathers, and infants for all live births to West Virginia residents, whether the birth occurred in West Virginia or outside of West Virginia. This study was focused on congenital anomalies reported electronically on birth certificates for this period.

The state of West Virginia is located in the central Appalachian mountains in the eastern United States. The 55 counties of West Virginia serve as the units of geographic aggregation in this analysis, and calendar years serve as the units of temporal aggregation.

The 1989 US birth certificate was used throughout the period 1990–2009. It has two parts. The first part asks demographic information about the parents and infant, e.g., name, date of birth, place of birth, residence, ethnicity, education, etc. The second part contains questions on maternal and infant health conditions, such as pregnancy history, prenatal care, pregnancy complications, obstetric procedures, and a section on congenital anomalies of child. Figure 1 shows the section for congenital anomaly information on the 1989 version of the birth certificate.

Section 43 is on page 2 of the birth certificate and contains a total of 22 items for information on congenital anomalies organized by six systems—(1) central nervous, (2) circulatory/respiratory, (3) gastrointestinal, (4) urogenital, (5) musculoskeletal, and (6) chromosomal—and with the category of ''Other'' (Fig. 1). For each system, certain specified birth defects (congenital anomalies) are given to be checked and an open-ended space is provided requesting the certifier to describe the details of ''other congenital anomalies'' in that system. There were a total of 15 specified birth defects (congenital anomalies), six spaces for ''other congenital anomalies'' for the six systems, and a seventh space for ''Other'' congenital anomalies (i.e., those that did not fit into the above categories), for a total of 23 entries. The entries are not


43. CONGENITAL ANOMALIES OF CHILD
(Check all that apply)

Anencephalus ..... 01 ☐
Spina bifida/Meningocele ..... 02 ☐
Hydrocephalus ..... 03 ☐
Microcephalus ..... 04 ☐
Other central nervous system anomalies
(Specify) _____ 05 ☐

Heart malformations ..... 06 ☐
Other circulatory/respiratory anomalies
(Specify) _____ 07 ☐

Rectal atresia/stenosis ..... 08 ☐
Tracheo esophageal fistula/ Esophageal atresia ... 09 ☐
Omphalocele/ Gastroschisis ..... 10 ☐
Other gastrointestinal anomalies
(Specify) _____ 11 ☐

Malformed genitalia ..... 12 ☐
Renal agenesis ..... 13 ☐
Other urogenital anomalies
(Specify) _____ 14 ☐

Cleft lip/palate ..... 15 ☐
Polydactyly/Syndactyly/Adactyly ..... 16 ☐
Club foot ..... 17 ☐
Diaphragmatic hernia ..... 18 ☐
Other musculoskeletal/integumental anomalies
(Specify) _____ 19 ☐

Down's syndrome ..... 20 ☐
Other chromosomal anomalies
(Specify) _____ 21 ☐

None ..... 00 ☐
Other _____ 22 ☐
(Specify)

Figure 1. The section for congenital anomaly information on birth certificate West Virginia (the second page).

ICD-coded. The 1989 revision was the first to include a checked-box format for reporting birth defects (Tolson et al., 1991). Previously, the birth certificate only used the open-ended question format which required only the reporting of the presence or absence of a congenital anomaly and allowed for a description.

During 1989–1997, West Virginia birth certificates were filled in at the hospitals and were transmitted to the state's bureau of vital statistics within a few days of delivery for electronic entry into the computer system and microfiche storage. Data entry was a Yes or No for each specified birth defect and a Yes or No for each category of ''other congenital anomaly.'' The literal strings for the ''other'' congenital anomalies were not added. The electronic files were then transmitted to the National Center for Health Statistics (NCHS), a part of the Centers for Disease Control and Prevention (CDC), for integration into the national dataset (Starr and Starr, 1995).

In 1998, the health department expanded its system to a fully electronic birth registry program in which birth certificate information could be electronically entered locally by hospitals rather than centrally at the health department. This program, with the capacity for capturing the text strings, was introduced among the various West Virginia hospitals over a number of years.

Review of the text strings showed that they fit into a number of common groups across the systems based on the quality of information they contained. (A) Malformation—these text strings gave sufficient information to classify them as congenital anomalies, either specified ones or other generally recognized malformations that did not fit into the categories of the specified birth defects. Most of the malformations were entered under the correct system. (B) Unknown—either there was no text string or the text string said unknown, doctor refused, or other similarly none informative statement. (C) Neonatal condition—the text string was sufficiently informative to show that the report was of a neonatal condition or observation rather than of a congenital anomaly, or that it referred to a common variant, a transient condition related to childbirth maturation, or an administrative report. The recorded neonatal conditions included both neonatal symptoms such as arrhythmias, murmurs, gastrointestinal reflux, hip click and neonatal diseases such as sepsis of the newborn, respiratory distress, necrotizing enterocolitis, and system-specific diseases. Common variants included birth marks, hemangiomas, Mongolian spots, skin tags, and tongue tie. Transitional observations included patent ductus arteriosus (PDA), secundum atrial septal defect (s-ASD) or patent foramen ovale (PFO), hydrocele, and undescended testicle. Administrative reports, such as transfer to the neonatal intensive care unit (NICU) or another hospital, prematurity, death, and the ordering of laboratory studies, were considered as neonatal information. Together, these conditions were grouped in the category of neonatal condition. The coding of text strings was done blindly with respect to both the county of residence and the facility where the delivery had occurred. Coding was conducted by two of the authors (SHL and SAR) with resolution of discrepancies.

Statistical Analysis

The 15 congenital anomalies specified on the birth certificate form were called ''specified birth defects'' and the 7 other indications on the birth certificate for other congenital anomalies were analyzed as ''other congenital anomalies.'' Birth certificates with specified birth defects and/or with other congenital anomalies were analyzed as reporting ''any congenital anomaly.'' Analyses were conducted for each system and for all systems combined.

Congenital anomaly birth prevalence rates were calculated for live births as the number of newborns with the congenital anomaly report per 100 live births. Rates were calculated (1) for each specified birth defect, singly and collectively; (2) for each ''other congenital anomaly,'' singly or collectively; and (3) for ''any'' congenital anomaly, singly or collectively, i.e., with a specified birth defect and/or an ''other congenital anomaly.''

