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Status of population-based birth defects surveillance programs before and after the Zika public health response in the United States

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Abstract

Background: The 2016 Zika public health response in the United States highlighted the need for birth defect surveillance (BDS) programs to collect population-based data on birth defects potentially related to Zika as rapidly as possible through enhanced case ascertainment and reporting. The National Birth Defects Prevention Network (NBDPN) assessed BDS program activities in the United States before and after the Zika response.

Methods: The NBDPN surveyed 54 BDS programs regarding activities before and after the Zika response, lessons learned, and programmatic needs. Follow-up emails were sent and phone calls were held for programs with incomplete or no response to the online survey. Survey data were cleaned and tallied, and responses to open-ended questions were placed into best-fit categories.

Results: A 100% response rate was achieved. Of the 54 programs surveyed, 42 reported participation in the Zika public health response that included BDS activities. Programs faced

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CONFLICTS OF INTEREST

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DISCLAIMER

The findings and conclusions in this report are those of the authors and do not necessarily represent the official position of the Centers for Disease Control and Prevention.

challenges in expanding their surveillance effort given the response requirements but reported mitigating factors such as establishing and enhancing partnerships and program experience with surveillance and clinical activities. Beyond funding, reported program needs included training, surveillance tools/resources, and availability of clinical experts.

Conclusions: Existing BDS programs with experience implementing active case-finding and case verification were able to adapt their surveillance efforts rapidly to collect and report data necessary for the Zika response. Program sustainability for BDS remains challenging; thus, continued support, training, and resource development are important to ensure that the infrastructure built during the Zika response is available for the next public health response.

Keywords

birth defects; congenital anomalies; population-based surveillance; public health emergency response; Zika virus

1 | INTRODUCTION

Birth defects are structural malformations present at birth with mostly unknown etiology (Toufaily, Westgate, Lin, & Holmes, 2018). The recent discovery of Zika virus as a novel teratogen (Rasmussen et al., 2016) and the national and international emergency public health response to understand the impact of the Zika virus infection on adverse birth outcomes, especially brain anomalies, highlighted the need for surveillance systems to collect data rapidly on potentially affected populations (Honein et al., 2017).

While the United States lacks a national population-based surveillance system to monitor major birth defects, over 40 states have established birth defects surveillance (BDS) programs. They vary in their case inclusion criteria, ascertainment methodologies, degree of case verification, and comprehensiveness of data variables collected (Mai et al., 2016).

As part of the national Zika virus emergency response, the Centers for Disease Control and Prevention (CDC) funded state health departments and other jurisdictions to establish or enhance birth defects surveillance (BDS) programs to conduct active surveillance of birth defects potentially related to the Zika virus infection. Programs were required to use standard case definitions, expand data sources to include nontraditional sources, such as specialty clinics and state laboratory reports, conduct case verification via medical record abstraction, and use innovative and timely approaches to rapid data collection (CDC, 2016).

For many BDS programs, the emergency response environment was different from the scope and speed of their routine surveillance activities. In particular, the response requirements of standardized data collection, an expansion of surveillance activities for complex birth defect cases with comprehensive data collection forms and case verification presented BDS programs with both challenges and opportunities.

The National Birth Defects Prevention Network (NBDPN) works to maintain a national network of state and other population-based BDS programs and serves as a forum for guidelines and standards, data collaborations, and development of resources to improve surveillance practice and data utilization (www.nbdpn.org). Given the impact of the Zika

response on BDS programs in the United States, the NBDPN sought to assess BDS program activities before and after the Zika response, identify lessons learned to prepare BDS programs for the next public health response, and determine program needs to support BDS activities.

2 | METHODS

In fall 2017, an ad hoc NBDPN workgroup developed a survey to assess population-based BDS program activities in the United States before and after the Zika response. The survey included questions about population-based BDS surveillance methodologies before 2016, participation in the Zika emergency response, resources and tools used, lessons learned, and challenges moving forward after the emergency response.

