article

Prospective assessment in newborns of diabetes autoimmunity (PANDA): Maternal understanding of infant diabetes risk

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Purpose: To assess accuracy of mothers' understanding of their newborns' genetic risk for type 1 diabetes and to identify predictors of the comprehension and retention of genetic information. **Methods:** Mothers of 435 newborns genetically screened at birth were informed of the infant's risk for type 1 diabetes using a standard script that provided both categorical and numerical risk information. The mothers' comprehension and retention of this information were assessed by structured interview on two occasions, ~3.6 weeks and ~3.9 months postnotification. **Results:** At the initial interview, 73.1% of mothers gave a correct estimate of their child's genetic risk, 3.2% overestimated risk, 13.3% underestimated risk, and 10.3% could not recall risk at all. At the follow-up interview, fewer mothers (61.9%) correctly estimated their child's risk and more mothers (24.4%) underestimated their child's risk. Maternal accuracy was associated with maternal education, ethnic minority status, infant risk status, maternal ability to spontaneously recall both categorical and numerical risk estimates, and length of time since risk notification. Underestimation of risk was associated with maternal education, family history of diabetes, time since risk notification, and maternal anxiety about the baby's risk. **Conclusion:** The accuracy of mothers' recall of infant risk declines over time, with an increasing number of mothers underestimating the infant's risk. Effective risk communication strategies need to be developed and incorporated into genetic screening programs. ***Genet Med*** **2003:5(2):77–83.**

Key Words: *newborn, genetic screening, type 1 diabetes, risk communication, parental understanding*

A genetic risk is defined as the degree of association between particular characteristics and a disease within a defined population.[1] The ability to predict genetic risk depends on the sensitivity and specificity of the test. Although tests of genetic risk are available for a number of diseases, little research has been devoted to the best method of communicating genetic risk information to the general public nor have barriers to risk comprehension been clearly identified. Furthermore, research is needed to assess whether risk communication techniques impact decisions people make about genetic testing or other health-related behaviors.

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The available literature suggests that the relationship between genetic counseling and emotional outcomes is, at best, unclear.[2,3] The accuracy of information recalled after genetic counseling may be influenced by many variables, including education,[4–7] ethnicity,[8–11] and family history of the disease.[12,13] In general, better understanding of risk information is associated with higher education, membership in the majority culture, and a family history of the disease. Several studies suggest that recipients of genetic information demonstrate decreased retention across time[14–19] and a tendency toward underestimation of risk.[20] As only 32% of the US population has a college education,[21] it is likely that many people will have trouble understanding and retaining complex genetic risk information.

Investigators have only recently begun to search for the best method of communicating genetic risk information. Studies have focused primarily on adults at increased risk for breast cancer or Huntington disease, as well as carriers for diseases such as sickle cell or cystic fibrosis (CF).[5,22–27] More recently, decoding of the human genome has led to screening for childhood-onset conditions such as Duchenne muscular dystrophy,[28,29] familial adenomatous polyposis,[30] and multiple endocrine neoplasia type 2.[31]

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Unfortunately, our knowledge of genetic risk has outpaced our knowledge of how to communicate that risk information to the general public.[32,33] In a 3-year follow-up of those who had undergone population screening for CF carrier status, 20% of carriers and 50% of those with negative results were inaccurate in their recall of their test results.[27] It is commonly reported that when parents of a child with CF learn their chances of having another child with the disease are one out of four, many are relieved as they believe they have already "used up" their one chance of having a child with CF.[34] It is apparent that studies of factors affecting the comprehension and retention of genetic risk need to be a priority, not only with at-risk adults but with parents of at-risk children.[35–38]

Newborn genetic screening for type 1 diabetes

Nearly 90% of type 1 diabetes occurs in families with no history of the disease,[39] and the prevalence of type 1 diabetes is greater than that of all other serious childhood chronic diseases.[40] Currently, several research programs are screening newborns from the general population to identify babies at risk for type 1 diabetes, and these high-risk children are then followed over the course of many years to elucidate factors contributing to the pathogenesis of disease.[41–45] Knip[46] reviewed the advantages of population screening for this disease, including the fact that for 95% of children, families may learn that their child is at a decreased risk for disease development. Other advantages include the possibility of identifying at-risk individuals for possible disease prevention as well as the potential to increase public understanding about diabetes and its symptoms and increase early detection. As population-based genetic screening efforts increase, it becomes important to document participant understanding of, and reactions to, the news of risk and to develop appropriate educational or counseling interventions.[47]

Previous research conducted in our laboratory has documented significant psychological sequelae associated with the use of screening programs to identify persons at risk for the development of type 1 diabetes in both adults and children.[47–51] In general, at-risk notification has been found to induce clinically significant anxiety in the at-risk individual and in family members, especially mothers of at-risk children.[49–52] However, no one has yet examined whether parents of children who have been genetically tested for their risk of developing type 1 diabetes truly understand the risk information they are given. The present study design allowed for controlled presentation of infant risk information and subsequent follow-up with mothers to assess their understanding of the infant's risk for developing type 1 diabetes.

