

Does provision of comprehensive information about newborn screening increase parents' decisional conflict?

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Newborn screening

- Most widespread example of population screening
- Expansion of screening panels – shift from “public health emergency” to “public health service”¹
- What do parents need to know?



¹Grosse SD, Boyle CA, Kenneson A, Khoury MJ, Wilfond BS. From public health emergency to public health service: the implications of evolving criteria for newborn screening panels. *Pediatrics* 2006;117: 923-9.

More extensive parental education

Pros

- Consistent with informed consent (where this is the model)
- May support the screening process in action
- May promote appropriate follow up of screen-positive results
- May mitigate psychosocial harms
- *Meets parent expectations and preferences for communication – psychological preparation, usefulness in informing other decisions, feeling of being respected, promoting trust*

Cons

- Concern about promoting parental anxiety
- Concern about lowering screening uptake

In practice

- NBS programs in US and Canada vary widely in the educational messages they send parents – cover **purpose, benefits**, process, harms, incidental findings, secondary use of samples
- Evidence base suggests generally low knowledge of process, potential outcomes, diseases screened for
- No clear evidence on which educational content is important to parents²

²Araia MH, Wilson BJ, Chakraborty P, Gall K, Honeywell C, Milburn J, Ramsay T, Potter BK. Factors associated with knowledge of and satisfaction with newborn screening education: a survey of mothers. *Genet Med* 2012;doi:10.1038/gjm.2012.87

Study rationale

Given the positive arguments that educational efforts may promote positive effects beyond being necessary for “informed consent”, should we be concerned that education might have negative effects?

Objective

To measure and compare expecting mothers’ responses to different educational messages about NBS, so as to identify those that are most relevant with respect to decision-making.

Study setting

- ~140,000 babies born in Ontario each year
- Newborn Screening Ontario screens all infants born for 28 conditions
- Not mandated but highly recommended
- Extremely high uptake rates



Methods

- **Factorial survey** - participants receive different combinations of specific educational messages, and their association with outcomes of interest is assessed
- Study population – pregnant women attending for routine ultrasound
- Survey completed in waiting room, following which standard information brochure provided
- Approved by research ethics boards of The Ottawa Hospital and the Children’s Hospital of Eastern Ontario

Factorial survey³

- Standard sample survey modified to permit manipulation of factors of interest (independent variables)
- Analysis explores importance of each factor, and their interactions, in relation to outcome(s) of interest
- Useful for studying opinions, attitudes and decisions related to complex situations

³Rossi HR, Anderson AB. The factorial survey approach. An introduction. In: *Measuring social judgments. The factorial survey approach*. Eds Rossi PH, Nock SL. Beverly Hills/London/New Delhi: Sage Publications, 1982: 15-17.

Survey structure

- Introduction
- Information about NBS ***“please read before answering the questions”***:
 - Four standard paragraphs describing Ontario NBS
 - One paragraph with “consent” message
 - Up to four additional paragraphs with other messages
- Questions – attitude, knowledge, views on health care
- Demographics
- **32 versions**

“Consent”

Newborn screening is not mandatory in Ontario. It is considered part of standard newborn care. ...

“choice version”:

Parents have the right to choose to accept or decline newborn screening for their baby. A parent may wish to discuss this decision with a health care provider.

“directive version”:

Parents have the right to choose not to have their baby screened. If a parent wants to decline newborn screening they should first discuss this with a health care provider.

“Pain”

The heel prick used to obtain a blood sample for NBS causes a very small amount of pain to the newborn baby. Various methods are used to comfort the baby. These include feeding and/or cuddling the baby right after the sample is taken.

“False positive/negative”

Most babies will receive a ‘screen negative’ NBS test result, which is normal. Some babies will receive a ‘screen positive’ result, which could indicate a problem. A ‘screen positive’ result does *not* mean that a baby definitely has a disease. The baby will need further tests to find out for certain if he or she has the disease. It is possible for the NBS tests to miss a baby with a disease. However, this is very rare.