First, overall rates for specified birth defects, other congenital anomalies, and any congenital anomaly were calculated for the 1990–2009 period for each county ($n = 55$) and for each facility of birth (i.e., place of occurrence) ($n = 44$) [Not shown]. Facility-specific rates were calculated for the 44 facilities that had more than 1,000 live births to West Virginia residents during the 20-year period of 1990–2009. These 44 facilities accounted for 98% of the live births and 97% of the congenital anomaly mentions. Z scores (z: Mean/ Std Dev = $\bar{x}$/SD) were calculated to identify extreme outliers ($z \geq 4$; or $z \leq -4$) for both county rates and facility rates.

Second, distributions of county rates were mapped for geographic analyses and shown graphically. Annual rates (specified, other, and any) for the state were shown graphically for temporal analyses.

Third, where analysis of facility rates identified a hospital as an extreme outlier, its rates for individual malformation categories or groups were compared with the rates for the rest of the births to West Virginia residents and with those of its catchment area using prevalence rate ratios (PRR). The catchment area was defined as the set of counties each of which had at least 20% of its births for its new residents occurring at that hospital. PRRs were calculated as the prevalence rate for births at that hospital (congenital anomalies/100 live births) divided by the prevalence rate for the births elsewhere. The number of excess reports of malformations were calculated as the difference between the observed and the predicted where the predicted was determined by applying the rates for nonoutlier hospitals to the population of the outlier hospital. The temporal analysis of the rates was also examined with the exclusion of the data from the extreme outlier facilities.

Tabulations were developed using STATA (SE11). Calculations were made using Microsoft Excel. Statistical significance for the parameter coefficients was set as a two-tailed condition with $P < 0.05$. Temporal patterns were analyzed using Joinpoint regression analysis [http://surveillance.cancer.gov/joinpoint/example.html].

RESULTS

The Rates of Congenital Anomalies in West Virginia

The 20-year (1990–2009) birth registry dataset for West Virginia contained records for 418,385 live births to West Virginia residents, 91% of which occurred in West Virginia and 9% of which occurred in immediately neighboring states (Virginia > Maryland > Ohio). Rates for all specified birth defects, other congenital anomalies, and

any congenital anomalies were calculated for the state and for each county (Table 1). Specified birth defects were reported on 2,381 (0.57%) of the birth certificates and ''other congenital anomalies'' were indicated on 5,659 (1.35%) of the birth certificates. In total, congenital anomalies were reported on 7,597 (1.82%) of the birth certificates. Thus, the proportion of newborns recorded with congenital anomalies is elevated three-fold (1.82%/0.57 % = 3.19) if ''other congenital anomalies'' are included as well as specified birth defects.

Geographical Distribution of County-level Rates of Congenital Anomalies in West Virginia

The geographical distribution of the congenital anomaly rates for the 55 West Virginia counties has been mapped, both for the specified birth defects (Fig. 2a) and for the other congenital anomalies (Fig. 2b). The geographic distribution of the rates for any congenital anomalies (not shown) was similar to that for other congenital anomalies.

The county-specific rates for the specified birth defects, as a group, showed a band of higher rates across the middle of the state with lower levels on the west side of the Ridge and Valley mountains and higher rates on the east side. The rates were normally distributed across the 55 counties (Fig. 3a) with a median rate of 0.58%. There were no extreme outliers with z score of ≥ 4.

The county-specific rates for other congenital anomalies (Fig. 3b) showed a distribution (median 1.08%) with a long tail to the right. Raleigh County had the only extreme outlier z scores (≥ 4), both for any congenital anomaly and for other congenital anomalies (5.78%).

Temporal Trends of the Rates of Congenital Anomalies

The 20-year temporal trends for the West Virginia birth defect and other congenital anomaly rates are presented in Figure 4. The rates for specified birth defects generally approximate a prevalence of 0.70% during 1990–1997 and of 0.50% during 1998–2009. The rates for other congenital anomalies and for any congenital anomaly were reasonably steady during 1992–1997 and during 1997–1999 as the system changes were introduced. Those rates nearly doubled from 1999 to 2003 and then showed some ebb and flow. Year 1990 had increased rates of congenital anomaly reports, which probably reflect the setting up of standard practices in registering congenital anomalies during the first full year of use of the new birth certificate. The temporal pattern for any mention of congenital anomaly is clearly driven by the ''other congenital anomalies'' rather than the specified birth defects.

Analysis of Congenital Anomaly by Facility

We analyzed the reported congenital anomaly rates by facility. The median facility-specific rates were 0.24% for specified birth defects, 0.81% for other congenital anomalies, and 1.62% for any congenital anomalies. One hospital, Hospital 22 in Raleigh County, stood out with a z score of ≥ 4 for other congenital anomalies and for any congenital anomaly. With the exclusion of the extreme outlier Hospital 22 from the data, the visual bulges of the ''other congenital anomaly'' rate and of the ''any congenital anomaly'' rate between 1999 and 2003 were no longer evident (Fig. 5).

Such an observation raises the issue of—in what ways did the congenital anomaly reporting for Hospital 22 distinguish itself from that of the rest of the state? We have analyzed the 1998–2009 West Virginia birth certificate dataset that included electronically the text string for the entry of other congenital anomalies, comparing the case counts and rates for Hospital 22 with those for the rest of West Virginia State (Table 2). Of the 250,159 live births in 1998–2009 recorded for West Virginia residents, 19,626 (7.8%) were recorded from Hospital 22. Hospital 22 reported an ''other congenital anomaly'' more frequently than reported a specified birth defect compared as the rest of the state (1,518/155 = 9.8 vs. 2,186/1,305 = 1.7) (Table 2). While Hospital 22 reported 7.8% of the live births for West Virginia residents, it reported 11% of the specified birth defects and 41% of the ''other congenital anomalies.'' Overall, it accounted for 33% of all the infants reported to have congenital anomalies (Table 2).

The prevalence rate ratio (PRR) for specified birth defects (Hospital 22 births compared to non-Hospital 22 births) was 1.40, but for ''other congenital anomalies'' the PRR was 8.16 overall and 10–25 fold for most systems (circulatory, gastrointestinal, urogenital, and musculoskeletal). Thus, the reported rate for having a congenital anomaly for Hospital 22 births were about six times as great as that for the rest of the state, primarily due to a reported ''other congenital anomaly'' rate that was about eight times as great as that of the rest of the state.

