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Using State and Provincial Surveillance Programs to Reduce Risk of Recurrence of Neural Tube Defects in the United States and Canada: A Missed Opportunity?

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Abstract

Background—Once a woman has had a fetus or infant affected with a neural tube defect (NTD), the risk of recurrence is approximately 3%. This risk can be significantly reduced by folic acid supplement consumption during the periconceptional period; however, this requires women at risk to be adequately informed about the appropriate dosage and timing of supplement intake before planning another pregnancy. As birth defects surveillance programs are tasked with identifying and documenting NTD-affected pregnancies and births, they are in a unique position to support recurrence prevention activities.

Methods—In 2015, we surveyed state and provincial birth defects surveillance programs to assess their NTD recurrence prevention activities. The online survey was sent to programs in 52 United States (U.S.) jurisdictions and all 13 provinces and territories in Canada. Findings were compared with a similar survey conducted in 2005 among U.S. programs.

Results—In 2015, of the 44 U.S. and Canadian surveillance programs that responded, only 9 programs (7 U.S. and 2 Canadian) reported currently having activities specifically directed toward preventing NTD recurrence. Compared with a 2005 survey of U.S. programs, the number of U.S. programs working on NTD recurrence prevention decreased by almost 50% (from 13 to 7 programs).

Conclusion—The number of birth defects surveillance programs with NTD recurrence prevention activities has decreased over the past decade due to a range of barriers, most notably a

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lack of resources. However, while some recurrence prevention activities require part-time staff, other activities could be accomplished using minimal resources.

Keywords

neural tube defect; recurrence; prevention; spina bifida; anencephaly; folic acid

Introduction

Consumption of supplemental folic acid has been shown to be effective in preventing the recurrence of neural tube defects (NTDs) (Grosse and Collins, 2007), especially those not part of a syndrome or other pattern of multiple anomalies. The most common forms of NTDs are anencephaly, spina bifida, and encephalocele, although rarer forms also occur. These birth defects arise during the first 28 days of gestation due to faulty closure of the embryonic neural tube (Sadler, 2012). NTDs that decline in frequency with increased consumption of folic acid, through food fortification or use of supplements that contain folic acid, are considered to be folate sensitive and thus potentially preventable. In 1991, the U.S. Centers for Disease Control and Prevention (CDC) issued guidelines for preventing the recurrence of NTD (CDC, 1991, 1992). These guidelines recommend that women with a previous affected pregnancy and *planning* a subsequent pregnancy speak with their doctors about consuming 4.0 milligrams of folic acid daily, beginning a minimum of 1 month before conception and continuing through the first 3 months of pregnancy. These guidelines are referenced by other U.S. public health and medical organizations (ACOG, 2003; USPSTF, 2009; Toriello, 2011; Zolotor and Carlough, 2014).

Recommendations by the Society of Obstetricians and Gynecologists of Canada and Health Canada are similar, but advise that women begin consuming 4.0 milligrams folic acid at least 3 months before conception (Van Allen et al., 2002; Wilson et al., 2015). For women at elevated risk but *not planning* a pregnancy, CDC (2002) recommends 0.4 mg of folic acid per day, and Health Canada (Van Allen et al., 2002) recommends 1.0 mg folic acid daily in a multivitamin-multimineral supplement. Previous studies reported that women with a history of a NTD-affected pregnancy were under-informed about the preventive role of folic acid (Rinsky-Eng and Miller, 2002). Furthermore, recent evidence shows that many women remain inadequately supplemented. This would indicate that many either remain unaware of the recommendations to take folic acid or fail to initiate usage at the appropriate time (Arth et al., 2015).

With few exceptions (Felkner et al., 2005; Bupp et al., 2015), the prevalence of recurrent NTD in the United States has not been well published or studied in the era after the initiation of folic acid fortification. The risk factors underlying the *recurrence* of NTD are only partially understood, and may differ from the factors associated with primary *occurrence* (Mitchell, 2005; Agopian et al., 2013). Recurrent cases may result from inadequate consumption of folic acid or are due to mechanisms unrelated to folic acid supplementation or fortification, for example, those where the NTD is one component of a chromosomal syndrome or a single gene disorder. Nonetheless, it has been shown that periconceptional folic acid consumption reduces the risk considerably, especially for isolated spina bifida.