MATERIALS AND METHODS

Prospective Assessment of Newborns for Diabetes Autoimmunity

The longitudinal Prospective Assessment of Newborns for Diabetes Autoimmunity (PANDA) is a National Institutes of Health and Juvenile Diabetes Research Foundation International–supported registry that uses genetic testing to identify newborns at risk for type 1 diabetes.[53] Most mothers were contacted at the time of their child's birth and asked permission to screen the newborn for the presence of the high-risk HLA-DQB1 alleles using blood spots on filter paper (obtained by heel stick at the time of state-mandated phenylketonuria testing). Occasionally, mothers of older babies were approached or requested that their babies be genetically screened. Mothers were told they would be re-contacted only if their child was at increased risk for type 1 diabetes. Informed consent was obtained.

The risk of diabetes development in the general population is estimated at 1–2/300.[40] Infants tested were assigned to one of six risk categories depending on the child's HLA-DQB1 allele status and family history of type 1 diabetes (Table 1). Mothers in the low, very low, and protected risk categories were not recontacted. Mothers in the moderate, high-risk, and extremely high-risk groups were sent letters asking them to call for their infant's test results. If mothers did not call, efforts were made to contact the mothers by telephone. Once telephone contact was made, mothers were provided their infant's risk status using a script (see below), any questions were answered, and mothers were asked permission for our research team to contact them for a telephone interview.

The script read as follows: "Hello, Ms. ____, my name is ____ with the University of Florida Diabetes Research office. As you recall, your baby participated in a study prior to leaving the hospital to see if he/she had any genes that would put him/her at risk for developing diabetes. We have found that your baby is at moderate risk for developing diabetes. Please keep in mind that this does not mean that your baby will definitely get diabetes. Out of 100 babies with the same genetic markers as your baby there would be approximately 2 babies who would develop diabetes. We would like to continue to follow your baby to see if we can gain more information as to why some

Table 1
Infant risk categories by DR/DQ status and family history of type 1 diabetes

DR/DQ	With first-degree type 1 relative	Without first-degree type 1 relative
DR 3/4	1/4–5 Extremely high risk	1/15 High risk
DR 4/4	1/6 Extremely high risk	1/20 High risk
DR 3/3	1/10 High risk	1/45 Moderate risk
DR 4/X	1/15 High risk	1/60 Moderate risk
X/X	1/125 Low risk	1/600 Very low risk
DQ 0602 JDR 0403	1/400 Protected	1/15,00 Protected

Ratios represent the likelihood the child will develop type 1 diabetes.

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babies may go on to develop diabetes and some babies never develop diabetes. Do you have any questions?" (*Note: numbers used for high-risk infants were 10 out of 100 for infants with a first-degree relative with type 1 diabetes and 5 out of 100 for infants with no first-degree relative with type 1 diabetes; number used for extremely high-risk infants was 20–25 out of 100.*)

At-risk notification

Using the standardized script, mothers of babies identified as moderate, high, or extremely high risk were informed of their child's results by PANDA staff using both categorical terminology (i.e., moderate, high, extremely high) and numerical estimates (2–25%) of risk, as suggested by the literature.[6,7,54] At the time of notification, the infants ranged in age from 1 month to 3 years with a mean age of 6 months. Mothers' questions were answered. PANDA staff then requested permission to follow the infant and asked whether the mother could be recontacted by telephone. The length of the notification telephone call ranged from 2 to 30 minutes, with an average of 6.6 minutes (SD = 4.4 minutes).

Participants

Those mothers of infants at increased risk for diabetes (moderate, high, or extremely high) who agreed to be interviewed were contacted. Although some mothers were Spanish-speaking, all mothers interviewed could speak English. In this at-risk sample, the majority of infants were at moderate risk (60.0%), 34.7% were at high risk, and 5.3% were at extremely high risk. Most of the mothers (74.7%) reported the infant had some family history of diabetes (type 1 or type 2). Participation rate was high; 90.3% ($n = 435$) of the mothers we were able to contact agreed to be interviewed and 79% consented to participate in a follow-up interview ($n = 344$). The demographic characteristics of the study participants are shown in Table 2. The sample was largely Caucasian (80.4%), married (75.4%), and highly educated (72.2% reported having some college education). The mean maternal age at time of the child's birth was 28.7 years (range 17–43).