Comparison of the Rates of Congenital Anomalies between Hospital 22 and Surrounding Hospitals

To investigate whether the observed high rates were due to an area effect or to a hospital effect, we examined the congenital anomaly reports for Hospital 22 catchment area. The Hospital catchment area was defined as the set of counties for which more than 20% of their live births had occurred in Hospital 22. Based on this criterion, the catchment area was comprised o Raleigh, Fayette, Wyoming, Summers, and Nicholas counties. Demographically, the Hospital 22 and non-Hospital 22 births were similar with respect to distribution of maternal age, education, Hispanicity, marriage, and paternity. Proportion black and proportion on Medicaid were higher for Hospital 22 births than for non-Hospital 22 births.

For residents in the Hospital 22 catchment area, the reporting rate for specified birth defects were similar for Hospital 22-born infants and for non-Hospital 22-born infants (0.80% vs. 0.67%; $P > 0.05$) with a prevalence rate ratio of 1.19 (Table 3). However, the reporting rate for other congenital anomalies was considerably different (7.76% vs. 1.21%) with a prevalence rate ratio of 6.41 (Table 3). The rates for individual types of ''other congenital anomaly'' were also compared. The rate of reported cases for individual types of ''other congenital anomaly'' was significantly greater for catchment area births among Hospital 22 births than among non-Hospital 22 births for all conditions with the exceptions of congenital hip dislocation and of urogenital (based on single cases). The rates for individual types of other congenital anomalies for the non-Hospital 22 births for the catchment area were generally similar to those of the non-Hospital 22 births to residents elsewhere in West Virginia

Table 1
Congenital Anomaly (Specified, Other, and Any) Counts and Rates for West Virginia County 1990 to 2009

		Counts				Rates		
Number	County	Live births	Specified birth defects[a]	Other congenital anomaly[b]	Any congenital anomaly[c]	Specified birth defects[a] (%)	Other congenital anomaly[b] (%)	Any congenital anomaly[c] (%)
1	Barbour	3,478	26	42	61	0.75	1.21	1.75
2	Berkeley	20,931	120	276	375	0.57	1.32	1.79
3	Boone	6,486	25	41	61	0.39	0.63	0.94
4	Braxton	3,083	30	30	52	0.97	0.97	1.69
5	Brooke	4,858	21	53	68	0.43	1.09	1.40
6	Cabell	23,655	115	125	223	0.49	0.53	0.94
7	Calhoun	1,590	15	19	32	0.94	1.19	2.01
8	Clay	2,648	9	14	21	0.34	0.53	0.79
9	Doddridge	1,521	6	18	22	0.39	1.18	1.45
10	Fayette	11,507	89	571	641	0.77	4.96	5.57
11	Gilmer	1,382	6	18	23	0.43	1.30	1.66
12	Grant	2,574	13	34	45	0.51	1.32	1.75
13	Greenbrier	7,641	49	260	300	0.64	3.40	3.93
14	Hampshire	4,247	20	58	73	0.47	1.37	1.72
15	Hancock	6,680	37	50	81	0.55	0.75	1.21
16	Hardy	2,755	16	27	41	0.58	0.98	1.49
17	Harrison	16,732	111	179	275	0.66	1.07	1.64
18	Jackson	6,358	25	38	58	0.39	0.60	0.91
19	Jefferson	10,866	58	173	217	0.53	1.59	2.00
20	Kanawha	48,165	126	154	262	0.26	0.32	0.54
21	Lewis	3,953	25	37	60	0.63	0.94	1.52
22	Lincoln	5,612	31	28	51	0.55	0.50	0.91
23	Logan	9,322	35	47	80	0.38	0.50	0.86
24	Marion	12,638	98	130	210	0.78	1.03	1.66
25	Marshall	7,371	40	57	89	0.54	0.77	1.21
26	Mason	5,904	34	44	75	0.58	0.75	1.27
27	McDowell	6,768	46	63	102	0.68	0.93	1.51
28	Mercer	15,384	82	137	204	0.53	0.89	1.33
29	Mineral	5,723	43	100	137	0.75	1.75	2.39
30	Mingo	8,108	43	82	120	0.53	1.01	1.48
31	Monongalia	17,702	102	232	311	0.58	1.31	1.76
32	Monroe	2,648	10	47	54	0.38	1.77	2.04
33	Morgan	2,821	21	37	53	0.74	1.31	1.88
34	Nicholas	5,912	39	136	173	0.66	2.30	2.93
35	Ohio	10,445	62	95	142	0.59	0.91	1.36
36	Pendleton	1,588	12	24	34	0.76	1.51	2.14
37	Pleasants	1,568	13	18	28	0.83	1.15	1.79
38	Pocahontas	1,878	14	34	47	0.75	1.81	2.50
39	Preston	6,558	48	110	148	0.73	1.68	2.26
40	Putnam	11,905	45	41	77	0.38	0.34	0.65
41	Raleigh	17,861	174	1,032	1,160	0.97	5.78	6.49
42	Randolph	6,486	48	78	114	0.74	1.20	1.76
43	Ritchie	2,171	17	16	30	0.78	0.74	1.38
44	Roane	3,303	10	35	44	0.30	1.06	1.33
45	Summers	2,398	14	99	110	0.58	4.13	4.59
46	Taylor	3,266	33	33	59	1.01	1.01	1.81
47	Tucker	1,437	14	17	31	0.97	1.18	2.16
48	Tyler	1,926	10	19	26	0.52	0.99	1.35
49	Upshur	5,435	33	55	82	0.61	1.01	1.51
50	Wayne	9,697	54	46	92	0.56	0.47	0.95
51	Webster	2,162	22	24	40	1.02	1.11	1.85
52	Wetzel	4,155	40	45	78	0.96	1.08	1.88
53	Wirt	1,146	2	18	20	0.17	1.57	1.75
54	Wood	20,227	100	207	295	0.49	1.02	1.46
55	Wyoming	5,750	50	256	290	0.87	4.45	5.04
	Total	418,385	2.381	5,659	7,597	0.57	1.35	1.82

[a]Number of live births with one or more specified birth defect (congenital anomaly) on birth certificate.
[b]Number of live births with one or more ''other congenital anomaly'' on birth certificate.
[c]Number of live births with one or more specified birth defect and or other congenital anomaly on birth certificate.

