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Scientific impact of the National Birth Defects Prevention Network multistate collaborative publications

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Abstract

Background: Given the lack of a national, population-based birth defects surveillance program in the United States, the National Birth Defects Prevention Network (NBDPN) has facilitated important studies on surveillance, research, and prevention of major birth defects. We sought to summarize NBDPN peer-reviewed publications and their impact.

Methods: We obtained and reviewed a curated list of 49 NBDPN multistate collaborative publications during 2000–2022, as of December 31, 2022. Each publication was reviewed and classified by type (e.g., risk factor association analysis). Key characteristics of study populations

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and analytic approaches used, along with publication impact (e.g., number of citations), were tabulated.

Results: NBDPN publications focused on prevalence estimates ($N = 17$), surveillance methods ($N = 11$), risk factor associations ($N = 10$), mortality and other outcomes among affected individuals ($N = 6$), and descriptive epidemiology of various birth defects ($N = 5$). The most cited publications were those that reported on prevalence estimates for a spectrum of defects and those that assessed changes in neural tube defects (NTD) prevalence following mandatory folic acid fortification in the United States.

Conclusions: Results from multistate NBDPN publications have provided critical information not available through other sources, including US prevalence estimates of major birth defects, folic acid fortification and NTD prevention, and improved understanding of defect trends and surveillance efforts. Until a national birth defects surveillance program is established in the United States, NBDPN collaborative publications remain an important resource for investigating birth defects and informing decisions related to health services planning of secondary disabilities prevention and care.

Keywords

birth defects; birth defects network; chromosomal abnormalities; congenital anomalies; data utilization; non-syndromic; prevalence ratios; registries; surveillance programs

1 | BACKGROUND

Birth defects are congenital abnormalities of body structure or function (including genetic abnormalities) that affect approximately 3% of births in the United States. As a leading cause of infant mortality in the United States, birth defects contribute substantially to adverse outcomes and morbidity, in many instances, requiring surgeries and/or contributing toward life-long disability, as well as psychosocial stress for affected individuals and family members (Almli et al., 2020; Banu et al., 2022; Christianson et al., 2006; Diseth & Emblem, 2017; Petrini et al., 2002). The National Birth Defects Prevention Network (NBDPN) is a collaborative consortium of birth defects surveillance programs, research centers, and individuals involved in population-based birth defects surveillance, research, and prevention throughout the United States. NBDPN was formed in 1997 to address the need for (1) multi-state, population-based birth defects surveillance datasets, (2) harmonization of data quality and surveillance practices across programs, (3) development of national prevention strategies, and (4) promotion of national research collaborations for birth defects. NBDPN has provided many opportunities for members of the birth defects surveillance and research community to enhance surveillance, advance science, and utilize population-level data for public health practice.

Given the lack of a national, population-based, birth defects surveillance program in the United States, multistate collaborative publications have been a major NBDPN activity requiring pooling and analyzing individual-level data from state birth defects surveillance programs. Unlike clinic-based populations or case–control studies for which individuals must consent to participate, population-based data are robust to potential selection bias,

which make them the gold-standard for birth defects epidemiological research. The increased sample size provided by NBDPN pooled data has created analytic opportunities for specific defects, population subgroups, and outcomes that are relatively rare in a single state (e.g., improved statistical power). Thus, these collaborations are designed to improve the accuracy of scientific information on birth defects, which inform decisions related to healthcare service planning of secondary disabilities prevention and intervention. Herein, we summarize NBDPN multistate collaborative publications (henceforth, NBDPN publications) to assess their contribution and impact to the literature on birth defects surveillance and research.

2 | METHODS

NBDPN provided a curated list of all peer-reviewed NBDPN publications from 2000 to 2022 ($N = 49$, available at https://www.nbdpn.org/collab_projects.php), and all co-authors reviewed the list to ensure that no publications were missing. We reviewed and summarized the study population characteristics (delivery years, sites, primary exposure[s] and outcome[s] of interest, and number of case children) for each publication as well as the journal's name. A publication was assigned to one of five categories, based on the primary research question(s) addressed: prevalence analyses, descriptive epidemiology association analyses, risk factor association analyses, outcome association analyses, or surveillance methods. Because a publication may span multiple categories, particularly the first three listed categories, hierarchical assignment was used (prioritization order: risk factor association analyses, descriptive epidemiology association studies, prevalence studies).

