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A Quality Assessment of Reporting Sources for Microcephaly in Utah, 2003 to 2013

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Abstract

Background—Obtaining accurate microcephaly prevalence is important given the recent association between microcephaly and Zika virus. Assessing the quality of data sources can guide surveillance programs as they focus their data collection efforts. The Utah Birth Defect Network (UBDN) has monitored microcephaly by data sources since 2003. The objective of this study was to examine the impact of reporting sources for microcephaly surveillance.

Methods—All reported cases of microcephaly among Utah mothers from 2003 to 2013 were clinically reviewed and confirmed. The UBDN database was linked to state vital records and hospital discharge data for analysis. Reporting sources were analyzed for positive predictive value and sensitivity.

Results—Of the 477 reported cases of microcephaly, 251 (52.6%) were confirmed as true cases. The UBDN identified 94 additional cases that were reported to the surveillance system as another birth defect, but were ultimately determined to be true microcephaly cases. The prevalence for microcephaly based on the UBDN medical record abstraction and clinical review was 8.2 per 10,000 live births. Data sources varied in the number and accuracy of reporting, but a case was more likely to be a true case if identified from multiple sources than from a single source.

Conclusion—While some reporting sources are more likely to identify possible and true microcephaly cases, maintaining a multiple source methodology allows for more complete case ascertainment. Surveillance programs should conduct periodic assessments of data sources to ensure their systems are capturing all possible birth defects cases.

Keywords

microcephaly; quality assessment; birth defects; Utah; hospital discharge data; reporting

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Introduction

Population-based birth defects surveillance programs are useful for monitoring trends and understanding demographic, maternal, and environmental risk factors related to birth defects (Feldkamp et al., 2005). Surveillance programs often need to cast a broad net, seeking information from a wide variety of data sources, for case ascertainment. Understanding the strengths and limitations of reporting sources can guide programs in determining where to prioritize efforts, such as determining the key reporting sources needed for rapid reporting in response to a public health emergency.

As part of an urgent public health response to Zika virus, the Centers for Disease Control and Prevention (CDC) is funding population-based birth defects surveillance programs to rapidly and closely monitor microcephaly and other brain abnormalities to better understand the potential impact of Zika virus. With the need to establish baseline prevalence estimates for microcephaly and to understand the contributions of different reporting sources to those estimates, the Utah Birth Defect Network (UBDN) evaluated its reporting sources for microcephaly cases from 2003 to 2013.

Materials and Methods

UBDN

The Utah Birth Defect Network (UBDN) is a statewide active case-finding surveillance program within the Children with Special Health Care Needs (CSHCN) Bureau at the Utah Department of Health (UDOH) that monitors birth defects for all Utah resident births. Established in 1994, UBDN initially monitored only neural tube defects, but expanded in 1999 to include all major structural malformations. The Utah Administrative Rule R398-5 provides UBDN legal authority to collect information on births for Utah resident mothers with a reportable birth defect. Sources for case-finding include vital records (birth certificate, death certificate, and fetal death certificate), hospital disease index (delivery and tertiary hospitals), logbooks (ultrasound, labor and delivery, neonatal intensive care units, newborn nursery), specialty clinics, cytogenetic laboratory reports, pathology results, and champions. A champion is a volunteer clinician recruited by UBDN at hospital facilities to report possible birth defect cases to the UBDN.

Case definition for a possible case includes any infant or fetus diagnosed with at least one of the structural birth defects collected by the UBDN (http://bit.ly/2egayvb) born in Utah to a resident mother. Pregnancy outcomes include live births, stillbirths ≥ 20 weeks and elective terminations. Possible cases submitted by reporting sources are first reviewed before abstraction by a tracking specialist and then entered into a Microsoft Access database. Maternal and infant medical records are abstracted from delivery and tertiary hospitals, prenatal records, specialty clinics, and pathology records. UBDN abstractors compile a comprehensive record for a possible case in preparation for clinical case review. Cases are reviewed and assigned final birth defects diagnosis by the UBDN clinical case reviewer.

