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Cross-Sectional Survey on Newborn Screening in Wisconsin Amish and Mennonite Communities

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Abstract

Old order Amish and Mennonites, or Plain populations, are a growing minority in North America with unique health care delivery and access challenges coupled with higher frequencies of genetic disorders. The objective of this study was to determine newborn screening use and attitudes from western Wisconsin Plain communities. A cross-sectional survey, with an overall response rate of 25%, provided data representing 2,010 children. In households with children (n=297), the rate of newborn screening was 74% and all children were screened in 40% of these households. Lack of access to testing was the most common reason for not screening all children and parental age was inversely associated with testing. The majority of respondents reported some or more knowledge of screening, viewed screening as important, and had access to screening in their communities. Households with children who had never received newborn screening (26%) reported lower frequencies of favorable responses in all categories compared to households that had at least one child screened. The difference in access to newborn screening was less marked between the groups compared to differences on knowledge and consideration of its importance. Moreover, 55% of households who had never screened any of their children reported being unlikely or unsure of screening any future children. A focus on improving access to newborn screening alongside establishing approaches to change parental perceptions on the importance of newborn screening is necessary for increasing newborn screening in these Plain communities.

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Introduction

Newborn screening is needed to identify disorders that can potentially cause severe disabilities or death, are treatable, and are not readily apparent at birth (1). While newborn screening is important for all infants, it is particularly important in populations with known founder effects, a wide variety of inheritable genetic disorders, and carrier rates higher than the general population (2). The Plain people of North America are such a population and include the Old Order Amish and Mennonite groups. Plain people belong to conservative Christian sects descended from small groups of immigrants who fled Europe beginning in the 17th century to avoid religious persecution (2). Plain people live in closed, well-defined communities with a lifestyle that separates them from mainstream society. Marriage outside of the community is prohibited and new converts to the faith are uncommon. Thus, there is frequently a high degree of genetic relatedness between parents. Over time, genetic drift of founder mutations has resulted in a higher prevalence of certain genetic disorders compared to the general population. These include genetic disorders that are part of the recommended uniform newborn screening panel such as phenylketonuria (PKU), maple syrup urine disease (MSUD), and severe combined immunodeficiency (SCID) (2).

The Amish population in North America is located in thirty U.S. states and the Canadian province of Ontario with an estimated population at 290,100 in 2014 (3). Amish women have an average 8.3 live births (4) and the Amish population size is estimated to double every 21.25 years in North America (5). Wisconsin has the fourth largest Amish population in North America with forty-two settlements and approximately 17,025 individuals (5). These Plain communities are located throughout numerous counties of western Wisconsin.

In Wisconsin, approximately 2% of annual births occur in out-of-hospital settings and the majority of these births occur in Plain communities (6). While our clinical experiences and published reports using vital records data suggest low newborn screening rates among Plain communities in Wisconsin, formal evaluation of newborn screening practices in Plain populations has not been undertaken (7, 8). In general, Plain people are not opposed to standard medical care and technologies. However, preventive care is not widely embraced by the Plain culture. For example, a survey of Old Order Amish communities in Ohio revealed that their low immunization rates arose from fears surrounding immunizations, rather than poor access (9). More accurate newborn screening information from Plain communities and survey of Plain people's views on newborn screening would likely lead to improved outreach activities and educational materials aimed increasing newborn screening rates in these communities. Herein we report results from a Plain community survey designed to determine newborn screening practices, knowledge, and views in western Wisconsin Plain communities.

Materials and Methods

Instrument

We adapted a survey from the Community Health Clinic in Topeka, Indiana with input from Wisconsin Plain community members (10). The paper survey consisted of 37 questions. The question formats included Likert rating scales, yes/no, open-ended, and multiple choice. The University of Wisconsin Health Sciences IRB determined this study was not human subjects research because the primary intent was quality improvement.

Distribution

Survey distribution to communities in western Wisconsin used a variety of approaches depending on the needs and requests of each community. Our distribution was focused on 1) communities in Vernon and Monroe counties suspected to have low utilization of newborn screening, 2) Old Order Mennonite households in northwestern Wisconsin, and 3) other Amish communities outside of Vernon and Monroe counties. Old Order Amish and Mennonite community members who engaged in advisory boards and community event planning established and organized by members of our team served as liaisons to community households for survey distribution. Cultural reasons precluded use of electronic or phone technology for survey distribution. Likewise, mass mailings were judged to be impersonal with a high likelihood of poor participation, in addition to uncertainty about the accuracy of existing community directories. In general, distribution involved either hand-delivery or completion of the survey in-person.