Selected characteristics of analytical methods used were summarized for each association publication (whether analyses were adjusted for at least one covariate; accounted for case children with chromosomal abnormalities via stratification, restriction/exclusion, or adjustment; or accounted for case children with isolated versus multiple defect phenotypes via the same approaches). Information from publications was initially abstracted by J.T.B. and confirmed by S.B.S., with additional review from A.J.A. when there was uncertainty and/or disagreement. For each publication, we also determined the journal's impact factor for the most recent available year, per the Clarivate Analytics 2021 Journal Citation Report (Clarivate Analytics, 2023a), and, for publications before January 1, 2021, the number of times the publication was cited by February 28, 2023, per the Clarivate Analytics' Web of Science Core Collection (Clarivate Analytics, 2023b), formerly known as the Thomson Reuters Web of Science.

3 | RESULTS

3.1 | Overview

Of the 49 NBDPN publications reviewed (Table 1), 11 reported on prevalence of individual major birth defects, 6 on prevalence of a spectrum of defects, 5 on descriptive epidemiology association analyses, 10 on risk factor association analyses, 6 on outcome association analyses, and 11 on surveillance methods. Differing numbers of US surveillance programs contributed data to these publications, ranging from 4 to 54 programs across the different publication types, including some programs that ascertained fetal deaths and/or terminations

in addition to live births. The pooled samples in many of these studies represent one-quarter or more of all birth defects in the United States (Parker et al., 2009), which emphasizes both the datasets' expected national generalizability and high statistical power. Publications with the highest numbers of citations were prevalence analyses of a spectrum of defects (e.g., N = 1161 citations) or individual defects (e.g., N = 339), followed by risk factor association analyses (e.g., N = 245 citations).

3.2 | Prevalence analyses

3.2.1 | Individual defects—Eleven publications (using pooled data from 12 to 41 surveillance programs) focused primarily on US prevalence estimates for specific major defects or groups of defects (Table 1), including critical congenital heart defects (CCHDs) (Mai et al., 2012; Stallings et al., 2022), neural tube defects (NTDs) (Williams et al., 2015), gastroschisis (Kirby et al., 2013), orofacial clefts (IPDTOC Working Group, 2011; Mai et al., 2014), eye and ear defects (Stallings et al., 2018), gastrointestinal defects (Lupo et al., 2017), congenital microcephaly (Cragan et al., 2016), and trisomy chromosomal abnormalities (Heinke et al., 2021; Mai et al., 2013; Stallings et al., 2022). Publications reporting population-based period prevalence and prevalence trends have been instrumental in delineating the impact of changes in birth defects screening and prevention. The most cited publication (Williams et al., 2015) suggested a continued impact of mandatory folic acid fortification in the United States during the post-implementation period, with years of relatively stable, reduced prevalence of NTDs. Although most defects described across the 11 publications were relatively common compared to rarer defects, the pooled data allowed estimating prevalence of relatively rare trisomies stratified by maternal age categories, time periods, and/or other variables. To illustrate, one publication tabulated the prevalence of trisomy 13 simultaneously stratified by both pregnancy outcome and time period (2000–2004 vs. 2006–2010) (Mai et al., 2013). NBDPN data also have been used to contribute to worldwide estimates of birth defects prevalence, as was the case in one highly cited publication that combined NBDPN data with data from the European Surveillance Systems of Congenital Anomalies and International Clearinghouse for Birth Defects Surveillance and Research to establish an estimate of the worldwide prevalence of cleft lip with or without cleft palate (IPDTOC Working Group, 2011).