We piloted the survey with contacts in new and established birth defects programs in six states: Colorado (CO), Florida (FL), Massachusetts (MA), Pennsylvania (PA), Texas (TX), and Utah (UT). We solicited additional feedback from the NBDPN Executive Committee and developed a final version in SurveyMonkey (www.surveymonkey.com).

In October 2017, the online survey was sent to population-based BDS program contacts in 50 states, District of Columbia (DC), Puerto Rico (PR), and two federally administered programs—CDC Metropolitan Atlanta Congenital Defects Program (MACDP) and Department of Defense Birth and Infant Health Registry (DoD). Follow-up emails were sent and/or phone calls were made to programs that had not responded to the online survey or had incomplete or unclear responses. By mid-December 2017, all respondents had completed the survey.

We cleaned and analyzed the survey data using Microsoft Excel. Questions with multiple answers were analyzed using both mutually exclusive and nonmutually exclusive groupings. Responses to open-ended questions were reviewed and placed into best-fit themes/categories.

3 | RESULTS

3.1 | Program characteristics before Zika

Survey responses were collected from all states, DC, PR, MACDP, and DoD (N = 54; 100% response rate). Before 2016, there were 45 BDS programs across the United States; nine states had no BDS activities. Of the 45 programs, 15 (33%) accepted cases as reported through administrative data sources or passive reporting from facilities, and 14 programs (31%) obtained cases through administrative data sources or facility reporting and performed case verification for some birth defects. The remaining 16 programs (36%) had active case-finding in which program staff went to birthing and other facilities to identify cases and obtain information from medical records.

Selected characteristics of BDS programs in the United States before 2016 (N = 45) are presented in Table 1. The most common case-finding strategy for identifying potential cases examined administrative databases, while the most common case verification included checking medical records to ensure case inclusion. When examining the number of methods

used for case-finding or case verification, 35 (78%) programs reported only one or two methods for flagging potential cases, and 16 (36%) programs reported only one procedure for verifying cases while 14 (31%) reported no procedures for case verification.

Thirty-five (78%) programs linked to selected data elements from administrative data sets, such as the birth certificates (Table 1). Over half of BDS programs (58%) reported medical record abstraction at birthing facilities, and most of them also abstract at in-patient specialty hospitals and a smaller subset abstract at outpatient specialty sites. About one-third of programs reported case review by a clinical geneticist or other specialized clinical expert.

Most programs had a program-specific database that was developed in-house (67%), and the most commonly used statistical analytic software (not mutually exclusive) included SAS (80%), Excel (44%), and Access (38%). Before 2016, over 95% of BDS programs (43/45) reported incorporating some type of data quality assurance strategy into their program. The most commonly cited data quality activities were quality “runs” performed periodically to check the data set, reported by 30 (67%) programs, and the front-end editing built into the abstraction form and/or data system, reported by 26 (58%) programs. In addition, 21 (47%) programs reported review of case record(s) by other staff; 11 (24%) reported reassessment of a sample of records by other staff (e.g., reabstracting); and 2 (4%) reported other method (e.g., staff training and quality assurance audits) (Table 1).

BDS program contacts (N = 54) were also asked what public health initiatives they had participated in prior to 2016. The most frequently reported initiatives were participation in maternal and child health engagements, such as working with Maternal and Child Health/Children with Special Health Care Needs (MCH/CSHCN) (70%), and newborn screening for critical congenital heart defects (67%) (data not shown). Other initiatives mentioned included Neonatal Abstinence Syndrome (17%), cytomegalovirus infection prevention efforts (13%), and programs to reduce infant mortality(6%). However, seven programs reported not participating in any other public health initiative.

3.2 | BDS program activities during the Zika public health emergency response

Of the 54 programs surveyed, 42 programs reported a Zika public health response in their state that included the BDS program. Figure 1 presents Zika-related BDS activities that programs initiated or changed. Performing case identification, verification, and reporting in a rapid manner was a new undertaking for most BDS programs. Even programs that already performed certain required activities (e.g., medical record review) had to adjust their procedures to meet the Zika BDS requirements. When these responses were stratified by type of BDS program in place before 2016, programs with active case-finding or passive case-finding with case verification were more likely to have key surveillance elements (e.g., medical record abstraction, clinical review) in place that could be adapted to the ZBDS response while passive programs without case verification had to build these elements into their system (data not shown).