Families were encouraged to complete the survey whether or not they had a family member with a special health care need or any familiarity with newborn screening. The hand-delivered surveys were distributed by a Plain individual within their district, midwives, public health nurses, or health care providers. In-person surveys were obtained during a local Plain auction event, outreach clinics, or community meetings organized by members of our team aimed at improving health care delivery for Plain communities in Wisconsin. The hand-delivered surveys included an informational letter with instructions, the survey, and a self-addressed prepaid return envelope. Households were instructed to complete only one survey. After receipt of a survey, families received \$5 financial compensation.

Database

The information received from the surveys between August, 2014 to March 1, 2015 were entered into a secure electronic database developed using REDCap (Research Electronic Data Capture).

Statistical Analysis

Descriptive statistics, such as means, counts, and percentages were exported from the REDCap database. Percentage values were rounded to the nearest whole number. Student t-test (two-sided) was used for age comparisons. Chi-squared (χ^2) tests and Fisher's Exact Tests examined relationships between categorical variables. SAS version 9.4 and R version 3.1.1 software packages were used for statistical analysis. A p value 0.05 was considered statistically significant.

Results

Response Rate and Demographics

Three hundred fifteen of the 1,239 delivered surveys were completed and returned to the survey team for an overall response rate of 25%. The respondent demographics are shown in Table 1. For the Amish respondents (n=269), 90% resided in our target areas of Vernon and Monroe counties. Amish respondents outnumbered Mennonite respondents 6:1.

Newborn Screening Practices

To assess historical newborn screening practices in Wisconsin Plain communities, all households were asked if any of their children ever received a newborn screen. 74% households reported receiving newborn screening for at least one of their children, and 40% of those reported that all of their children were screened. A significantly higher proportion of Old Order Mennonite households received newborn screening compared to Old Order Amish households (93% vs. 70%, $p = 0.002$). Households that reported either any or all children received newborn screening had a significantly younger parental age compared to households that reported never receiving newborn screening for any child (mean maternal 35.3 vs 48.2 years, $p < 0.001$; mean paternal age 36.4 vs. 48.7 years, $p < 0.001$, respectively).

Reasons for Lack of Newborn Screening

Households with child were asked to indicate the reason(s) for not having a child receive newborn screening. Seventy percent of surveyed households with children had one or more children that did not receive newborn screening. The most common reasons were “I did not know about newborn screening” (32%) and “Newborn screening was not offered by my health care provider” (23%). Other concerns regarding beliefs, cost, travel, privacy, or follow-up testing and care collectively comprised 16% of household responses to this question. “Other” was listed as a reason for 21% of these household responses. A variety of reasons were provided in the “other” write-in section (Table 2). Multiple responses could be provided and families that had all of their children screened selected the “I had all of my children screened” response.

Newborn screening community knowledge

Knowledge of newborn screening was assessed by asking, “How much do you know about newborn screening?” The majority of households reported having “some” or “a lot” of knowledge about newborn screening (Figure 1A). There was not a statistically significant difference in reported knowledge between Amish and Mennonite households. For households with knowledge of newborn screening, the most commonly identified resource for information was a midwife (64%). Mennonites households were more likely to learn about newborn screening from a midwife than were Amish households (84% and 57% respectively; $p = 0.001$). The second most common information resource listed by Amish households was a family member (18%).

Newborn screening community views and access

The majority of households reported newborn screening was “important” or “very important” in preventing death and disability for their family (Figure 1B). Mennonites were significantly more likely to regard newborn screening as “important” or “very important” than were Amish (87% and 72% respectively, $p = 0.043$).

Most households reported favorable views of newborn screening within their communities. The majority of households reported newborn screening is either “encouraged” or “many families do it” in their community (Figure 1C). A higher proportion of Mennonite households reported “many families do it” compared to Amish households, though the difference was not statistically significant (54% vs 38%, $p = 0.056$).