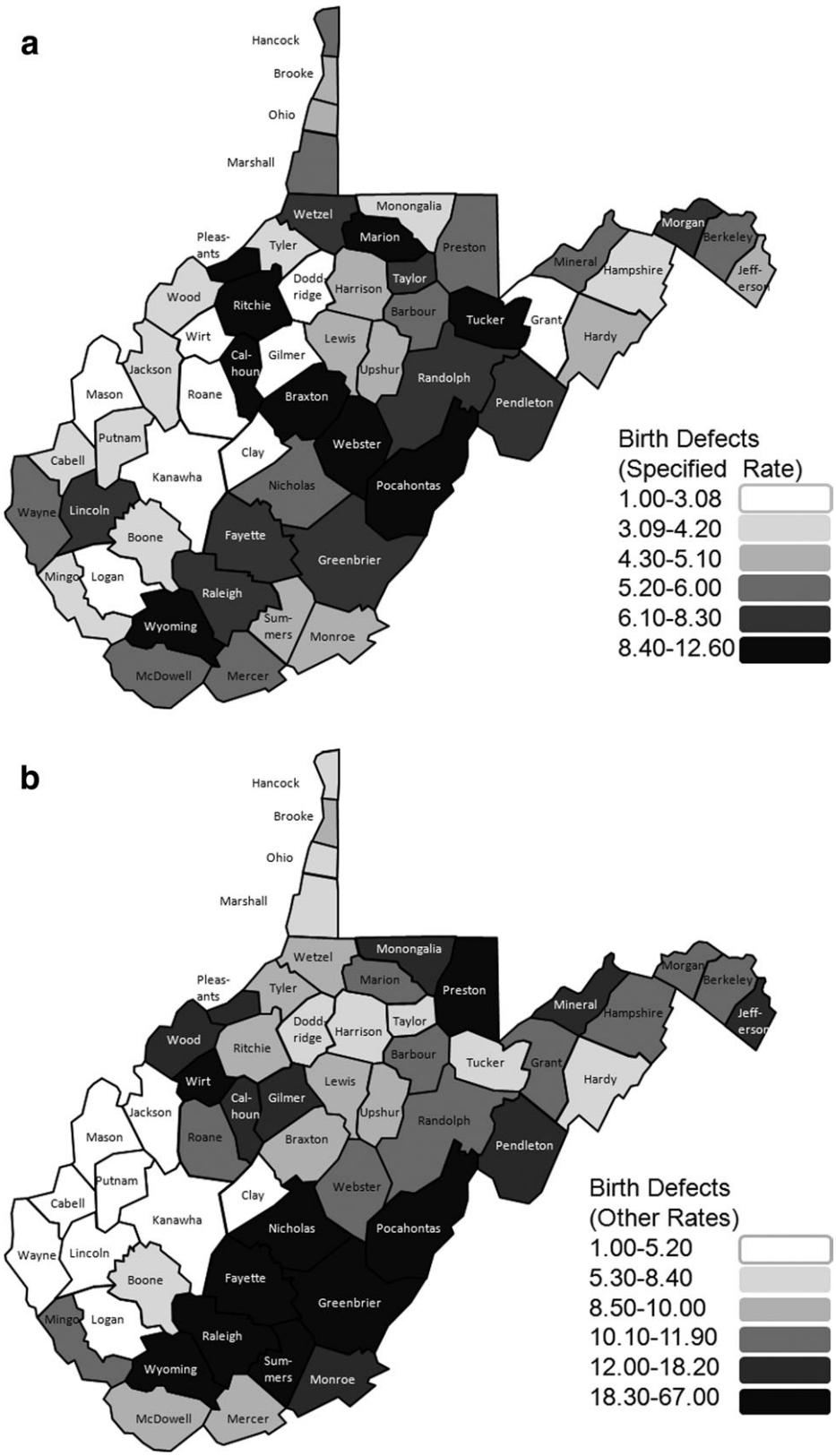


Figure 2. (**a**) Congenital anomaly birth prevalence rates for specified birth defects as reported on birth certificates. (**b**) Congenital anomaly birth prevalence rates for ''other congenital anomaly'' as reported on birth certificates.

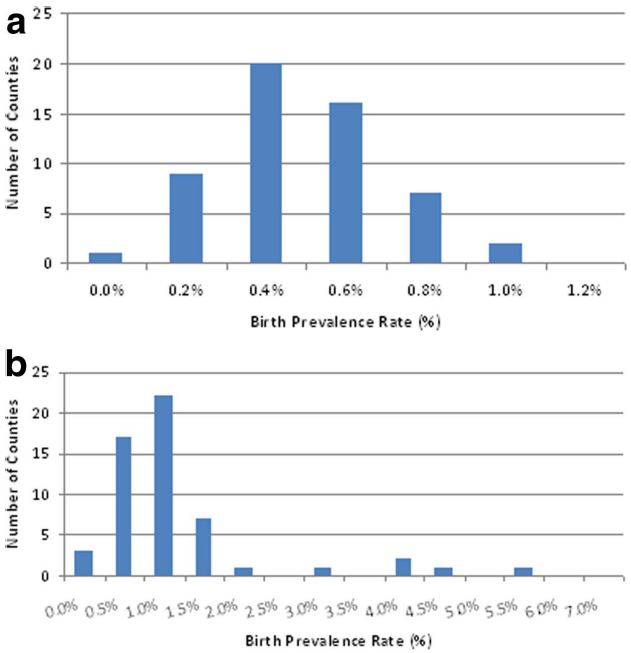


Figure 3. (**a**) Distribution of reported rates per 100 live births for specified birth defects (West Virginia Counties, 1990–2009). (**b**) Distribution of reported rates per 100 live births for ''other congenital anomaly'' (West Virginia Counties, 1990–2009). [Color figure can be viewed in the online issue, which is available at wileyonlinelibrary.com.]

Thus, the rates reported by Hospital 22 did not represent a characteristic of the area but rather a reporting characteristic of Hospital 22.