CDC provided tools and resources to assist BDS programs during the Zika response. Most programs reported using the birth defects case inclusion guidance for medical record review

(74%) and reporting guidance (76%). Additionally, more than half used the data abstraction guide (62%) and database tools (about 50%).

In addition to BDS programmatic enhancements and modifications, programs reported shifts in their partnership engagement (Figure 2). Most BDS programs enhanced their working relationships with existing MCH/CSHCN programs (79%) and agency's legal office (43%), while establishing new collaborations with infectious disease programs (67%) and local health departments (17%). Over half (52%) forged enhanced or new partnerships with pediatric neurology specialists given the birth defects of interest (brain and other central nervous system conditions).

As they moved toward active Zika BDS, programs reported challenges and barriers most frequently associated with rapid medical record abstraction and data management. The most common way that programs mitigated challenges was by establishing and enhancing partnerships and collaborations. Programs also added staff, improved reporting systems, and accessed additional data sources for case ascertainment. Examples included obtaining faster access to records through secure remote access and working with other public health programs such as the Early Hearing Detection and Intervention program to obtain more complete case information.

3.3 | Moving forward after Zika emergency response

Programs were asked to identify the top lessons learned from their program's Zika response experience that could help them prepare for another emerging public health response. The most common lesson reported by programs was engagement with new partners or enhancing relationships with existing partners (52%), followed by better planning (21%), communication (9%), and training of staff (7%).

Without future Zika BDS funding, 14% of programs responded that they could continue to conduct active, population-based Zika-related birth defects monitoring. Among the remaining programs, half said they could continue but with some modifications, while the other half would have to halt active case ascertainment surveillance.

All programs surveyed were asked the extent to which they are now better prepared to respond to another public health emerging issue (Figure 3). The programs indicated better preparedness due to new and improved partnerships, increased experience, and enhanced databases. However, programs indicated a need for assistance with several surveillance activities. Activities including ascertaining prenatally diagnosed cases, training data collectors on medical record abstraction, following-up on infants (typically through 1 year), and locating and training clinical reviewers would require assistance. Programs also reported their greatest needs in order to meet their upcoming challenges within the next 2 years (Table 2); these included staffing (65%), training/resource development (63%), funding (54%), program public health/legislative authority (33%), data issues (33%), and buy-in / program support (28%).

4 | DISCUSSION

Although most states conducted population-based BDS prior to the emergency Zika public health response in the United States, the states which had BDS programs that incorporated elements, such as active case-finding or case verification from medical records were poised to adapt and respond rapidly to an emerging public health threat. Only 36% of BDS programs performed active case-finding and 62% indicated experience in obtaining at least some birth defects data from medical records. The rapid, multisource, active case-finding, verification, and reporting required for Zika BDS presented challenges for many programs.

Additional factors enhanced a BDS program's ability to successfully respond and to rapidly provide high quality data to inform the response. These factors included: existing collaborations and partnerships with other public health programs, BDS program visibility and leadership support, existing program authority (state statutes and rules/regulations that underlie the program's ability to perform surveillance activities), flexibility of the program, and staff to manage change, and program readiness (in terms of experience, training, technical expertise). States and other jurisdictions with population-based BDS programs are encouraged to build and sustain their programs with these qualities to be better prepared to respond to another public health emerging issue, such as the opioid epidemic.

Traditionally, BDS programs have participated in other public health initiatives that were mostly restricted to the maternal and child health arena. However, the Zika public health response required a shift in routine BDS activities and engagement with a wider, multidisciplinary cadre of partners and collaborators. Partnerships were vital to BDS program success. Partners supported BDS programs in a number of ways, including the provision of resources and buy-in regarding the value of BDS for multiple purposes. Examples of partnership engagement were highlighted as part of CDC's "features from the front line" and "stories from the field" (CDC, 2018).