To determine community access to newborn screening, households were asked to indicate their level of agreement with the following statement, “I have access to newborn screening”. The majority of households either “agreed” or “strongly agreed” they had access to newborn screening (Figure 1D). Reported access to newborn screening was significantly higher in Mennonite communities compared to Amish (95% vs. 79%; $p = 0.012$).

Associations between knowledge, views, and access with newborn screening practices

Responses to questions regarding knowledge, views, and access to newborn screening were stratified between households that ever received newborn screening for any or all children or that never received newborn screening for any of their children. Households that ever had newborn screening reported a higher frequency of favorable responses in all categories compared to households that never had newborn screening (Figure 2). Households that reported “some” or more knowledge about newborn screening, viewed newborn screening as “very important” or “important” in preventing death and disability, and had access to newborn screening were statistically more likely to have received newborn screening compared to households that never had newborn screening for a child ($p < 0.001$, χ^2 analysis for each variable [i.e. knowledge, views, access]).

Community attitudes on newborn screening for future children

To assess possible future newborn screening practices, households were asked how likely they would be to have newborn screening with a future birth. Collectively, the majority of households reported they would be “very likely” (53%), “likely” (17%), or “somewhat likely” (7%), to have newborn screening with a future birth. Fourteen percent of households reported they would be “unlikely” to have newborn screening in the future and 9% households felt “unsure.”

When responses were stratified based on reported historical newborn screening pattern, households that never had newborn screening for any of their children reported a higher frequency of “unlikely” or “unsure” responses about future newborn screening (Figure 3, $p < 0.001$, χ^2 analysis). No significant difference was found in age of the parents between those who reported the likelihood of having newborn screening for possible future children as “very likely” or “likely” compared to those who responded “unlikely.” A significantly

higher proportion of Mennonite households would have newborn screening with possible future children versus Amish households (95% vs 81%, $p = 0.021$).

Discussion

In this study, we sought to improve our understanding of newborn screening practices and provide insights into newborn screening knowledge, views, and access in Plain communities of western Wisconsin. Our findings are the first to address this issue in Plain communities. Despite being small minorities, Old Order Amish and Mennonites have settlements throughout North America with increasing population growth rates and our findings in western Wisconsin likely have implications for the other Plain communities throughout North America. Our results suggest merely providing access to newborn screening is not sufficient and improved approaches to change parental perceptions of the importance of newborn screening are needed.

In our survey respondents, 74% of households with children received newborn screening for at least one of their children, but the overall newborn screening rate is lower because only 40% of those respondents reported screening all their children. In comparison, the overall newborn screening rate for newborns in Wisconsin is approximately 99% (11–13). The reasons provided for not obtaining newborn screening by households varied; the most commonly reported reason was a lack of awareness of newborn screening. The younger mean parental age for families reporting newborn screening for a child may imply that awareness and access to newborn screening has improved over time. This is further supported by the high frequency of affirmative responses on the question about access to newborn screening.

The responses from households that never received newborn screening for any of their children are a valuable subset for informing ongoing efforts to improve newborn screening utilization in Plain communities. This subset reported less favorable responses in knowledge, importance, and access to newborn screening compared to households that ever had received newborn screening. The differences between “ever screened” versus “never screened” households in these responses suggest knowledge and views on its importance impact household perspectives on newborn screening more than poor access does. The majority of this respondent subset also reported being unlikely or unsure about receiving newborn screening in the future. As these relationships were not attributable to age (data not shown), continued efforts to increase newborn screening access along with approaches to provide culturally appropriate newborn screening education with an emphasis on the importance of newborn screening could make a difference for future children in these communities.

Our survey distribution was primarily focused on counties in Wisconsin with perceived low newborn screening use. Survey responses were also obtained from Mennonite communities located outside these targeted counties. Even though this sample size was smaller, the Mennonite respondents provided more favorable responses on knowledge, views, and access compared to Amish respondents in all newborn screening parameters surveyed. This is associated with greater utilization of newborn screening by Mennonite respondents. The

reasons for these differences are not readily apparent from our survey responses and were not related to age differences between the two sects (data not shown). One possible difference may be the source of information about newborn screening since most Mennonites received their knowledge from a midwife. Another possible difference may be broader community support for newborn screening in Mennonite communities. Efforts are ongoing to increase our Mennonite sample size and gather information from health care providers in these communities to better understand key differences between these sects related to newborn screening.