Misclassification Bias of Congenital Anomaly Reports for Hospital 22

Each report of ''other congenital anomaly'' has been classified as either (A) Malformation, (B) Unknown, or (C)

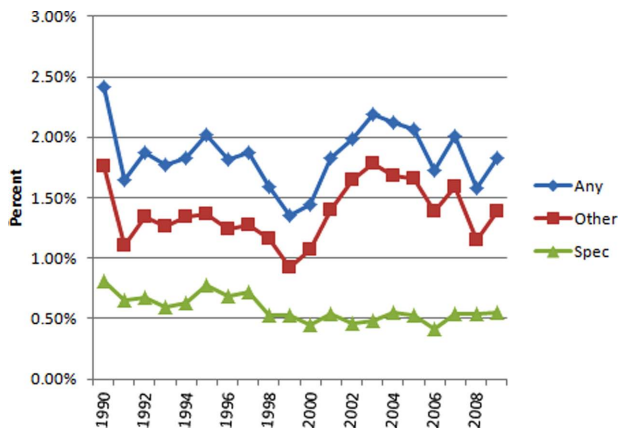


Figure 4. Rates of specified, other, any congenital anomaly for West Virginia (1990–2009). [Color figure can be viewed in the online issue, which is available at wileyonlinelibrary.com.]

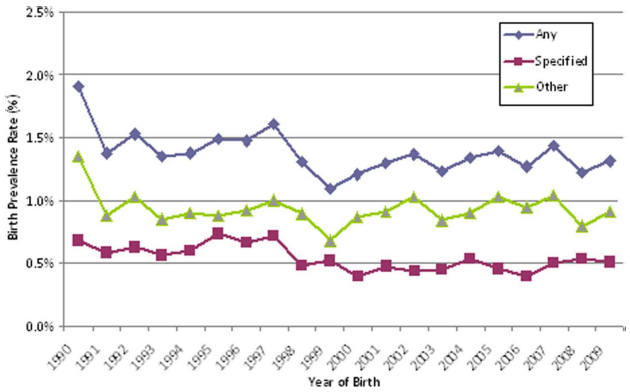


Figure 5. Congenital anomaly rates (specified, other, and any) for West Virginia by year with the exclusion of data for hospital 22. [Color figure can be viewed in the online issue, which is available at wileyonlinelibrary.com.]

Neonatal condition. Table 4 shows the distribution of ''other congenital anomalies'' reports across these three classes for the state and separately for Hospital 22 births and for non-Hospital 22 births for the 1998–2009 period. Hospital 22, which accounted for 7.8% of the live births, accounted for 41% of the ''other congenital anomaly'' reports with 29% of the malformations, 15% of the unknown, and 59% of the neonatal conditions. While neonatal conditions accounted for 72% of ''other congenital anomaly'' reports for the Hospital 22 (1,860/1,515 = 72%), they still accounted for 35% for the non-Hospital 22 births (764/2,187 = 35%). Hospital 22 was not unique in reporting neonatal conditions as congenital anomalies, but it was the major contributor. The reporting rates per 100 live births for the three classes were similar for the non-Hospital 22 births at 0.3% but dissimilar for the Hospital 22 births (0.6–5.6%). The prevalence rate ratios (PRRs) were 4.7 for (A) malformations, 2.1 for (B) unknown, and 16.9 for (C) neonatal conditions. Thus, while all three PRRs were elevated, the major difference between Hospital 22 and non-Hospital 22 was the markedly elevated reporting of neonatal conditions as other congenital anomalies.

The measure of excess reporting from Hospital 22 can be calculated. If the reporting rates for Hospital 22 had been the same as those for the non-Hospital 22 births for West Virginia residents, Hospital 22 would have had 1,329 fewer ''other congenital anomaly'' reports, three-quarter of which (1031/1,329 = 78%) would have been of neonatal conditions. Further analysis by specific condition follows.

Certain conditions were reported much more frequently for births from Hospital 22 than from West Virginia births elsewhere. The PRR was greater than 100 for diagnoses of reflux (Infinity), tongue tie (499), newborn sepsis (250), s-ASD (200), arrhythmia (115) and hip click (108) (not shown). Additionally, the PRR was found to be greater than 50 for hemangioma (96), skin tag (69), birth mark (54), and PDA (53). These diagnoses alone account for 42% of the excess diagnoses of ''other congenital anomalies''. Including additional conditions with PRR > 30 (hydrocele, undescended testes, musculoskeletal disease, chordee, Mongolian spot, circulatory-respiratory disease, and murmur) accounts for 64% of the excess diagnoses of ''other congenital anomalies.''

Table 2
Congenital Anomaly (Specified, Other, and Any) Counts and Rates for West Virginia, for Hospital 22 births and for Non-Hospital 22 births (1998–2009)

		Non-Hospital 22		Hospital 22		West Virginia				
		Number	Rate[a] (%)	Number	Rate[a] (%)	Number	Rate[a] (%)	% Hospital 22	PRR[b]	Excess[c]
Specified Birth Defect										
Live births		230,533		19,626		250,159		7.3		
ca (01)	Anencephaly	39	0.02	6	0.03	45	0.02	13	1.81	3
ca (02)	Spinabifida	46	0.02	4	0.02	50	0.02	8	1.02	0
ca (03)	Hydrocephaly	53	0.02	5	0.03	58	0.02	9	1.11	0
ca (04)	Microcephaly	128	0.06	3	0.02	131	0.05	2	0.28	−8
ca (06)	Heart	221	0.10	5	0.03	226	0.09	2	0.27	−14
ca (08)	Rectal atresia	15	0.01	3	0.02	18	0.01	17	2.35	2
ca (09)	Tracheoesoph	19	0.01	2	0.01	21	0.01	10	1.24	0
ca (10)	Omphalo/Gastroc	79	0.03	1	0.01	80	0.03	1	0.15	−6
ca (12)	Malgenital	93	0.04	3	0.02	96	0.04	3	0.38	−5
ca (13)	Renal agenesis	24	0.01	5	0.03	29	0.01	17	2.45	3
ca (15)	Cleft	228	0.10	35	0.18	263	0.11	13	1.80	16
ca (16)	PSAdactyly	103	0.04	38	0.19	141	0.06	27	4.33	29
ca (17)	Clubfoot	146	0.06	22	0.11	168	0.07	13	1.77	10
ca (18)	Diaphragmatic	25	0.01	7	0.04	32	0.01	22	3.29	5
ca (20)	Down syndrome	86	0.04	16	0.08	102	0.04	16	2.19	9
	Sum[d] - Specific	1,305	0.57	155	0.79	1,460	0.58	11	1.40	44
	Any[e] - Specific	1,093	0.47	148	0.75	1,241	0.50	12	1.59	55
Other congenital anomaly										
ca (05)	Other CNS	56	0.02	14	0.07	70	0.03	20	2.94	9
ca (07)	Other Circulatory	162	0.07	341	1.74	503	0.20	68	24.73	327
ca (11)	Other gastrointestinal	69	0.03	88	0.45	157	0.06	56	14.98	82
ca (14)	Other Urogenital	239	0.10	279	1.42	518	0.21	54	13.71	259
ca (19)	Other Musculoskeletal	208	0.09	415	2.11	623	0.25	67	23.44	397
ca (21)	Other Chromosonal	62	0.03	10	0.05	72	0.03	14	1.89	5
ca (22)	Other	1,390	0.60	371	1.89	1,761	0.70	21	3.14	253
	Sum - Other	2,186	0.95	1,518	7.73	3,704	1.48	41	8.16	1,332
	Any - Other	2,083	0.90	1,366	6.96	3,449	1.38	40	7.70	1,189
	Sum - Any[f]	3,491	1.51	1,673	8.52	5,164	2.06	32	5.63	1,376
	Any - Any	2,975	1.29	1,473	7.51	4,448	1.78	33	5.82	1,220