3.2.2 | Spectrum of defects—Six publications (using pooled data from 11 to 39 surveillance programs) described US prevalence estimates across a spectrum of selected major defects, at various temporal periods, including 1999–2001 (Canfield et al., 2006; Centers for Disease and Prevention, 2006), 1999–2007 (St. Louis et al., 2017), 2004–2006 (Parker et al., 2010), 2008–2012 (Mai et al., 2015), and 2010–2014 (Mai et al., 2019). The estimates generated highlight the importance of NBDPN as a unique dataset, as some of these are the most cited of all NBDPN publications (i.e., four of the six most cited publications). The initial study aggregated pooled data over a 3-year period to provide the first US prevalence estimates for 21 major defects and estimated the number of births affected by these defects each year in the United States (Canfield et al., 2006). Many of these publications reported prevalence stratified by maternal characteristics; for example, a higher prevalence of anencephaly, spina bifida without anencephaly, encephalocele, gastroschisis, and Down syndrome was observed among infants born to Hispanic compared

to non-Hispanic White women (Centers for Disease and Prevention, 2006). As a group, these publications facilitated comparison of prevalence estimates for several major defects side by side, examining temporal changes in prevalence (e.g., to confirm increases in the prevalence of gastroschisis in recent years; Mai et al., 2019), and comparison of estimates between programs with different surveillance methods (e.g., active versus passive case finding; Parker et al., 2010). In fact, a key theme across these publications has been the ability to show variation in prevalence estimates by ascertainment methodology and other key surveillance system characteristics (Mai et al., 2015; Parker et al., 2010), which has important implications for birth defects surveillance worldwide, such as better understanding the potential for under-ascertainment of certain defects.

3.3 | Descriptive epidemiology association analyses

Several NBDPN publications (using pooled data from 10 to 20 surveillance programs) focused on hypothesis-generating analysis of associations between different parental/case descriptive characteristics and risk for birth defects in offspring, such as reporting on prevalence ratio estimates across several maternal characteristics. Five publications used NBDPN data to describe the epidemiology of abdominal/gastrointestinal defects (Kapoor et al., 2019;Kirby et al., 2013; Marshall et al., 2015; Stallings et al., 2019) or limb/musculoskeletal defects (Parker et al., 2009).

NBDPN descriptive epidemiology publications have contributed important evidence toward confirming suspected associations between young maternal age and gastroschisis and increases in gastroschisis prevalence over time (Kirby et al., 2013; Stallings et al., 2019), a trend also reported in several countries throughout the world (Calderon et al., 2019; Castilla et al., 2008; Loane et al., 2007). NBDPN descriptive studies also helped to establish differences between the distinct epidemiological profiles of gastroschisis versus omphalocele, an abdominal wall defect that does not seem to have a similarly increasing prevalence over time (Kirby et al., 2013; Marshall et al., 2015; Stallings et al., 2019). In fact, given the potential for risk profiles for defects such as gastroschisis to change over time, NBDPN pooled data have allowed opportunities to conduct adequately-powered comparisons restricted to fairly narrow, recent time windows (e.g., births in 2012 and later) (Stallings et al., 2022). Of note, the large pooled samples have likely allowed for analysis of variables with rare categories that would have too sparse of cells to be considered in smaller datasets; for example, there were $N = 30$ case children with club foot and mothers with pregestational diabetes across pooled data from 10 surveillance systems (Parker et al., 2009).

3.4 | Risk factor association analyses

Ten NBDPN publications (using pooled data from 4 to 24 surveillance programs) evaluated associations for potential risk factors with orofacial cleft defects (Zhou et al., 2017), gastrointestinal defects (Jones et al., 2016), NTDs (Boulet et al., 2008; Williams et al., 2002, 2005), and a spectrum of defects (Canfield et al., 2005, 2014; Kirby et al., 2019; Liberman et al., 2022; Marengo et al., 2018). The most cited of these include four association studies that estimated the relative prevalence of NTDs before versus after fortification of enriched grain products in the United States with folic acid in 1998 (Boulet et al., 2008; Canfield et al., 2005; Williams et al., 2002, 2005), which is a major contribution and impact of NBDPN.

Collectively, these publications confirmed the effectiveness of the US folic acid fortification policy as a successful national public health intervention for birth defects prevention, as evidenced by a sustained reduction in the prevalence of spina bifida and anencephaly in particular, as well as other defects, to a lesser extent.