Almost all programs (95%) performed some data quality activities prior to 2016. The high proportion of programs with data quality activities may reflect, in part, the NBDPN work on data quality including the establishment and implementation of data quality standards for BDS programs in 2014, which have been measured annually via program self-assessments (Anderka et al., 2015). However, enhancements in routine data quality activities are needed as only 58% of programs reported having front-end editing built into their abstraction form and /or data system and only 67% reported that data quality runs were periodically performed to check the data set.

Access to clinical experts to assist with complex birth defects cases was another key element for successful participation in the Zika response. Prior to 2016, only one-third of the programs reported access to a clinical geneticist or other specialized clinical expert for conducting clinical case reviews. This is less than what was reported previously by Lin, Forrester, Cunniff, Higgins, and Anderka (2006). Clinicians, in particular clinical geneticists, can assist BDS programs with review of medical records, case classification, coding, staff education/training, and networking with other physicians (Lin, Rasmussen, Scheuerle, & Stevenson, 2009). Their participation can contribute to improvements in the accuracy and

completeness of BDS data. Thus, BDS programs are encouraged to prioritize working with these clinicians and participate in training them for public health surveillance of birth defects.

Established population-based BDS programs enable a response to local and national issues with birth defects and related adverse outcomes, provide ongoing monitoring for program planning and evaluation, help communities through referral to services, outreach, and improved care, and contribute to research to determine risk factors for birth defects. Funding for Zika BDS has improved the BDS infrastructure in several ways. BDS programs have forged new partnerships and improved existing relationships, gained experience participating in a public health response, upgraded their databases and put better processes and procedures in place. However, program sustainability will be challenging once Zika BDS funding is expended. There is a need for alternative strategies and tool development for those who plan to halt completely (or reduce) the new or enhanced activities that were implemented for Zika BDS. Many programs need resources to continue to sustain activities at the level they had during the Zika response. Therefore, continued assistance through resource and tool development and training is essential to support state BDS programs. An NBDPN Action Plan will be developed to prioritize support to state programs based on the needs expressed in this survey.

The Zika response highlights the need for core, well-resourced public health surveillance programs, and provides a lesson with relevance across the public health spectrum. The Zika response demonstrated that having a population-based BDS program in place was important—those programs with existing active case-finding or case verification were the ones capable of responding most swiftly in the face of a public health emergency (Delaney et al., 2017). BDS programs are encouraged to consider the key program components that influenced a successful Zika response and strive to incorporate those features into their program to improve their readiness to respond to a future public health threat.