[a]Birth prevalence rate, i.e. number/live births.
[b]Prevalence rate ratio, i.e., Hospital 22/non-Hospital 22.
[c]Reported - Expected based on non-Hospital 22 rates.
[d]Number of such congenital anomalies reported on the birth certificates.
[e]Number of live births with such congenital anomaly reported on the birth certificate.
[f]Specified birth defect and/or other congenital anomaly.

The Neonatal Corrected Annual Rates of Congenital Anomalies

We have calculated neonatal-corrected annual rates of congenital anomalies by removing the reports of neonatal conditions from the other congenital anomaly reports uniformly across West Virginia for all hospitals and recalculated corrected West Virginia congenital anomaly rates for the 1998–2009 period (Fig. 6). The rise in the congenital anomaly rates for 1999–2003 seen in the uncorrected dataset is absent in the corrected dataset. No change in the congenital anomaly birth prevalence rates for the 12-year period 1998–2009 is seen after the data have been corrected with the exclusion of neonatal conditions.

The inclusion of neonatal conditions as congenital anomalies increased the average annual (1998–2009) proportion of infants with a reported congenital anomaly from 1.14 per 100 live births to 1.78 per 100 live births, a more than 50% increase. This effect was not limited to Hospital 22, as the change in rate for the other hospitals was about 30%, an increase from 1.0 to 1.3 per 100 live births. With the exclusion of neonatal conditions, the county rates for ''any congenital anomaly'' showed a normal distribution with no significant outliers.

Joinpoint regression analysis of the 1998–2009 data set that included the neonatal conditions demonstrated a (+9.00) annual percent change (APC) prior to 2003 ($P < 0.05$) and a (−3.52) APC after 2003. The second slope was significantly different from the first slope ($P = 0.025$) but not from zero ($P = 0.21$). In contrast, joinpoint regression analysis excluding the neonatal conditions showed a (+0.62) APC ($P = 0.47$) for the full 1998–2009 period. Thus, the bulge in the annual rate of congenital anomalies could be explained by the erroneous inclusion of the neonatal conditions as if they were congenital anomalies.

Our investigation into the textual strings for the ''other congenital anomalies'' entry into the West Virginia birth certificate has revealed that the geographic and temporal clustering of the initial epidemiological pattern reflected the incorporation of neonatal conditions into the domain of congenital anomalies. We have demonstrated that the

Table 3
Congenital Anomaly (Specific, Other, and All) Counts and Rates for Hospital 22 Catchment Area, for Hospital 22 Births and for Non-Hospital 22 Births (1998–2009)

	Non-Hosp 22		Hospital 22		Catchment area				
Diagnosis	Number	Rate[a] (%)	Number	Rate[a] (%)	Number	Rate[a] (%)	% Hosp 22	PRR[b]	Excess[c]
Live births	7,264		18,598		25,862		72		
Sum[d] - Specific	49	0.67	149	0.80	198	0.77	75	1.19	24
Any[e] - Specific	46	0.63	142	0.76	188	0.73	76	1.21	24
Sum - Other	88	1.21	1,444	7.76	1,532	5.92	94	6.41	1219
Any - Other	80	1.10	1,304	7.01	1,384	5.35	94	6.37	1099
Sum- Any[f]	137	1.89	1,593	8.57	1,730	6.69	92	4.54	1242
Any - Any	122	1.68	1,408	7.57	1,530	5.92	92	4.51	1096

[a]Birth prevalence rate, i.e. number/live births.
[b]Prevalence rate ratio, i.e., Hosp 22/non-Hosp 22.
[c]Reported - Expected, based on non-Hospital 22 rates.
[d]Number of such congenital anomalies reported on the birth certificates.
[e]Number of live births with such congenital anomaly reported on the birth certificate.
[f]Specifically-named and/or other congenital anomaly.

clustering related primarily to one facility but the misclassification was generally across the state. The inclusion of the reports of neonatal conditions had increased the apparent overall congenital anomaly rate for West Virginia from 1.1 per 100 live births to 1.8 per 100 live births. The opportunity to include neonatal conditions and other unknown conditions as congenital anomalies was a consequence of having open-ended entries for otherwise unspecified conditions included in the form. More uniform reporting may occur with the newer birth certificate that does not permit open-ended entries.

DISCUSSION

We analyzed the congenital anomaly information reported on the West Virginia birth certificates for the 20-year period 1990-2009. Among the 418,385 live births to West Virginia residents, the prevalence rates of infants with specified birth defects, other congenital anomalies, and any congenital anomaly were 0.57 %, 1.35 % and 1.82 %, respectively. Starting in 1998, the electronic records included the textual strings for the ''other congenital anomaly'' reports and that permitted an examination of the types of conditions that were being reported under the rubric of ''other congenital anomaly.'' We found that 50 % of the ''other congenital anomaly'' reports referred to neonatal conditions rather than to congenital anomalies and that an additional 21% were not classifiable as there was no description of the anomaly. Geographic and temporal patterns in congenital anomaly distributions that had initially been observed disappeared after the data were corrected by excluding the neonatal conditions. Thereafter, the pattern for both specified and other congenital anomalies were similar.

We concluded that the inclusion of data collected by use of a checked box, in the absence of a quality review of the conditions reported, had led to the acceptance of erroneous data and the development of faulty epidemiological patterns. We recommend either that open-ended entries be individually reviewed or that the use of checked boxes without specific criteria should be eliminated from the form.