Table 2
Demographic characteristics of study participants ($n = 435$)

Characteristic	No.	%
Marital status		
Married	325	75.4
Single, involved father	69	16.0
Single, noninvolved father	23	5.3
Separated	5	1.2
Divorced	9	2.1
Maternal ethnic group		
Caucasian	348	80.4
Hispanic	38	8.8
African American	26	6.0
Asian/other	21	4.8
Maternal educational level		
Some high school	30	7.0
High school/GED	90	20.9
Some college	158	36.7
Completed college	111	25.8
Some graduate school	10	2.3
Completed graduate school	32	7.4
Infant risk classification		
Moderate risk (2%)	261	60.0
High risk (5–10%)	151	34.7
Extremely high risk (20–25%)	23	5.3
Family history of diabetes		
No family history	110	25.3
Family history		
Maternal gestational	9	2.0
First-degree relative	49	11.3
≥Second-degree relative	267	61.4

GED, general equivalency diploma.

Procedure

Those mothers who agreed to be interviewed were contacted twice: an average of 3.6 weeks (SD = 3.9 weeks) and an average of 3.9 months (SD = 1.8 months) after risk notification. On both occasions, a structured interview was used to assess maternal understanding of the infant's risk and maternal beliefs about the likelihood the infant would develop diabetes. Maternal anxiety about the child's risk was assessed using the state component of the State-Trait Anxiety Inventory (STAI).[55] The STAI has proven to be a reliable and valid measure of both trait and situation-specific anxiety. In our sample, this measure was found to be highly reliable at the initial ($\alpha = 0.93$) and follow-up ($\alpha = 0.92$) interviews. The study procedures were approved by the University of Florida Health Science Center Institutional Review Board.

RESULTS

Maternal understanding of infant risk

As the study design included a standardized "scripted" presentation of infant risk status by PANDA staff, we could compare mothers' reports of the baby's risk status with what the mother was actually told. Accuracy was determined by first allowing the mother to provide a spontaneous response if she was able, with some prompting such as "Do you remember a specific category or any numbers you were told about?" A correct response, either categorical or numerical, was considered accurate. If the mother could not remember, or had conflicting estimates, a recognition task was given to the mother to determine whether she was able to recognize her child's risk category. At the initial interview (~3.6 weeks after the mother was

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first informed of the baby's at-risk status by the PANDA staff), 73.1% of mothers gave a correct estimate of their child's genetic risk information, 3.2% overestimated their child's risk, 13.3% underestimated their child's risk, and 10.3% could not recall any risk at all, even after given six combined categorical and numerical risk estimates for recognition purposes.

At the follow-up interview, approximately 3.9 months after at-risk notification, more mothers (24.4%) underestimated their child's risk. There was no substantive change in the number of mothers who overestimated the baby's risk (3.2%) or who could not remember the baby's risk (10.5%).

The McNemar test was used to assess the change in maternal accuracy of risk estimates across time. This analysis was limited to mothers with accuracy data for both interviews ($n = 344$) (Table 3). Results indicated that there was a significant decline in accuracy from 73.4% accurate at the initial interview to 61.9% accurate at the follow-up interview ($\chi^2 = 76.9$, $P < 0.001$).

The McNemar test also documented a significant increase in the number of mothers who underestimated their child's risk at the follow-up interview (24.4%) compared with the initial interview (13.3%) ($\chi^2 = 40.1$, $P < 0.001$). No changes over time were observed for those mothers who initially overestimated or who were unable to recall or recognize their child's risk status (Table 3).

At both interviews, we also asked mothers whether they believed their child would ever develop diabetes. Initially, most mothers (71.8%) indicated they were unsure about their child's ultimate health status, although 20.6% stated their child would never develop diabetes and 7.9% indicated they expected the child to ultimately become diabetic. At the second interview, fewer mothers (64.7%) remained unsure of their baby's health status. There were small increases in the percentage of mothers (10.8%) who were convinced the child would ultimately develop diabetes and the percentage of mothers who believed that the child would *never* develop the disease (23.3%).