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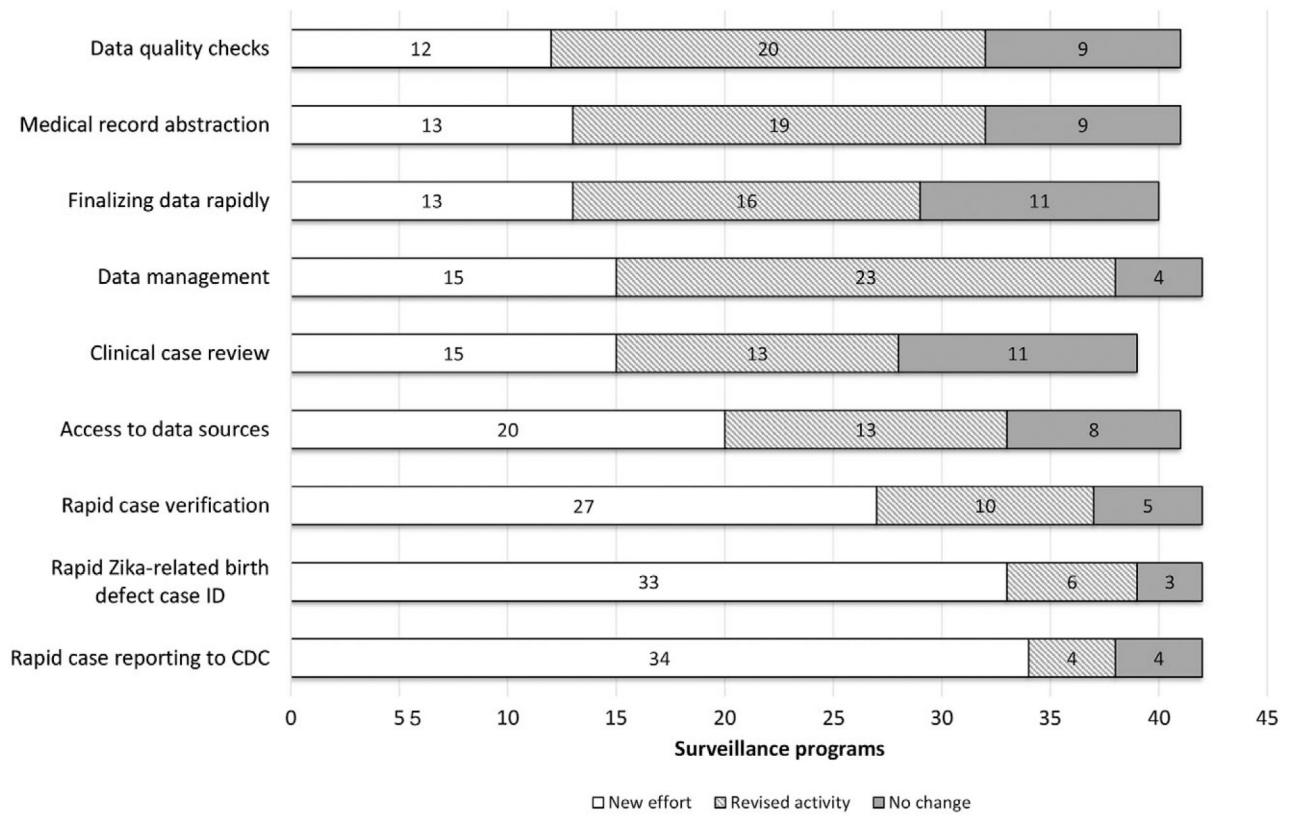


FIGURE 1. Birth Defects Surveillance Activities Implemented During Zika Public Health Response, United States (N = 42[†])[‡]
[†]Number of programs that reported a Zika public health response in their state that included the BDS program. [‡]Activities are not mutually exclusive. [§]ID-identification