Felkner et al. (2005) detected no recurrent NTD among 130 women receiving folic acid supplementation, and Bupp et al. (2015) reported a 0.4% NTD recurrence risk among mothers who took folic acid, and an 8.5% recurrence risk among mothers not taking folic acid.

As part of their routine functions, birth defects surveillance programs not only record the occurrence of NTD-affected pregnancies, but some also record mothers' contact information. These data could be used to identify mothers and families who could benefit from educational materials and messages. As such, surveillance programs might be well-positioned to use their registries to educate high risk mothers about the role of folic acid in preventing the recurrence of an NTD.

In a 2005 survey of state birth defects surveillance programs, Collins et al. (2009) contacted 34 states and Puerto Rico and identified 13 programs with current activities to prevent the recurrence of an NTD. In this study, we updated the 2005 survey concerning NTD recurrence prevention activities in both the United States and Canada to determine the number of surveillance programs in 2015 engaged in NTD recurrence prevention, the educational activities conducted, the resources dedicated to this prevention topic, and the obstacles identified in implementing NTD recurrence prevention activities.

Materials and Methods

In January 2015, the NTD Surveillance and Folic Acid Education Committee of the National Birth Defects Prevention Network (NBDPN) conducted a survey of state and provincial birth defects surveillance programs. Similar to the 2005 version, this current survey was created and conducted online using SurveyMonkey (SurveyMonkey.com; Portland, OR). The Committee invited the programs in the jurisdictions of all 50 U.S. states, the District of Columbia, Puerto Rico, and all 13 provinces and territories in Canada to participate in the survey. The Committee emailed an invitation to the birth defects surveillance point-of-contact in each state, province, or territory. Email reminders were sent from February to April 2015.

As in 2005, the new survey asked programs to indicate the status of state/provincial activities related to NTD recurrence prevention (none, previously had or planning, current activity). If the initial point-of-contact was unable to provide this information, the Committee requested that they forward the survey to a knowledgeable respondent. For programs denoting current activities, the Committee considered the response valid if activities included one or more of the following activity components: (a) provided educational materials for NTD recurrence prevention at events, through community advocacy groups, or by means of social media; (b) conducted outreach to clinics that, in turn, offered recurrence prevention information to high risk women; or (c) used registry data to contact women who had an affected pregnancy, directly or indirectly (e.g., by means of a health care practitioner).

For programs with current activities, responses concerning activity components, including the proportion of families of NTD cases they contacted, whether families are contacted in cases of fetal death, assessment of the mothers' understanding of NTD recurrence risk, the

interval from birth to contact, the method (e.g., in-person, mail, clinic) used for the contact, and whether the program devotes staff to recurrence prevention activities were assessed. For the programs that either never had activities or did not have current ones, the survey asked about the barriers they encountered in trying to conduct recurrence prevention activities. Analyses consisted of tabulations of categorical responses and review of open-ended text responses.

Results

A total of 35 (67.3%) U.S. jurisdictions and nine (69.2%) Canadian provinces or territories responded to the survey (Fig. 1). Current NTD recurrence prevention activities were reported from 15 programs in 12 U.S. states, Puerto Rico, and two Canadian provinces. Upon review of the 15 responses, six programs did not have targeted recurrence prevention components. For example, six programs reported that their activities consisted solely of disseminating general folic acid materials at a Birth Defects Prevention Month event held annually each January. As these materials were not recurrence specific, these six programs were removed from further evaluation. This left 38 programs for analysis, 29 respondents with no current program plus the 9 respondents with NTD recurrence prevention activities. Of the 29 respondents with no current program, 14 programs either had activities in the past, including 1 Canadian program, or reported planning future initiatives. The remaining 15 programs, 9 in the United States and 6 in Canada, reported no recurrence prevention activities involving their birth defects surveillance systems. Overwhelmingly, these surveillance programs cited a lack of funds and staff as the reason for this lack of activity (Fig. 2). These programs without recurrence prevention activities also noted a lack of expertise, low priority, and concerns about privacy among the reasons for not implementing activities. These responses are similar to those reported in the previous 2005 survey.