MICROCEPHALY CASE DEFINITION

Case definition for congenital microcephaly for this study includes: occipital frontal circumference (OFC) at birth (or at delivery for stillbirths and elective terminations) < 10th percentile, OFC at 1 year of age ≤5th percentile or OFC at 18 months ≤ 2nd percentile for gestational age and gender based on standard growth charts, or diagnostic code 742.1.

STUDY DESIGN

This study includes all reported possible microcephaly cases diagnosed at ≤ 2 years of age occurring among live births, stillbirths ≥ 20 weeks gestation, or elective terminations to Utah residents from January 1, 2003, to December 31, 2013, identified through UBDN surveillance. Any possible case of microcephaly was abstracted by the UBDN from 2003 to 2010. From 2011 to 2013, cases of microcephaly were abstracted only when the condition co-occurred with another birth defect; however, for this study, UBDN staff retrospectively reviewed and abstracted all reported possible cases of microcephaly from 2011 to 2013 to ensure complete case ascertainment.

Traditional Sources: Clinical and Administrative Data—Clinical reporting sources report directly to the UBDN based on a list of diagnostic codes from their medical record databases (UBDN hospital disease index); these sources include hospital reported data from major birth and tertiary hospitals, clinics including pediatric ophthalmology and pediatric cardiology, pathology, neonatal intensive care units, prenatal diagnostic databases, cytogenic laboratory reports, and facility champions. Administrative sources include vital records data (birth, fetal death, and death certificates) and UDOH statewide hospital discharge dataset (Utah Department of Health, 2016). Cases identified by either a clinical or administrative source are considered “source-acquired possible cases,” which then starts the UBDN active case-finding process for tracking and abstraction.

Supplemental Source: Hospital Discharge Data—The UDOH hospital discharge dataset is not an established reporting source for routine surveillance by the UBDN; however, for this study, microcephaly cases in the UBDN database were linked to UDOH hospital discharge data to assess the utility of this database as an independent case source. The UDOH hospital discharge encounter-based data from 2003 to 2014 were used for birth cohorts occurring from 2003 to 2013. Since 2015, discharge data were not available, all births born in 2013 were detected only through 2014 (could not complete the criteria for up to 2 years of age). Because these data are encounter-based, they were de-duplicated using medical record numbers by facility and then matched to the UBDN database by means of two linkage methodologies: “electronic” and “hybrid.” “Electronic” linkage was performed using statistical software that combines deterministic and probabilistic methodologies. “Hybrid” linkage approach was used as a comparison and validation of the “electronic” method. The “hybrid” linkage was completed by matching the UDOH hospital discharge dataset to the UBDN database on exact matches in Micro-soft Access, and then exact manual match for the remaining cases to ensure maximum completion.

Reporting source accuracy was evaluated by comparing each reporting source to the reference source, the UBDN database. The reference source prevalence of microcephaly was

calculated as the number of true cases abstracted by the UBDN divided by the total live birth population for the study period (422,160). The reporting source prevalence was calculated as the number of observed cases by a reporting source divided by the total live birth population for the study period. The prevalence ratio was calculated as the reporting source prevalence divided by the referent source prevalence. The positive predictive value was calculated as the number of true cases divided by the number of possible cases reported by source(s). Sensitivity was calculated as the number of true cases detected by reporting source divided by all true cases confirmed by UBDN tracking, abstraction, and clinical case review. Positive predictive values and sensitivities were calculated using SAS 9.3 (SAS Institute, Cary, NC).