Factors associated with accuracy of maternal understanding of infant diabetes risk

Logistic regression analyses were used to assess maternal and infant variables that may be associated with maternal accuracy about the baby's risk status. We tested the following predictors: maternal demographic variables (ethnic minority status, education, age, whether this is her first child, family history of diabetes, marital status); study variables (infant risk, PANDA staff member who informed the mother of risk, length of PANDA staff notification telephone call, length of time since risk notification); and maternal reaction variables (recall of a risk category or number, expectations about whether the child would develop diabetes, and anxiety associated with risk notification). At the time of our initial interview, mothers with lower levels of education, and those from an ethnic minority group including African-American and Hispanic women, were less likely to be accurate about their child's risk (Table 4). Sixty-one percent of mothers with a high school education or below had accurate recall of their infant's risk, while 78% of mothers with some college education and beyond reported accurate infant risk results. Seventy-eight percent of Caucasian mothers were accurate while 38.5% of African-American mothers and 57.9% of Hispanic mothers were accurate.

At the initial interview, mothers who spontaneously recalled both a category and a numerical risk estimate (27.4%) were significantly more likely to be accurate (93.3% vs. 65.5% accurate). The longer the time elapsed between risk notification by PANDA personnel and the initial interview, the less likely the mother's risk estimate would be accurate. Of interest, mothers with extremely high-risk infants tended to be marginally less accurate than mothers of either high-risk or moderate-risk infants (60.9% accurate for extremely high risk vs. 71.6% for moderate risk and 77.5% for high risk).

At the follow-up interview, maternal accuracy was best predicted by initial accuracy of risk perception; mothers who were accurate at the initial interview were more likely to be accurate at the second interview (Table 5). Mothers of infants from lower-risk groups were again more likely to be accurate than those from the extremely high-risk group (36.4% for extremely high risk, 62.4% for moderate risk, 65.8% for high risk). Time elapsed since risk notification remained a significant predictor, with longer time intervals associated with lower accuracy. With all of these factors controlled, African-American mothers

Table 3
Accuracy of recall for mothers with both initial and follow-up interviews ($n = 344$)

	Time 1		Time 2	
Recall	No.	%	No.	%
Accurate	254	73.4	213	61.9
Underestimate	45	13.1	84	24.4
Overestimate	12	3.5	11	3.2
Don't know/don't remember	33	9.6	36	10.5

Table 4
Summary of logistic regression analysis for variables predicting accuracy of maternal risk perception at initial interview ($n = 431$)

Variable	*B*	SE *B*	*P*
Maternal education	0.30	0.13	0.019
African American	−1.72	0.48	0.001
Hispanic	−0.76	0.38	0.044
Extremely high risk	−0.88	0.48	0.065
Both category & number recalled/recognized	1.97	0.42	0.001
Time since risk notification	−0.41	0.12	0.001

This model's correct classification rate = 77.5%. Categorical variables African American, Hispanic, extremely high risk, and both category & number recalled/recognized were coded as 1.

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Table 5
Summary of logistic regression analysis for variables predicting accuracy of maternal risk perception at follow-up interview ($n = 344$)

Variable	*B*	SE *B*	*P*
Initial accuracy	2.48	0.32	0.001
African American	1.15	0.65	0.076
Extremely high risk	−1.07	0.53	0.044
Time since notification	−0.26	0.07	0.001

This model's correct classification rate = 76.4%. Categorical variables initial accuracy, African American, and extremely high risk were coded as 1.

were slightly more accurate than other mothers (64.7% of African-American mothers were accurate vs. 61.7% of all other mothers). Maternal age, whether this was the mother's first baby, family history of diabetes, the PANDA staff member who informed the mother of the infant's risk status, the length of PANDA staff notification telephone call, maternal expectations about diabetes development, marital status, and mother's initial anxiety were unrelated to accuracy of maternal risk perception.

Factors associated with maternal underestimation of infant diabetes risk

Logistic regression analyses were used to assess maternal and infant variables associated with maternal underestimation of infant diabetes risk (Tables 6 and 7). At the time of the initial interview, mothers with a lower level of education were more likely to underestimate infant risk (22.5% of mothers with a high school education and below underestimated their infant's risk compared with 9.6% of mothers with some college education or beyond). Mothers whose child had a first-degree relative with diabetes were also more likely to underestimate their child's risk (24.5% underestimated their infant's risk compared with 13.9% of mothers with infants who had a ≥ second-degree diabetic relative and 8.2% of mothers with infants who had no diabetic relative). The longer the time elapsed since risk notification, the more likely mothers were to underestimate their child's risk. Mothers who were unable to spontaneously recall both a risk category and a number at the initial interview were more likely to underestimate their child's risk (16.5% underestimated risk compared with 5.0% of mothers who did recall both a risk category and a number).