CHARACTERISTICS OF CURRENT RECURRENCE PROGRAMS (*N* = 9)

The following results are restricted to the nine programs (seven U.S. and two Canadian) that reported specific recurrence prevention activities. Among the range of approaches used, seven programs provided educational materials for NTD recurrence prevention in one of the following ways: at events, through community advocacy groups, or by means of social media. Five programs conducted outreach to clinics that provide recurrence prevention information to high risk women. Three programs implemented other activities, specifically, conducting maternal interviews, providing continuing medical education and/or reproductive guidelines to health care providers, and using local health department staff to deliver the messages to mothers.

The survey sought information pertaining to NTD case ascertainment. Four of the nine programs used active case-finding methods (intense staff engagement in finding cases), one conducted only passive case-finding (program receives case information from data sources), and four combined active and passive ascertainment. Due to the variable number of births in their catchment areas, the range of NTD cases ascertained by these programs ranged from zero to 170. Of the current programs, eight could verify the NTD diagnosis with medical records review, and three of these also received physician confirmation. The one remaining

program did not verify the reported diagnosis. In cases of adoption, two programs attempted to contact the birth mother, but three programs excluded adoptions. The remaining respondents did not disclose how adoptions are handled within their program. Eight of the nine programs reported the ability to determine whether an ascertained NTD case can be classified as an NTD recurrence for that mother.

CONTACTING THE MOTHER

The nine current programs reported contacting, on average, 76% of case mothers (with contact ranging from 10% to 100%). While the programs focused on contacting women who delivered a live birth, four programs indicated that they also contacted women who had an affected pregnancy resulting in a non-live birth. Six of the nine programs used records from a confidential registry to contact case mothers directly, and two programs used local health departments to contact mothers. The various methods used to contact mothers with a child or pregnancy affected by an NTD are shown in Table 1, with in-person contact being the most common approach. With respect to the type of professional contacting the women, the most common option selected was a genetic counselor/social worker, or a nurse. The typical interval following birth in which contact was made ranged from immediately following hospital discharge to within 1 year of birth. On average, programs needed 4 months following delivery to reach the mothers, but some programs were unable to report the actual time.

EDUCATIONAL MATERIALS AND THEIR DISSEMINATION TO FAMILIES

Current programs used a variety of methods and educational materials to disseminate information about reducing the risk for NTD recurrence. Six of the programs used brochures and fact sheets developed by CDC or produced by the program itself. Brochures/fact sheets developed by the March of Dimes Foundation, the NBDPN, and the Spina Bifida Association of America were slightly less common, with three of the programs using materials from these groups. One program used materials from another source and one program used a video as the educational media. Programs most frequently disseminated educational materials by mail (five programs), in person (four programs), or at a clinic (four programs). Two programs used family or case physicians to distribute materials; one program disseminated materials at targeted gatherings and events for persons with spina bifida. All nine programs recommended that mothers take 4.0 mg of folic acid for NTD recurrence prevention. No program mentioned educating women about NTD risk factors unrelated to folic acid.

PROFESSIONAL EDUCATION COMPONENT

Two of the programs included a component targeting providers and health professionals for professional education. Both programs targeted the same groups: the mother’s health care professional, family practitioners, and midwives.

RESOURCES, EFFECTIVENESS, EVALUATION, AND BARRIERS

Only one of the nine programs devoted a portion of time from both a contractor and full-time employees toward NTD recurrence prevention. The remaining programs dedicated

minimal staff time to the activities. While six programs (67%) identified their activities as being cost effective because their interventions were not resource intensive, one said their activities were not cost effective due to the high percentage of unplanned pregnancies (two programs did not respond). Most of the programs (78%) said they evaluated or were beginning to evaluate their activities. Of the nine current programs, five identified barriers or issues they have encountered. Two programs cited staffing limitations and one program each identified lack of funds, small numbers of cases for analysis, and challenges in obtaining the correct maternal contact information as barriers for conducting program evaluation.

Discussion

This survey inquired about NTD recurrence prevention activities since 2005, and expanded the focus from the United States to include Canadian programs. However, only 9 programs in 44 responding jurisdictions (7 in the United States, 2 in Canada) were conducting recurrence prevention activities in early 2015. Compared with the Collins et al. (2009) survey conducted in 2005, the number of programs conducting NTD recurrence prevention activities in the United States decreased by almost half (from 13 to 7 programs).