Other risk factors investigated included environmental factors, such as ambient fine particulate matter with aerodynamic diameter ≤2.5 microm (PM 2.5), an exposure which was associated with cleft palate in offspring (Zhou et al., 2017). Interpregnancy interval is another exposure that has been evaluated, and associations between short interpregnancy interval and gastroschisis as well as tetralogy of Fallot and cleft lip with or without cleft palate have been reported (Liberman et al., 2022). A racial/ethnic difference was reported for anotia/microtia, with a higher prevalence in offspring of American Indians/Alaska Native women compared to non-Hispanic White women (Canfield et al., 2014; Marengo et al., 2018). Finally, associations between delivery year and gastroschisis by maternal age and race/ethnicity strata (Jones et al., 2016) and between maternal nativity status and 27 select birth defects (Kirby et al., 2019) were reported. In aggregate, these risk factor publications were instrumental in delineating directions for further research examining the complex etiology of birth defects in the United States.

3.5 | Outcome association analyses

Given the lack of national population-based data for outcomes among individuals with major defects, NBDPN outcome association analyses have started to address this gap. Six publications (using pooled data from 6 to 15 surveillance programs) used NBDPN data to examine outcomes among neonates and infants with various birth defects. Most focused on neonatal, post-neonatal, and/or infant survival, including among infants with chromosomal abnormalities (Meyer et al., 2016), NTDs (Bol et al., 2006), or a spectrum of defects (Lopez et al., 2018; Wang et al., 2015). Areas of national concern include defining life expectancy and mortality, which is critical information for families and clinical practitioners, as well as identifying risk factors and disparities in survival. NBDPN publications have addressed these national needs by implicating gestational age as a risk factor for mortality among infants with trisomies 13 or 18 (Meyer et al., 2016), showing improved survival and decreased lesion severity in infants with spina bifida following folic acid fortification (Bol et al., 2006; Mai et al., 2022), and reporting on higher post-neonatal mortality risk among offspring of non-Hispanic Black and Hispanic women compared to non-Hispanic White women for several defects evaluated (Wang et al., 2015). Disparities were further highlighted by a publication that examined mortality risk by Hispanic ethnic subgroups, noting that survival among children with CHDs was lowest among the offspring of Mexican American women and highest among those of Cuban American women (Lopez et al., 2018). Beyond mortality, preterm birth has been evaluated, with neonates delivered preterm in the general population observed to be twice as likely to have at least one major defect compared to those born at term (Honein et al., 2009).

3.6 | Surveillance methods

NBDPN has been instrumental in furthering birth defects surveillance methodology for birth defects surveillance systems and best practices for surveillance and reporting. Eight

publications surveyed 31–54 surveillance programs about methodologies, barriers, and programmatic activities. Two publications assessed NTD recurrence prevention activities among surveillance systems in 2005 and 2015, respectively (Collins et al., 2009; Flood et al., 2016), highlighting that few systems conducted activities to prevent NTD recurrence (e.g., 9 of 44 in 2015). Other publications identified that few surveillance programs (7 of 34) have interstate data exchange agreements with other state surveillance programs (Cassell et al., 2007), and only a slight majority (20 out of 38) of programs conduct geocoding of maternal address (Wang et al., 2010). Furthermore, a self-administered data quality assessment tool was used to highlight similarities and differences for data completeness, timeliness, and accuracy across surveillance programs (Anderka et al., 2015). Additional surveys focused on other aspects of clinical review or public health practice activities among surveillance programs (Anderka et al., 2018; Lin et al., 2006; Mai et al., 2016).

Beyond this work, NBDPN publications have led to methodologic contributions that established recommendations for the role of clinicians in birth defects surveillance (Lin et al., 2006); collection, protection, and use of population-based birth defects surveillance data (Mai et al., 2007), and reporting of key metrics such as 95% confidence intervals for birth defects prevalence estimates (Correa-Villasenor et al., 2003). Many of these recommendations have now become the national standard for birth defects surveillance and reporting activities in the United States, and enable programs to self-assess, chart progress, and set clear aspirational targets for excellence.