Table 6
Summary of logistic regression analysis for variables predicting maternal underestimation of infant risk at initial interview ($n = 431$)

Variable	*B*	SE *B*	*P*
Maternal education	−0.44	0.16	0.005
Family history of diabetes (first-degree relative)	1.19	0.40	0.003
Time since notification	0.34	0.13	0.009
Both category & number recalled/recognized	−1.31	0.34	0.000

This model's correct classification rate = 87.5%. Categorical variables family history of diabetes and both category & number recalled/recognized were coded as 1.

Table 7
Summary of logistic regression analysis for variables predicting maternal underestimation of infant risk at follow-up interview ($n = 342$)

Variable	*B*	SE *B*	*P*
Underestimation at initial interview	2.02	0.36	0.001
Family history of diabetes (≥second-degree relative)	−0.58	0.28	0.035
Time since notification	0.16	0.07	0.033
Anxiety at follow-up interview	−0.38	0.02	0.088

This model's correct classification rate = 78.4%. Categorical variables underestimation at initial interview and family history of diabetes were coded as 1.

At the time of the follow-up interview, those who initially underestimated the infant's risk were more likely to continue to underestimate the child's risk (Table 7). Mothers with infants who had no family history of diabetes (30.2% underestimated risk) or who had a first-degree relative with diabetes (28.3% underestimated risk) were more likely to underestimate the infant's risk than mothers of children with a ≥ second-degree diabetic relative (21.1% underestimated risk). The longer the time elapsed since risk notification, the more likely mothers were to underestimate their child's risk. Mothers who reported little anxiety about their child's risk status at the follow-up interview were marginally more likely to underestimate the child's risk.

Maternal age and ethnicity, whether this was the mother's first baby, marital status, the PANDA staff member who informed the mother of the infant's risk status, the length of the PANDA staff notification telephone call, the infant's degree of risk (moderate, high, or extremely high), and maternal expectations about diabetes development were unrelated to maternal underestimation of infant risk.

DISCUSSION

Only 73.1% of mothers were able to accurately recall their infant's diabetes risk shortly after risk notification. Furthermore, the number who could accurately recall this information declined over time to 61.9%. Maternal education and ethnic minority status were important correlates of accurate recall; those who were inaccurate were less educated and from the ethnic minority community. These findings suggest that particular care is needed when communicating risk information to less-educated mothers and mothers from minority groups, such as African Americans and Hispanics. Simple interventions, such as using checklists, follow-up questions, follow-up letters,[56] or face-to-face education and/or counseling,[57–59] may be needed to ensure accurate initial understanding and continued accurate retention of risk information. Mothers who could recall both a categorical description and a numerical estimate of the infant's risk were more likely to be accurate, suggesting that assuring initial understanding of both types of risk infor-

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mation may promote accurate maternal perceptions of infant diabetes risk.

A substantial minority of mothers underestimated the infant's risk, and this number increased over time. Underestimation was more common among less educated mothers. It appears that many mothers who receive potentially alarming news about the infant's risk for developing diabetes respond by underestimating the baby's risk. Underestimation may be an attempt at psychological protection; previous studies of individuals given information about diabetes risk have also documented the use of risk minimization as a coping strategy.[51]

Family history and infant risk status had interesting and somewhat complex relationships to maternal comprehension and retention of infant risk information. Mothers of extremely high-risk infants tended to be the least accurate at both the initial and follow-up interviews. All of these mothers' infants had first-degree relatives with diabetes, so their inaccuracy was not a function of lack of familiarity with type 1 diabetes. In fact, mothers of infants who had a first-degree diabetic relative were more likely to underestimate the infant's risk (24.% underestimated risk at the initial interview and 28.3% underestimated risk at the follow-up interview). However, mothers of infants with no diabetic family history showed the largest increases in risk underestimation from the first (8.2% underestimated) to the follow-up interview (30.2% underestimated). These data suggest that some family history of a disease may be helpful in terms of risk communication and retention but that in cases of first-degree relatives with the disease, other factors may impede acceptance of the information.

Future studies should examine risk communication strategies and methods to ensure accurate understanding of risk information in persons from a variety of educational and ethnic backgrounds, as well as experience with the disease about which risk information is being communicated. Effective risk communication is essential if we are to ensure participants' decision-making is based on accurate understanding of risk information. Without accurate understanding, participants' treatment decisions or their decisions to continue or discontinue participation in longitudinal study protocols so important to understanding the etiology of a disease like type 1 diabetes will not be truly informed.

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