Results

For the birth cohort from January 1, 2003, through December 31, 2013, a total of 477 possible microcephaly cases were reported to the UBDN by means of 12 data sources. Of the 477 possible cases, 251 cases (52.6%) were confirmed as a true case and 226 cases (47.4%) did not meet the UBDN criteria for microcephaly and were classified as not a true case (NAC). The UBDN identified 94 cases of microcephaly that were reported as another defect by a reporting source and not reported by any reporting source as microcephaly, comprising 27.2% of all true cases of microcephaly in the UBDN database (considered nonreported source for microcephaly). In total, 345 true cases of microcephaly were confirmed in Utah from 2003 to 2013, 251 from reporting sources and 94 from abstraction only. Prevalence estimate based on reporting sources alone was 11.3 cases per 10,000 live births (477/422,160). The prevalence based on the UBDN tracking, abstraction, and clinical case review was 8.2 cases per 10,000 live births (345/422,160). Table 1 shows the distribution of possible and true cases by number of reporting sources, including single, multiple, and nonreported cases (identified by UBDN through medical record abstraction only). Of the 477 possible cases of microcephaly, 274 (57.4%) of them were only reported from a single reporting source. These cases account for 35.1% (121/345) of all true cases. However, a reported case was more likely to be a true case if reported from multiple sources than from a single reporting source (positive predictive value [PPV] 63.0% and 44.2%, respectively). Cases identified by four or more reporting sources had a PPV of 100%; that is, every possible case identified by four or more reporting sources was confirmed as a true case.

Table 2 presents the frequency of case ascertainment for all 12 reporting sources from 2003 to 2013, PPV, sensitivity, and prevalence ratio (defined as the reporting source prevalence divided by the referent prevalence). The sources for these case reports are not mutually exclusive. There are a total of 742 possible case reports for microcephaly, representing 477 possible cases from reporting sources. Of those, 64.0% (n = 475) were reported by clinical reporting sources and 35.9% (n = 267) by administrative data sources. The UBDN hospital disease index and pediatric genetics comprised the sources for 92.2% of all possible cases from a clinical reporting source while the UDOH hospital discharge data accounted for 85.4% of all possible case reports from an administrative source.

Table 3 shows the frequency of possible and true cases where each reporting source was the only source to report a possible case to UBDN. Pediatric genetics clinics were the largest

single reporting source for true microcephaly cases. They also had the highest sensitivity (23.8%) for reporting microcephaly cases and among the highest PPV (59%).

The UDOH hospital discharge dataset special linkage of microcephaly cases to the UBDN database identified 236 possible cases after de-duplication on encounter-based medical record number and UBDN identification number. Eight cases were identified by UDOH hospital discharge data as microcephaly but were not in the UBDN database. These eight cases were subsequently identified through retrospective abstraction: two cases were born out of state, which did not fit UBDN criteria, three were classified as NAC upon review by UBDN, and three were classified as true cases and should have otherwise been reported to the UBDN. The eight cases which could not be linked to the UBDN database were excluded in this present study. In total, this yielded 228 possible matches to the UBDN database to identify as possible case or true case. Forty of the 228 cases were identified by UDOH hospital discharge dataset only; of those 20 were NAC, 16 were classified as another defect, and four were identified as microcephaly as part of the UBDN tracking process before the UDOH hospital discharge data report.

Discussion

Identification of data sources effective for birth defects case ascertainment is important for continued surveillance and rapid-time monitoring in the event of a public health emergency. This study showed that a multiple source approach allowed for more complete case-finding; only 35.1% of all true cases were reported from a single source. The PPV of case reporting was 63% if multiple sources identified the potential case; it was only 44.2% if there was only one source. In approximately one quarter of the cases, relying on additional data sources pulled in cases with other birth defects, which, when abstracted, were found to be a true case of microcephaly. Active case-finding improved detection of cases that might be missed from reporting sources. Also, the PPV and sensitivity of the data sources were less useful in identifying the best data sources because larger administrative and clinical data sources provide a net for cases not reported by specialty clinics, pathology, and champions. The large change in PPV across reporting sources was due to variability in possible cases reported, not the best representation of accuracy of reporting sources.