4 | DISCUSSION

NBDPN publications have contributed to US birth defects surveillance and research across several domains by contributing gold-standard prevalence estimates, providing evidence about defect etiologies and outcomes among affected individuals, and evaluating the impact of public health practice of birth defects across the country. These projects produced several important datasets and facilitated collaborations across states.

Among the most cited NBDPN publications were those that provided US birth defects prevalence estimates, which can inform planning for interventions and resource needs. These publications also enabled systematic description of program data collection methodologies and current status of state surveillance efforts that are vital to understanding variation among surveillance programs, interpreting prevalence data, and understanding public health implications of birth defects in the United States. Some publications have been recognized with awards from professional societies; for instance, the Birth Defects Distinguished Scholar Award was awarded by the Society of Birth Defects Research and Prevention (formerly Teratology Society) to recognize authors for the importance, impact, and relevance of their work in the field of birth defects research (Canfield et al., 2006; Parker et al., 2010). Other highly cited NBDPN publications were those that helped establish the success of folic acid fortification policy as a US population-based birth strategy for the prevention of birth defects. These studies have been critical in informing ongoing efforts related to working toward folic acid fortification in other countries (Atta et al., 2016; Morris et al., 2021) with estimates suggesting that fortification in over 50 countries contributes toward NTD prevention in over 65,000 infants per year worldwide (Kancherla et al., 2021). NBDPN

is likely to remain a critical resource for assessing the success of future national initiatives for birth defects prevention.

Pooling data across states and leveraging the technical expertise of state programs are unique strengths of NBDPN publications. For example, having access to data for large numbers of case children allows researchers to analyze very rare birth defects and exposures that otherwise would not have sufficient numbers/statistical power within a single surveillance program. Aggregating data from multiple surveillance programs not only increases the size of the population but allows for better approximation of US population demographic characteristics, such as race/ethnicity. Additionally, NBDPN publications facilitate sharing of ideas, perspectives, and information that advance the field of birth defects research and surveillance.

Despite these strengths, results from these publications should be interpreted in consideration of several limitations. Potential differences in surveillance practices (e.g., case definitions, abstraction methods) across programs may contribute to differences in birth defects prevalence. Information on potential exposures and covariates was constrained to a small number of similar variables collected across states (typically basic demographic information collected from medical or vital records). Furthermore, some covariates could not be examined, such as population differences in individual states or differences in access to healthcare; thus, some degree of residual confounding may be present. Few studies (N = 3/21 association studies, Table 1) accounted for case children with chromosome abnormalities (e.g., excluding syndromic case children), likely due to limited data available, which is expected to potentially lead to bias in association analyses when the etiologies of syndromic and non-syndromic cases differ (Benjamin et al., 2022). Despite these limitations, however, findings from NBDPN publications provide critical clues about characteristics associated with major birth defects and provide a national snapshot of the impact of these conditions that could not otherwise be assessed.

Our assessment of NBDPN publications is somewhat limited as it does not fully describe the complex analytical designs used, especially those with secondary analyses or multiple goals. For example, our simple and hierarchical categorization framework may inadvertently imply a distinct line between prevalence and descriptive epidemiologic association analyses when some publications might not be so distinct. Our inclusion criteria likely eliminated several publications that did not explicitly mention NBDPN, but may have originated in part from prior NBDPN collaborations or predated the official formation of NBDPN (e.g., Kirby et al., 2000; Mason et al., 2005; Williams et al., 2002). Similarly, we did not include reports that were not published in peer-reviewed journals, though other NBDPN data uses include support of graduate student theses/dissertations, annual reports, and other program reports. However, recognizing the collective contribution of NBDPN publications as a group and considering the scope and focus of work to date may help prioritize and plan for future initiatives.