There are several limitations to our survey. Since distribution was focused on counties with perceived low newborn screening use, survey responses may be limited or biased. The results may represent select Amish community views and practices not shared with other Amish communities in Wisconsin and elsewhere in North America. For example, newborn screening views and practices may have skewed household completion of the survey. There may be potential cultural limitations secondary to the manner in which survey questions were written as suggested by write-in responses listed in Table 2. Despite these limitations, the data gathered from our cross-sectional survey provide previously unknown information about newborn screening practices and attitudes in Plain communities and thus will serve to improve ongoing activities to increase newborn screening use.

Access to confirmatory testing and follow up care with appropriate specialists for confirmed genetic disorders is an important aspect of promoting newborn screening (14). Provision of these critical aspects of newborn screening have notable challenges in Plain communities secondary to distance from tertiary care centers and cultural refusal of traditional health insurance. To this end, our ongoing outreach activities have coupled newborn screening education with health care models that provide culturally-appropriate care through partnership with local community providers and development of affordable genetic testing. This approach should be considered by others seeking to improve newborn screening in Plain communities.

Conclusion

Historical newborn screening practices appear to have improved over time in western Wisconsin Plain communities, but are considerably lower than the general population. Lack of awareness and access appear to be important barriers to newborn screening in the past that are being overcome. The majority of surveyed Plain households in western Wisconsin have knowledge, favorable views of, and access to newborn screening. However, approximately 23% of survey respondents with children remain unlikely or unsure of having newborn screening for any future children. In addition to ensuring access to timely newborn screening for all infants, newborn screening outreach efforts in Wisconsin Plain communities and elsewhere should address misperceptions about newborn screening and emphasize its importance in preventing death and disability. Based on our survey results, midwives are a valuable resource for disseminating this information in an effective manner.

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Abbreviations

NBS	newborn screening
REDCap	Research Electronic Data Capture
Amish	Old Order Amish
Mennonite	Old Order Mennonite

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Figure 1. Newborn Screening Community Knowledge, Views, and Access

A. Knowledge of NBS, n=296. **B.** Views on importance of NBS, n=283. **C.** Community views, n=282. Note: respondents were instructed to circle all that apply. **D.** Access to NBS, n=244. Abbreviations: NBS=newborn screening.

Figure 2. Favorable Newborn Screening Responses and Newborn screening practice

Households were stratified by ever receiving newborn screening for any or all of their children (NBS Ever, n=171 with all data points) and never receiving newborn screening for any of their children (NBS Never, n=56 with all data points). Favorable responses were categorized as follows: Knowledge about newborn screening: “a lot” and “some” were combined. Importance of newborn screening: “very important” and “important” were combined. Access to newborn screening: “strongly agree” and “agree” were combined. Abbreviations: NBS=newborn screening.

Figure 3. Future Newborn Screening Practice

Households were stratified by ever receiving newborn screening for any or all of their children (NBS Ever, n=200 with all data points) and never receiving newborn screening for any of their children (NBS Never, n=67 with all data points). Abbreviations: NBS=newborn screening.

Table 1

Demographics of Survey Respondents

Total Households	315
Old Order Amish	269 (86%)
Old Order Mennonite	44 (14%)
Married	309 (99%)
Have children	297 (94%)
Average number of children per household, [mean (95%CI), n=294]	6.8 (6.4–7.3)
Average household size, [mean (95%CI), n=314]	6.6 (6.3–7)
Total children	2,010

Table 2

Write in Responses for Lack of Newborn Screening

Write in answer	Number of surveys
We had home births	6
Did not feel it was necessary	6
Baby seemed healthy and normal	5
It was not available at the time my children were born	4
Newborn screening was not offered by provider	3
Our family and all our children are healthy	3
Did not realize its importance at the time	3
We did not know about it at the time/We do not know enough about it now	3
It was not encouraged at the time	3
Hearing and/or heart screening were not offered at the time	3
We did the PKU test	3
Failure to get it completed/Unable to draw blood	3
We were concerned about the expense	2
Religious beliefs and trust in God	2
Do not believe it is important	1
We did not have access	1
Newborn screening takes away parent's rights when treatment is forced	1