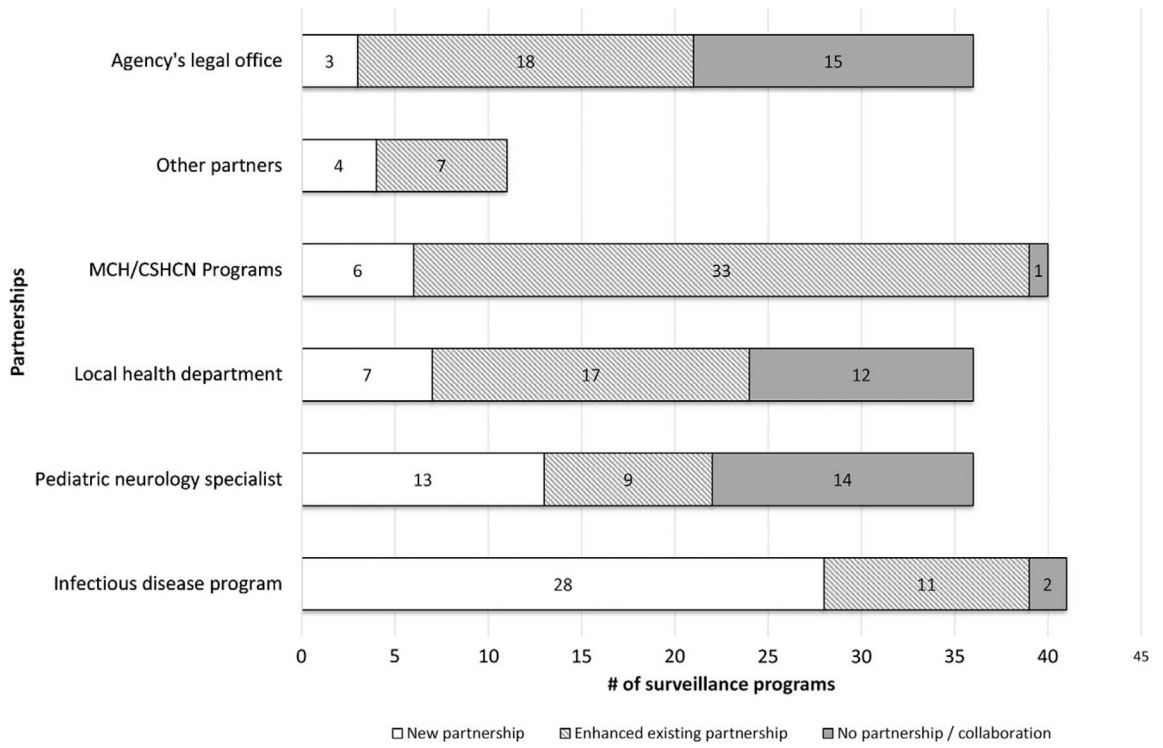


FIGURE 2. Partnerships Established by Birth Defects Surveillance Programs During Zika Public Health Response, United States (N = 42[†])[‡]
[†]Number of programs that reported a Zika public health response in their state that included the BDS program. [‡]Categories are not mutually exclusive. [§]MCH/CSHCN–Maternal and Child Health/Children with Special Health Care Needs