Possible directions for future work include increasing the number of state surveillance programs that contribute data to NBDPN publications, which may be assisted by developing processes to enable faster pooling of data to respond to emerging threats to mothers and

infants. For instance, administrative approval for each state to allow data to be stored centrally (e.g., in an ongoing data repository) may facilitate initiation of new project proposals and investigation of emerging research questions; the existing body of NBDPN publications represents only a small fraction of the scope of research questions that could be addressed to better understand the etiology and population-level impact of major birth defects. Additional direction includes standardizing analytic methods across projects (e.g., refined exclusion of syndromic cases and more frequent use of multi-variable modeling, when appropriate). However, while NBDPN projects can help develop and guide research questions, given limited information related to periconceptional maternal exposures in NBDPN, other data sources (e.g., genetic evaluations, maternal exposure assessments) may be helpful for assessing risk factors more in depth. Similarly, other data sources may be more helpful for assessing outcomes other than mortality throughout the life course (e.g., morbidities and health care utilization).

In summary, NBDPN publications have been critical for understanding the population-level implications of major birth defects in the United States. Future work will undoubtedly involve increasing efforts to broaden our understanding of the impact, causes, and consequences of these defects and increase our ability to improve the health and long-term outcomes of infants with birth defects. These studies may also inform hypothesis-testing research, thereby helping to serve as the groundwork for research delineating additional risk factors for birth defects in the United States.

DATA AVAILABILITY STATEMENT

Not applicable. This paper reviewed prior literature and did not assess other data.

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.

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TABLE 1

Characteristics Among All Published National Birth Defects Prevention Network (NBDPN) multi-state collaborative projects.

		Study characteristics				Multivariable analytic characteristics			Impact metrics	
Author and publication year	**Delivery years**	**Sites represented**	**Primary exposure of interest**	**Primary outcome of interest**	**Number of cases**	**Adjusted analysis[a]**	**Accounted for cases with chromosomal abnormalities**	**Accounted for isolated vs. multiple birth defects**	**Journal impact Factor[b]**	**Citations (N)[c]**
Prevalence analyses										
Individual defects										
Stallings et al., 2022[d]	2014–2018	19 States/ territories	N/A	12 CCHDs[e]	18,587	N/A	N/A	N/A	2.7	N/A
Stallings et al., 2022[d]	2013–2017	12 States/ territories	Maternal race/ ethnicity	Down syndrome	5836	N/A	N/A	N/A	2.7	N/A
Heinke et al., 2021[d]	2013–2017	25 States/ territories	N/A	Down syndrome	13,376	N/A	N/A	N/A	2.7	N/A
Stallings et al., 2018[d]	2011–2015	30 States/ territories	N/A	Eye and ear anomalies	5809	N/A	N/A	N/A	2.7	16
Lupo et al., 2017[d]	2010–2014	28 States	N/A	Gastrointestinal defects	Various	N/A	N/A	N/A	2.7	51
Cragan et al., 2016[d]	2009–2013	30 States/ territories	N/A	Congenital microcephaly	9678	N/A	N/A	N/A	2.1	45
Williams et al., 2015[f]	1999–2011	19 States	Folic acid	NTDs[g]	1326	N/A	N/A	N/A	35.3	339
Mai et al., 2014[d]	2007–2011	39 States	N/A	Orofacial clefts	N/A	N/A	N/A	N/A	2.1	83
Mai et al., 2013[d]	2006–2010	41 States	NA	Trisomy conditions	Various	NA	N/A	N/A	2.1	59
Mai et al., 2012[d]	2005–2009	34 States	N/A	CCHDs[e]	NR[h]	N/A	N/A	N/A	2.1	35
IPDTOC Working Group, 2011[i]	2000–2005	30 Countries	N/A	Cleft lip with or without cleft palate	7704	N/A	N/A	N/A	1.9	210
Spectrum of defects										
St. Louis et al., 2017[d]	1999–2007	11 States	Birth year	Select birth defects	Various	N/A	N/A	N/A	2.7	38
Mai et al., 2019[d]	2010–2014	39 States	Passive versus active case-finding	Select birth defects	Various	N/A	N/A	N/A	2.7	240