Survey response rates from the United States (67.3%) and Canada (69.2%) were similar, as were the proportions of programs that identified current activities (20.0% and 22.2%). Differences between U.S. and Canadian surveillance programs are apparent, with many U.S. jurisdictions having well established and long-standing birth defects registries. Few Canadian states or territories have such surveillance programs (Alberta being a notable exception), although there is an initiative under way to reestablish registries that had been discontinued or to implement new ones (Lowry and León, 2013).

The survey results identified opportunities to build and strengthen the core activities of birth defect surveillance programs. Our survey inquired about potential NTD recurrence prevention activities to reach mothers. All nine current programs used one or more of these activities. To expand the number of participating programs, these activities could serve as examples for other programs seeking to launch or expand recurrence activities.

The main barriers cited by both programs with and without recurrence prevention activities were inadequate funding and under-staffing. While more resources are needed for such initiatives, especially given their demonstrated cost-effectiveness (Grosse et al., 2008), it is acknowledged that many registries are being asked to do more with less. The following recurrence prevention activities are offered as suggestions for activities that might require only part-time staff or minimal resources: (a) Provide education about recurrence prevention at events attended by families of those affected by an NTD; (b) Partner with community groups (e.g., Spina Bifida Association) to deliver educational messages about recurrence prevention; (c) Include recurrence prevention messages in social media that specifically target mothers at high-risk; (d) Conduct outreach to high-risk women with information about recurrence prevention in a variety of clinical settings (e.g., obstetrics and gynecology, spina bifida clinic, genetics as well as family practice, general pediatrics); (e) Use information from the birth defects registry to contact families directly, such as via their healthcare provider or via a local health agency.

Efforts to use interventions are challenged also by the rarity of NTD recurrence. Surveillance programs should be able to present the magnitude of recurrent cases of NTD in their jurisdictions. Future surveys could ask about the ability of surveillance programs to publish their rate of recurrent NTD cases.

While our study shows that most birth defects surveillance programs are not engaged in recurrence prevention activities, opportunities for these programs to become engaged exist. First, there is a need for mothers with a history of NTD to be informed, educated, and empowered about the role of folic acid in preventing NTD in subsequent pregnancies. Because surveillance programs receive information about mothers who have had NTD-affected pregnancies, they may be in a unique position to help educate these women about recurrence risk before a subsequent pregnancy. Programs can explore and collaborate with interested partners to target the education messages. Second, there is also a need to link these mothers to health care providers (physicians, nutritionists, nurses, and others) who can reinforce this important intervention about recurrence risk.

Of additional importance is for healthcare professionals to be adequately informed. Several studies document that many healthcare professionals are lacking detailed and accurate information concerning the 4.0 milligram prescription dosage and timing of folic acid supplementation (Hauser et al., 2004; Abu-Hammad et al., 2008; Aggarwal et al., 2010). Our survey documented little to no attempt by surveillance programs to provide education to health professionals and this appears to be a worthwhile initiative to consider in the future. Also, seeking mothers' feedback concerning the recurrence prevention programs might add valuable perspective about the educational approaches.

With the emergence of interest in NTD risk factors other than folic acid, such as obesity and Hispanic ethnicity (Mitchell, 2005; Agopian et al., 2013), future surveys could assess whether jurisdictions are including these additional factors in their educational programs. Additional risk factors may be addressed by programs that promote optimal preconception health, healthy diet, exercise, and weight before and during pregnancy, and overall women's health.

STRENGTHS AND WEAKNESSES

A strength of this study was that it included all U.S. programs and its extension to Canadian provinces and territories. Also, given the paucity of information on this topic, this study presented useful data to guide public health interventions to help reduce NTD recurrence risk.

The study had some weaknesses. Because the survey was self-administered, six programs misclassified their recurrence prevention effort and had to be removed from the analysis. Also, we were not able to collect information in those areas that did not return a survey (approximately 65% of those contacted responded). However, because several the nonrespondents were from states without a birth defects program, we would expect that most of them had no recurrence prevention activities. Finally, we assumed that the programs had knowledge of their own jurisdiction's recurrence prevention activities or of similar activities conducted by other units in their jurisdiction.