Our analyses revealed that the geographical and temporal distribution of congenital anomaly reports on the West Virginia birth certificate was markedly affected by misclassification bias as a consequence of reporting neonatal conditions as if they were congenital anomalies and reporting biases observed as a hyper-reporting from Hospital 22 in Raleigh County. The inclusion of neonatal conditions as ''other congenital anomalies'' was shown to increase the proportion of West Virginia newborns reported to have congenital anomalies by over 50%, from 1.1 to 1.8%. Marked deviations in the geographic and temporal distribution of congenital anomaly reports disappeared after the dataset was corrected for either the misclassification or the reporting bias.

The purposes of birth defect surveillance systems are to establish normal patterns of occurrence, to retrospectively assess risk factors—e.g., occupational, environmental, and pharmaceutical—and to prospectively identify infants in need of medical, surgical, or rehabilitative interventions. Underlying assumptions in a surveillance system include both that reporting fractions are steady across institution and time and that consistent case defi-

Table 4
Reported ''Other Congenital Anomaly'' by Classification and Hospital (West Virginia, 1998–2009)

		Non-Hospital 22		Hospital 22		West Virginia				
Class	Diagnosis	Number	Rate (%)	Number	Rate (%)	Number	Rate (%)	% Hosp 22	PRR	Excess
A	Malformation	749	0.3	299	1.5	1,048	0.42	29	4.7	235
B	Unknown	674	0.3	120	0.6	794	0.32	15	2.1	63
C	Neonatal	764	0.3	1,096	5.6	1,860	0.74	59	16.9	1031
	Total	2,187	0.9	1,515	7.7	3,702	1.48	41	8.1	1,329
	Live births	230,533		19,626		250,159				

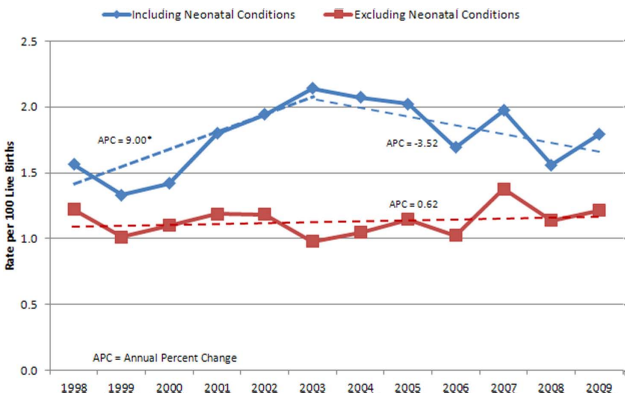


Figure 6. Rates for any congenital anomaly, both including and excluding neonatal conditions, per 100 live births with jointpoint analysis (West Virginia 1998–2009). [Color figure can be viewed in the online issue, which is available at wileyonlinelibrary.com.]

nitions are used. Violations of these assumptions would affect the validity and reliability of interpretive analyses.

We analyzed reporting fractions over three decades ago when we compared the national rates from a passive surveillance system [National Center for Health Statistics (NCHS), US birth certificates, 1974] to those from an active surveillance system [Centers for Disease Control (CDC]) Birth Defect Monitoring Program (BDMP), 1974] (Lamm, 1979). The prevalence rate ratio (PRR) varied between 27 and 77 % (median 47%), depending upon the specific birth defect.

This observation has been confirmed by others (Hexter et al., 1990; Boulet et al., 2011). Hexter et al. (1990) showed that while 1,623 infants among the San Francisco Bay five-county area 1983 births were in the California Birth Defect Monitoring Program (CBDMP) registry, the hospital discharge diagnosis index had identified 2,543 as having birth defects and the birth certificate had identified only 399 as having birth defects—clearly issues of over and under reporting. Boulet et al. (2011) showed, using 1995–2005 data from the Metropolitan Atlanta Congenital Defects Program (MACDP) registry, that the overall sensitivity for birth certificates was 23% with a range of 7–69%.

We can attempt a comparison for West Virginia. The West Virginia Department of Health submits data to the National Birth Defect Prevention Network (NBDPN) for 46 specific birth defects. For the period 2004–2008, 2,180 specific birth defects are reported for 96,533 live births for a prevalence of 2.26%. (NBDPN, 2011) This rate is more than four times the specified birth defect prevalence (0.50%) reported on the birth certificates, but the two systems are not reporting the same malformations. For the ten specified birth defects that are the same in both systems, the birth certificates reported 319 cases and the NBDPN reported 350 cases. Individually, the case count ratios (BC/NBDPN) ranged from 0.29 to 1.74 with a mean of 0.92. Because the data are deidentified, we are unable to assess the proportion of matching for each diagnosis.

In this study, we examine the second assumption. We have demonstrated that the case definitions for ''other congenital anomaly'' on the birth certificate have been used inconsistently over time and across hospitals. We have not examined the internal validity of the entries on the birth certificates. That is, are the specified birth defects confirmed from the hospital discharge or a review of the medical records? That would be a different study. We have examined whether the conditions entered as ''other congenital anomaly'' entries on the birth certificates are the conditions that are generally sought in congenital malformation surveillance programs? Do they answer the question asked? We have demonstrated for the 1998–2009 West Virginia birth defects that over 40% of these entries are erroneous responses or misclassifications.

Others have examined the issue of misclassification. Holmes and Westgate (2012) in assessing the use of ICD-9 codes 740 to 759.9 in establishing the prevalence of malformations in newborn infants excluded minor physical features such as birth marks, hemangiomas, Mongolian spots, torticollis, PDA, etc. They observed that hospital discharge records identified 1.3% of the infants as having malformations by ICD-9 coding, but found that more than half (53%) of them were minor features such as birth marks and minor anomalies.

A number of issues are raised in the published literature on factors affecting reliability and validity. Northam and Knapp (2006) proposed that birth certificate reliability and validity would be greater if completed by nurses rather than by physicians (Northam and Knapp, 2006). We examined that issue in the West Virginia data and found no difference for the reporting rates for specified birth defects [clerk/registrars (0.5%); physicians (0.6%); nurses and midwives (0.4%)]. We did find a difference for the reporting rates for ''other congenital anomalies'' [clerks and registrars (1.4%); physicians (0.8%); nurses and midwives (0.4%)], but that difference was attributable to a single hospital. When the data for Hospital 22 were removed, the subsequent reporting rates for other congenital anomalies were now similar [clerks/registrars (0.8%); physicians (0.7%); nurse/midwives (0.4%)]. Thus, the major discrepancies in ''other congenital anomaly'' entries were by clerks/registrars and were hospital-specific.