	Study characteristics					Multivariable analytic characteristics			Impact metrics	
Author and publication year	**Delivery years**	**Sites represented**	**Primary exposure of interest**	**Primary outcome of interest**	**Number of cases**	**Adjusted analysis[a]**	**Accounted for cases with chromosomal abnormalities**	**Accounted for isolated vs. multiple birth defects**	**Journal impact Factor[b]**	**Citations (N)[c]**
Mai et al., 2015[d]	2008–2012	38 States/ territories	Passive versus active case-finding	Select birth defects	Various	N/A	N/A	N/A	2.1	52
Parker et al., 2010[d]	2004–2006	30 States/ territories	Passive versus active case-finding	Select birth defects	Various	N/A	N/A	N/A	2.1	1,161
Canfield et al., 2006[f,j]	1999–2001	22 States	N/A	Select birth defects	Various	N/A	N/A	N/A	35.3/157.4	N/A
Canfield et al., 2006[d]	1999–2001	22 States	Passive versus active case-finding and race/ethnicity	Select birth defects	Various	N/A	N/A	N/A	2.1	462
Descriptive epidemiology association analyses										
Kapoor et al., 2019[d]	1999–2010	11 States	N/A	Infantile hypertrophic pyloric stenosis	29,554	Yes	No	Yes	2.7	13
Stallings et al., 2019[d]	2012–2016	20 States/ territories	N/A	Gastroschisis, omphalocele	5349 (gastroschisis) 2601 (omphalocele)	No	No	No	2.7	29
Marshall et al., 2015[k]	1995–2005	12 States	N/A	Omphalocele	2308	Yes	Yes	Yes	7.6	60
Kirby et al., 2013[k]	1995–2005	15 States	N/A	Gastroschisis	4713	Yes	No	No	7.6	98
Parker et al., 2009[d]	2001–2005	10 States/ territories	N/A	Clubfoot	6139	Yes	Yes	No	2.1	62
Risk factor association analyses										
Liberman et al., 2021[d]	2000–2009	9 States	Short and long interpregnancy intervals (IPIs)	Adverse birth outcomes/birth defects	Various	No	No	No	2.7	N/A
Kirby et al., 2019[d]	1999–2007	11 States	Maternal nativity status	27 select birth defects	Various	Yes	No	No	2.7	8
Marengo et al., 2018[d]	1999–2007	12 States	American Indian/Alaska Native race/ ethnicity	Select birth defects	Various	Yes	No	No	2.7	7
Zhou et al., 2017[l]	2001–2007	4 States	Maternal exposure to ozone and PM2.5	Orofacial clefts	7035	Yes	No	No	8.4	29
Jones et al., 2016[f]	1995–2012	14 States	Birth year	Gastroschisis	8866	No	No	No	35.3	103
Canfield et al., 2014[m]	1999–2007	12 States	Race/ethnicity	Select birth defects	Various	Yes	No	No	11.6	82

	Study characteristics					Multivariable analytic characteristics			Impact metrics	
Author and publication year	**Delivery years**	**Sites represented**	**Primary exposure of interest**	**Primary outcome of interest**	**Number of cases**	**Adjusted analysis[a]**	**Accounted for cases with chromosomal abnormalities**	**Accounted for isolated vs. multiple birth defects**	**Journal impact Factor[b]**	**Citations (N)[c]**
Boulet et al., 2008[d]	1999–2004	21 States/ territories	Birth year	Spina bifida and anencephaly	3311 (spina bifida), 2116 (anencephaly)	No	No	No	2.1	190
Canfield et al., 2005[d]	1995–2000	23 States/ territories	Folic acid fortification	Select birth defects	Various	No	No	No	2.1	163
Williams et al., 2015[f]	1995–2002	21 States/ territories	Folic acid fortification	Spina bifida and anencephaly	4468 (spina bifida), 2625 (anencephaly)	No	No	No	9.7	239
Williams et al., 2002[n]	1995–1999	24 States/ territories	Folic acid fortification	Spina bifida and anencephaly	5630	No	No	No	2.1	245
Outcome association analyses										
Survival										
Lopez et al., 2019[d]	1999–2007	12 States	Hispanic subgroups	Survival among infants with select birth defects	Various	Yes	No	No	2.7	6
Meyer et al., 2016[o]	1999–2007	9 States	N/A	Survival among infants with trisomy 13 or 18	693 (trisomy 13), 1113 (trisomy 18)	Yes	Yes	No	2.6	101
Wang et al., 2015[p]	1999–2007	12 States	Race/ethnicity	Survival among infants with select birth defects	Various	Yes	No	No	6.3	54
Bol et al., 2006[q]	1995–2001	15 States	Folic acid fortification	Survival among infants with NTDs[g]	2841 (spina bifida), 638 (encephalocele)	Yes	No	Yes	9.7	79
Other outcomes										
Mai et al., 2022[p]	1992–1996; 1999–2016	6 States	Folic acid fortification	Spina bifida lesion level	2593	Yes	No	No	6.3	N/A
Honein et al., 2009[r]	1995–2000	13 States	Preterm birth	Select birth defects	229,740	Yes	No	No	2.3	75
Surveillance methods										
Surveys of NBDPN surveillance program activities										
Anderka et al., 2018[d]	N/A	54 States/ territories	N/A	Public health response activities	N/A	N/A	N/A	N/A	2.7	0
Flood et al., 2016[d]	N/A	44 US jurisdictions/	N/A	NTD[e] recurrence	N/A	N/A	N/A	N/A	2.1	0