Summary

Although the United States and Canada have been pioneers in the establishment of folic acid food fortification programs that have had a major impact on NTD occurrence in both countries (Mills and Signore, 2004; De Wals et al., 2007), women remain at risk either because of inadequate folic acid intake or other causes. In particular, women who had an affected pregnancy are at greatly increased risk of recurrence. However, the results of this survey indicate that birth defects surveillance programs have not progressed since 2005 in their promotion and dissemination of NTD recurrence prevention messages and that significant barriers remain in using these entities to assure the delivery of this crucial information to mothers with NTD-affected pregnancies and the health professionals who care for them. Individual programs need to determine the best and most efficient ways to ensure that accurate and timely information about folic acid supplementation in the periconceptional period is provided to mothers with a previously affected pregnancy.

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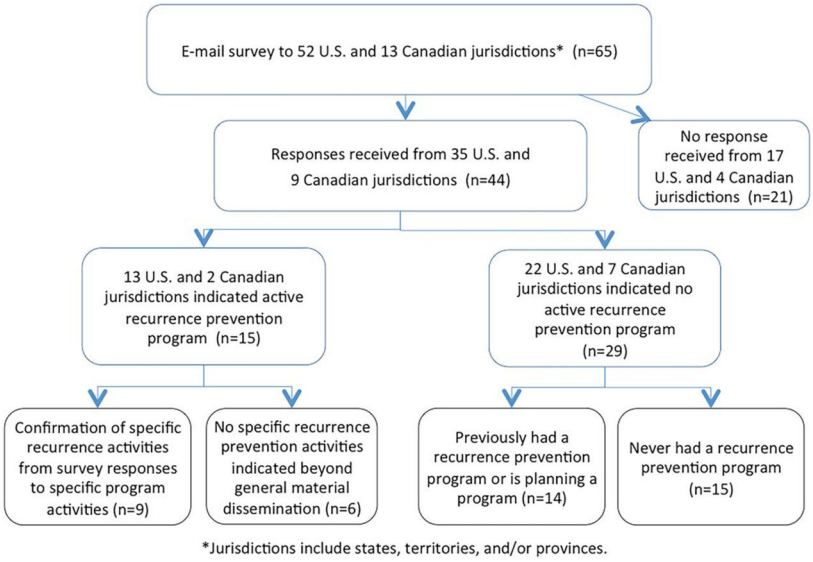

FIGURE 1.
Survey respondents from United States (U.S.) and Canadian jurisdictions.

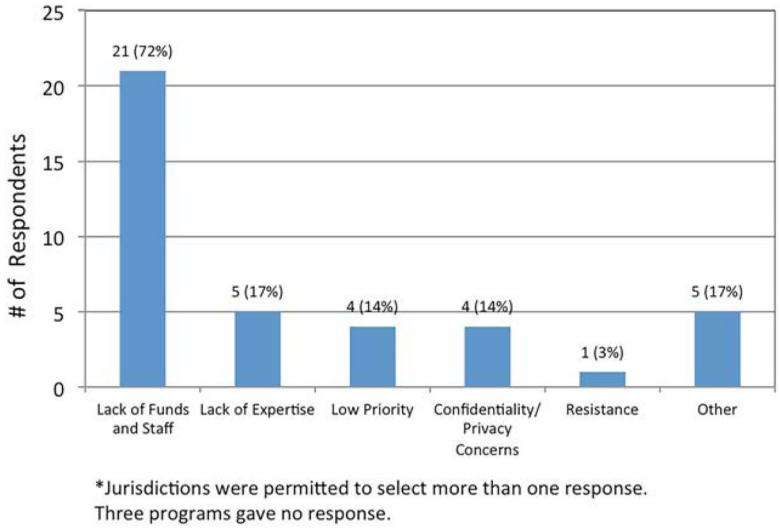

FIGURE 2.

Barriers cited for jurisdictions with no active neural tube defects recurrence prevention program (n = 29).

TABLE 1

Methods and Program Staff for Currently Active Recurrence Prevention Programs (n = 9) That Contacted Mothers Who Previously Had a Child or Pregnancy with a Neural Tube Defect

Program contact method	***n***	**% of all programs**
In person	5	56
Over the phone	4	44
At a clinic	3	33
In writing/by mail	3	33
Through physician of case/family	2	22
None	1	11
Type of program staff		
Genetic counselor/social worker	3	33
Nurse	3	33
Physician of case/family	2	22
Public health worker or administrator	2	22
Parent consultant	1	11

Respondents were permitted to select more than one option.