Extreme outliers of reporting rates were found for both Raleigh County and for Hospital 22 in Raleigh County, both for ''other congenital anomaly'' and for ''any congenital anomaly.'' When comparing the rates of Hospital 22 births with those of births to residents of the same five county catchment area, we concluded that the data reflected a hospital effect and not an area effect, as the observed high rates reported for Hospital 22 were not observed for the non-Hospital 22 births for the same geographical area.

There are multiple possible reasons that misclassification and reporting bias occur at hospital level. The NCHS requires that physicians complete or verify information on the certificate of live birth (USDHHS, 1987), but hospitals are known to record the data without any review by clinicians (Northam et al., 2003). The NCHS encourages states to train and certify the birth certificate data collectors who transmit data to the state's bureau of vital statistics. In reality, training may simply involve one medical record clerk helping another learn how to complete the form (Northam et al., 2003). Reliability assessments are not done routinely to monitor data quality (Northam et al., 2003). Format changes, such as the use of drop-down screens, should be examined to determine if they increase completeness and consistency in reporting.

In our study, other congenital anomalies were categorized based on the information in the text strings as (A)

Malformations, (B) Unknown, and (C) Neonatal conditions. The prevalence rate for reports of neonatal conditions was much more frequently (17-fold) for births from Hospital 22 than for other West Virginian births. The prevalence rate ratios (PRR) exceeded 100-fold for a number of conditions. Two-thirds of the excess reports of other congenital anomalies were explained by conditions with PRRs exceeding 30-fold. The inclusion of neonatal conditions as ''other congenital anomalies'' was shown to increase the proportion of West Virginia newborns reported to have congenital anomalies by 50%, from 1.1 to 1.8%.

There would be disputes among various clinicians as to which observations should be listed on the birth certificate where they would be interpreted as representing structural abnormalities with surgical, medical, or cosmetic importance. We cannot know the magnitude or severity of anomalous appearances based only on the diagnostic term. However, our analyses have the advantage over ICD-9 codes that our classification of individual neonatal conditions was based on the string text. We did not remove from the corrected dataset other congenital anomalies for which there was no diagnostic information as there was no basis to either accept or reject them, other than that they were indicated on the birth certificate. Those removals which we did perform were done uniformly without consideration of either facility of birth or county of residence.

Strengths and Limitations

The present study has evaluated the congenital anomaly information on the West Virginia birth certificate for the years 1990–2009. Compared with other research (Buescher et al., 1993; Piper et al., 1993; Watkins et al., 1996), our study has the following strengths: (1) large sample size: our analysis was based on records for 418,385 live births to West Virginia residents, which enabled sufficient statistical power to test various hypotheses; (2) long (20-year) study period: the 20-year birth certificate data from the state of West Virginia were obtained from the West Virginia Department of Health, which allowed us to examine the temporal tends of congenital anomalies over the past two decades; (3) analysis both of specified birth defects and of other anomalies: in addition to specified birth defects, we also examined the rates of other congenital anomalies for various systems. By reviewing the text strings of other congenital anomalies, we were also able to evaluate the quality and appropriateness of other congenital anomalies reported. We were fortunate that we were able to get the literal strings from the West Virginia Department of Health. Such information, if computerized, is retained only at the state level. Such a study could not be done through the national database since it does not collect the literal strings.

Our study also has limitations: (1) only partial correction of the misclassification errors: we excluded the reported neonatal conditions but were not able to assess for inclusion or exclusion the ''other congenital anomalies'' for which there was no diagnostic information since there was no evidence to either accept or reject them; (2) unable to correct for degree or severity of malformation: we did not have first-hand information on the degree or severity of malformations to see its variation across hospitals; and (3) validation: we did not obtain medical records to assess either completeness or accuracy of reporting which would not have been feasible to us under HIPPA medical confidentiality rules.

CONCLUSIONS

The 1989 revision of the US birth certificate introduced the checked-box for specified birth defects and the checked-box plus open-ended question format on an organ system basis for ''other congenital anomaly'' reports. Previous publications have evaluated accuracy of checked-box entries, but none have assessed the appropriateness of the entries in the organ-specific open-ended entries. The West Virginia data set provides such an opportunity.

We analyzed the 20-year birth certificate data from the State of West Virginia and identified both systemic bias due to changes in the data collection system and misclassification bias due to reporting neonatal conditions as if they were congenital anomalies. We found that the inclusion of neonatal conditions in ''other congenital anomaly'' reports increased the proportion of West Virginia newborns with a birth certificate report of a congenital anomaly by more than 50%. We also examined the geographical and temporal distribution of congenital anomaly reports on the West Virginia birth certification with or without exclusion of the misclassification bias. We concluded that surveillance data on other congenital anomalies was greatly susceptible to biasing influences and could only be used after misclassification bias had been accounted for. Such birth defect data should be interpreted with great caution.

We found that 1.1% of the birth certificates for West Virginia reported the occurrence of at least one congenital anomaly, after correction for misclassified neonatal conditions. We have found no significant variation in this for either specified birth defects or other congenital anomalies temporally or by county. This rate is consistent with other reported birth certificate rates but is inconsistent with the typical 3% congenital anomaly rate reported in active surveillance programs. While this degree of under-reporting is consistent with the known literature and encourages caution in the interpretation of rates, the consistency in pattern of reporting by both time and space allows for interpretive analyses.

We have demonstrated in the West Virginia data that the major source of variability across hospitals in the reporting of congenital anomalies on the 1989 version of the US standard birth certificate was related to the responses to the open-ended format queries on the ''other congenital anomalies.'' The 2003 revised birth certificate retained the checked-box format for specified birth defects and dropped the inquiry for other congenital anomalies, both the checked-box and the open-ended format. The decision in the newly revised 2003 US standard birth certificate to remove the open-ended query and restrict information collection to the checked-box format for specified birth defects was probably a wise one. We concur with the Report of the Panel to Evaluate the US Standard Certificates and Reports that the category for ''Other, specify'' would not result in useful data, and thus should not be included and coded by the States. Our study has demonstrated why, or in what way, these ''Other, specify'' data are not useful for analysis. The analysis in our article also cautions the usage of birth defects data obtained from other passive surveillance

programs, which may suffer from the misclassification issue as well.

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