Evaluation of reporting sources for specific defects is necessary because reporting sources vary among defects (Correa-Villaseñor et al., 2003; Tanner et al., 2010). Source ascertainment sensitivity and PPV also vary by birth defect type due to ascertainment bias. Ascertainment bias by reporting source should be considered due to several reasons (Correa-Villaseñor et al., 2003). Specialization continues to advance in medical practice, limiting the number of providers and in turn, reporting sources that provide care for conditions affecting a broader range of body systems. Another caveat to a large range in reporting source sensitivity and PPV is the variability of birth defects terminology and case definitions by reporting sources (Mai et al., 2015). Specific data sources have varying degrees of relationship with UBDN, resulting in potential information bias. Pediatric genetics is a unique data source due to the long-standing relationship with UBDN, and this source reports both inpatient and outpatient records. In contrast, the UBDN hospital disease index and UDOH hospital discharge database only report inpatient data. Logbooks were used before

the implementation of electronic medical records (EMR). Abstractors would visit hospitals to review logbooks and these abstractors were familiar with the specific criteria for each reportable birth defect. The transition to EMR has omitted the process of reviewing logbooks, allows for quicker case ascertainment and availability of more clinical data sources through electronic file transfer into the UBDN database. A limitation of the transition to EMR is that the EMR is not reviewed by abstractors before receipt by UBDN, leading to variability in the data quality (Mai et al., 2015).

Administrative data sources are necessary contributors to the UBDN database. Vital records data including birth and death certificates report routinely to UBDN. In comparison to studies evaluating other birth defects (Boulet et al., 2011), the sensitivity of vital records data for microcephaly ascertainment was low (5.8%) for this study. However, vital records data did identify three cases of microcephaly that were not reported by any other source.

Relying on a single source of reported cases without validation may lead to inclusion of false positives. In this study, the UDOH hospital discharge dataset prevalence estimate was most similar to the UBDN referent prevalence before linkage (8.2 per 10,000 live births), yet false positives accounted for approximately half of all possible cases. Further analysis is necessary to evaluate the false positives from hospital discharge data. Previous studies have identified false positives as a result of miscoding or contradictions in physician notes (Callif-Daley et al., 1995). In the need for rapid case ascertainment or further sub analysis, the UDOH Vital Records and hospital discharge datasets would not be the most accurate reporting source as the primary representation of population surveillance. These findings are similar to previous studies on administrative data sources. (Hexter and Harris, 1991; Watkins et al., 1996; Boulet et al., 2011; Li et al., 2013).

The study had several strengths. First, detailed microcephaly case information was abstracted from medical records and then reviewed by an experienced clinician for case accuracy. While laborious, this process allowed for more complete and accurate case information. Second, the program had the ability to examine the unique contributions of different types of data sources for microcephaly for more than a decade. Additionally, this study was able to compare a common data source that several population-based birth defects programs use for its primary birth defects data (statewide hospital discharge dataset) against a referent database (UBDN).

The study was also subject to some weaknesses. The generalizability of the study findings given the variations expected in data sources across population-based birth defects surveillance programs is unclear. For example, while approximately one-third of true cases were identified by only one reporting source for UBDN, this proportion could vary across state surveillance programs. Additionally, the lack of 2015 UDOH hospital discharge data limited the 2013 birth cohort linkage to one instead of two years; in contrast the 2003 to 2012 birth cohorts all had 2 years of discharge data linkage. This could have potentially biased the data linkage but the effect was most likely minimal because most cases of congenital microcephaly were captured within the first year of life (2014 discharge file). Finally, the applicability of these results to the Zika virus response could be impacted by the

fact that the UBDN case definition for microcephaly during the study period differs from the current recommended case definition used in the Zika virus response.

CONCLUSIONS

Birth defect surveillance systems need to ensure timely and accurate reporting of cases, especially for immediate response to public health emergencies. Regular validation of reporting sources will help ensure that the appropriate number and type of sources are accessed. Maintaining a diverse pool of case ascertainment sources is necessary to identify all possible cases of microcephaly.