	Study characteristics					Multivariable analytic characteristics			Impact metrics	
Author and publication year	**Delivery years**	**Sites represented**	**Primary exposure of interest**	**Primary outcome of interest**	**Number of cases**	**Adjusted analysis[a]**	**Accounted for cases with chromosomal abnormalities**	**Accounted for isolated vs. multiple birth defects**	**Journal impact Factor[b]**	**Citations (N)[c]**
		Canadian provinces		prevention activities						
Mai et al., 2016[s]	N/A	43 States/ territories	N/A	Population-based defect surveillance practices	N/A	N/A	N/A	N/A	2.7	10
Anderka et al., 2015[t]	N/A	43 States/ territories	N/A	Data quality assessment	N/A	N/A	N/A	N/A	4.1	20
Wang et al., 2010[u]	N/A	39 States/ territories	N/A	Barriers to geocoding birth defect data	N/A	N/A	N/A	N/A	NR	3
Collins et al., 2009[d]	N/A	34 States/ territories	N/A	NTD[e] recurrence prevention activities	N/A	N/A	N/A	N/A	2.1	5
Cassell et al., 2007[d]	N/A	52 States/ territories	N/A	Interstate birth defects data exchange agreements	N/A	N/A	N/A	N/A	2.1	3
Lin et al., 2006[d]	N/A	31 States/ territories	N/A	Clinical review capacity	N/A	N/A	N/A	N/A	2.1	4
Other										
Lin et al., 2009[d]	N/A	N/A	N/A	Role of clinicians	N/A	N/A	N/A	N/A	2.1	7
Mai et al., 2007[d]	N/A	N/A	N/A	Collection and protection of data	N/A	N/A	N/A	N/A	2.1	8
Correa-Villasenor et al,, 2003[d]	N/A	N/A	N/A	Reporting confidence intervals	N/A	N/A	N/A	N/A	2.1	11

[a] Adjusted or stratified for at least one covariate.

[b] Impaet Factor reported by Journal Citation Reports (NR when not reported).

[c] Only reported for articles published before 2021.

[d] Published in *Birth Defects Research/Birth Defects Research Part A: Clinical and Molecular Teratology*.

[e] Critical congenital heart defects.

[f] Published in *MMWR Morbidity and Mortal Weekly Report*.

[g] Neural tube defects.

[h] Not reported.

[i] Published in *The Cleft Palate Craniofacial Journal*.

[j] Republished in *JAMA (total citations not available)*.

[k] Published in *Obstetrics & Gynecology*.

[l] Published in *Environmental Research*.

[m] Published in *American Journal of Public Health*.

[n] Published in *Teratology*.

[o] Published in *American Journal of Medical Genetics Part A*.

[p] Published in *Journal of Pediatrics*.

[q] Published in *Pediatrics*.

[r] Published in *Maternal Child Health Journal*.

[s] Published in *Journal of Public Health Management and Practice*.

[t] Published in *BMC Public Health*.

[u] Published in *Journal of Registry Management*.