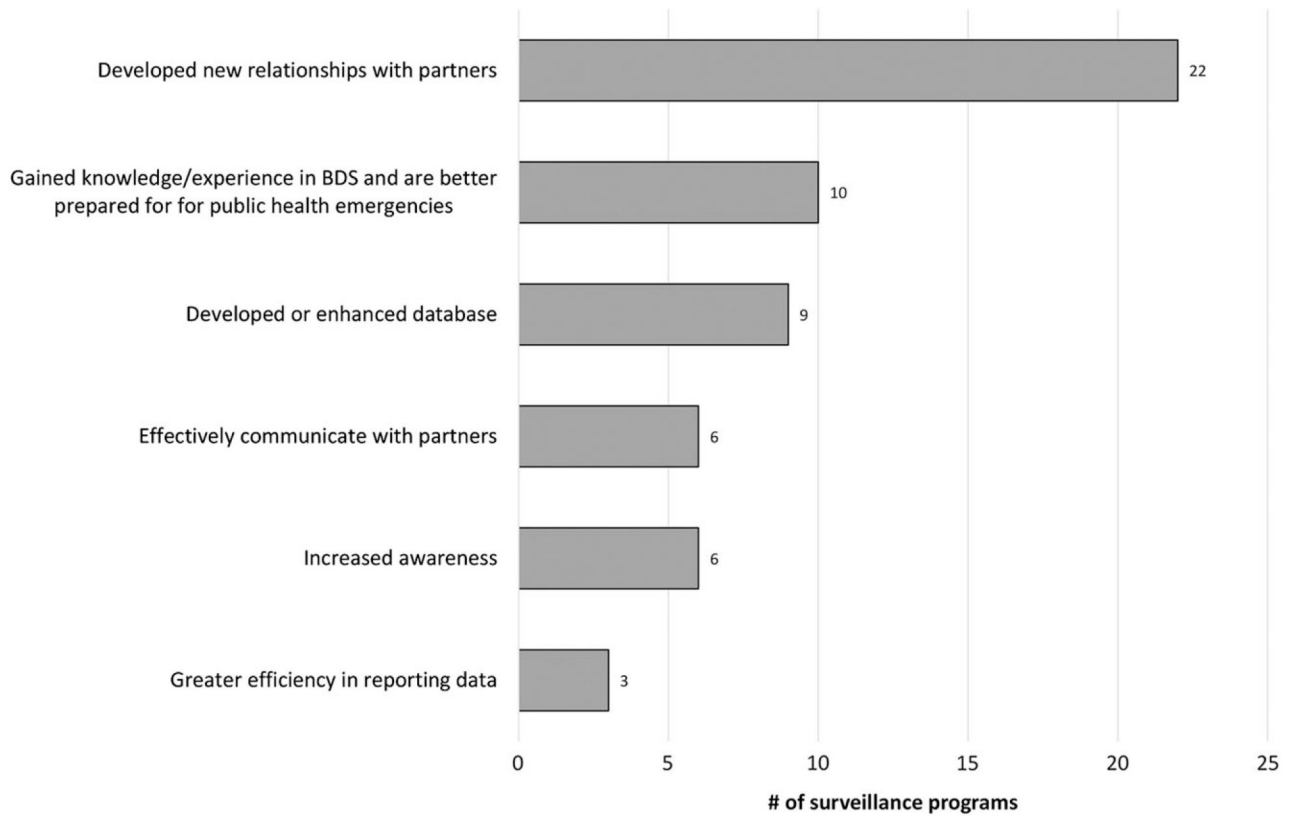


FIGURE 3. Areas that Birth Defects Surveillance Programs Reported to be Better Prepared for the Next Public Health Response (N = 54)[†]
[†]Categories not mutually exclusive. Ten programs did not respond

TABLE 1

Selected surveillance characteristics of population-based birth defects surveillance programs in the United States, before January 2016 (N = 45)

Characteristics (not mutually exclusive)	Programs number percent (%)	
Procedures for finding cases, and abstracting/processing data		
Flagging potential cases		
Administrative databases	33	73
Provider reporting of individual cases	22	49
Facility generated reports (e.g., disease index)	21	47
Search through facility unit logs	7	16
Verifying potential cases		
Checking medical records for verification of birth defect(s) meeting program's inclusion criteria	28	62
Checking verbatim diagnosis against reported code(s)	15	33
Confirm with provider	14	31
None, accepted case as reported	14	31
Completing case data		
Link to data elements from administrative data sets	35	78
Abstract infant medical records at birthing facilities	26	58
Abstract infant records at specialty hospitals (inpatient)	21	47
Abstract infant records at outpatient specialty sites	12	27
Abstract mom medical records	15	33
Conducting clinical case reviews		
Staff with disease coding or clinical training (e.g., RN)	16	36
Clinical geneticist or other specialized clinical expert	15	33
Staff with no or minimal clinical expertise	7	16
Tools for working with data		
Store/manage data		
Program-specific database developed in-house	30	67
Program-specific database or module developed by an outside vendor	15	33
Excel spreadsheet	6	13
Analyze data (software used)		
SAS	36	80
Excel	20	44
Access	17	38
SPSS	7	16
Other (e.g., R, SQL, tableau, STATA)	9	20
Quality assurance activities		
Data quality runs periodically performed to check data set	30	67
Front-end editing built into abstraction form and/or database	26	58
Review of the record(s) by other staff	21	47
Re-abstracting (a sample of records by other staff)	11	24

TABLE 2

Current or anticipated (within the next 2 years) program needs and challenges (N = 54)

Program needs or challenges (not mutually exclusive)	Programs number percent (%)	
Staffing	35	65
Examples include staff retention, succession planning		
Training/resource development	34	63
Examples include coding and case definitions, clinical review, database		
Funding	29	54
Authority	18	33
Examples include data access or cooperation from data sources, data sharing, and reporting compliance		
Data	18	33
Examples include data management, database development/compatibility, data quality, analysis, and reporting		
Buy-in/program support	15	28
Examples include internal public health department staff, reporters/external partners		
Other	8	15
Examples include infrastructure development, outreach		

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