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TABLE 1

Distribution of Number of Reporting Sources and Abstraction for Possible and True Cases of Microcephaly in the Utah Birth Defect Network, 2003 to 2013 (N = 571)

Number of reporting sources[a]	Possible cases (*n* = 571)	True Cases (*n* = 345)	PPV (%)	Sensitivity (%)[c]
Non-reported[b]	94	94	100.0	27.2
1	274	121	44.2	35.1
2	152	89	58.6	25.8
3	41	31	75.6	8.9
4	9	9	100.0	2.6
5	1	1	100.0	2.9
Multiple[d]	203	130	63.0	37.7

[a] Reporting sources include: champions at medical facilities, cytogenetics testing facilities, logbooks from hospital units, pathology reports, pediatric cardiology, pediatric center neonatal intensive care units, pediatric genetics, pediatric ophthalmology, prenatal diagnostics, UBDN hospital disease index, UDOH hospital discharge dataset, and vital records.

[b] Microcephaly was found during abstraction of these cases for other defects.

[c] Sensitivity calculated as number of true cases divided by all true cases confirmed by UBDN tracking, abstraction, and clinical case review.

[d] Multiple sources include all possible cases reported by two or more reporting sources.

TABLE 2

Cases of Microcephaly by Reporting Sources, Utah Birth Defect Network, 2003 to 2013

Type of reporting source[a]	All possible cases	True cases	PPV (%)	Sensitivity (%)[b]	Prevalence ratio[c]
Champions at medical facilities	5	3	60.0	0.9	0.014
Cytogenetics specialty clinic	1	1	100.0	0.3	0.003
Logbooks from hospital units	3	3	100.0	0.9	0.009
Pathology	3	1	33.3	0.3	0.009
Pediatric cardiology	1	0	0.0	0.0	0.003
Pediatric center neonatal intensive care unit	14	11	78.6	3.2	0.041
Pediatric genetics	191	128	67.0	37.1	0.554
Pediatric ophthalmology	7	5	71.4	1.5	0.020
Prenatal diagnostic	3	1	33.3	0.3	0.009
UBDN disease index	247	139	56.3	40.3	0.716
UDOH hospital discharge dataset	228	121	53.1	35.1	0.664
Vital records	39	20	51.3	5.8	0.113

[a] The reporting sources are not mutually exclusive.

[b] Sensitivity calculated as number of true cases divided by all true cases confirmed by UBDN tracking, abstraction, and clinical case review.

[c] The prevalence ratio is calculated by dividing the reporting source prevalence by the prevalence in the UBDN referent source prevalence (8.2/10,000 live births), based on 345 true cases and live birth population of 422,160.

TABLE 3

Cases of Microcephaly Identified Exclusively by a Single Reporting Source, Utah Birth Defect Network, 2003 to 2013 (N = 274)

Type of reporting source	Possible cases (exclusive reporting by single data source)	True cases	PPV (%)	Sensitivity (%)[a]
Champions at medical facilities	1	0	0.0	0.0
Cytogenetics specialty clinic	1	1	100.0	0.3
Logbooks from hospital units	1	1	100.0	0.3
Pathology	2	0	0.0	0.0
Pediatric cardiology	1	0	0.0	0.0
Pediatric center neonatal intensive care unit	5	3	60.0	0.9
Pediatric genetics	139	82	59.0	23.8
Pediatric ophthalmology	3	2	66.7	0.6
Prenatal diagnostic	2	1	50.0	0.3
UBDN disease index	63	24	38.1	7.0
UDOH hospital discharge dataset	40	4	10.0	1.2
Vital records	16	3	18.8	0.9

Possible cases = reported cases; true cases = clinically reviewed and confirmed as microcephaly cases.

[a]Sensitivity calculated as number of true cases divided by all true cases confirmed by UBDN tracking, abstraction, and clinical case review.