“Over-diagnosis”

Early diagnosis and treatment improves the health of many babies who have the diseases included in NBS. Some babies have a very mild form of the disease. This mild form of disease might not cause health problems later on. However, there is often no way to know which babies will develop health problems and which will not. Because of this, all babies diagnosed with a disease will be seen by a doctor for monitoring and sometimes treatment.

“Storage/secondary uses”

The blood sample taken from a baby’s heel is stored on a paper card. The card is sent to a laboratory in Ottawa for testing. Newborn Screening Ontario stores the paper cards in a secure facility for 19 years. After 19 years, the cards are destroyed. The blood samples on the stored cards can legally be used in these ways:

1. Helping the screening program ensure that the testing methods are working properly.
2. Re-testing the sample at the request of the baby’s doctor.
3. Use after a legal warrant or court order (e.g. by the Coroner’s Office if the baby has died unexpectedly)
4. Release to another laboratory. The sample may be used for testing if the parent or guardian makes a written request.
5. Anonymous research approved by a research ethics board. In this case, all identifying information has been removed. It is impossible to link a person with the research results. This is allowed under the Ontario Personal Health Information Privacy Act (2004). Identifiable samples can only be used for research if the person or their parent/guardian gives written consent.

Primary outcome – decisional conflict

Decisional Conflict Scale⁴ (‘question format’ version), measures personal perceptions of

- a. Uncertainty in choosing options
- b. Modifiable factors contributing to uncertainty such as feeling uninformed, unclear about personal values and unsupported in decision making
- c. Effective decision making such as feeling the choice is informed, values-based, likely to be implemented and expressing satisfaction with the choice

⁴O’Connor AM. *User manual - Decisional Conflict Scale*. Ottawa: Ottawa Hospital Research Institute; 1993, updated 2010.
http://decisionaid.ohri.ca/docs/develop/User_Manuals/UM_Decisional_Conflict.pdf

Interpretation

- 16 questions, scored 0-100, reported as continuous score
- In comparing groups, meaningful effect sizes ~10 points

- Scores <25 generally reflect low decisional conflict
- Scores > 37.5 generally reflect high decisional conflict

Secondary outcomes

1. Current preference - whether respondent thinks she will want to have her baby screened
2. Actual screening status (follow up with Newborn Screening Ontario, with participants' consent)



Analysis

Focuses on the association between **being offered specific messages** and **decisional conflict**.

Target sample size = 500. Study powered to allow evaluation of multiple interactions between messages, and with demographic and other participant characteristics

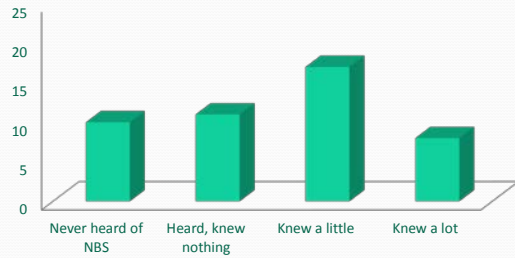
First 46 participants

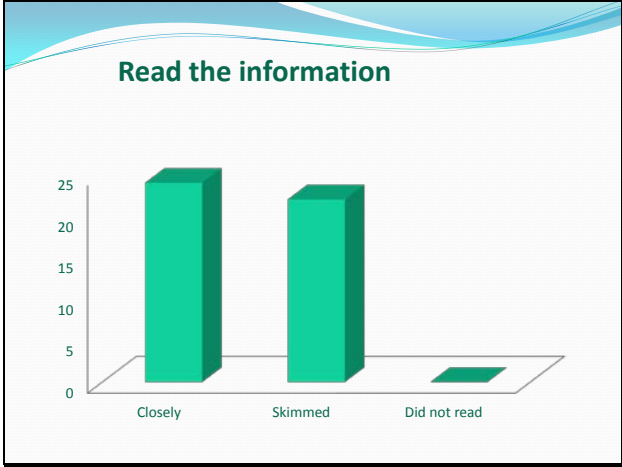
	N	% (95% CI)
Primary caregiver during pregnancy		
Obstetrician	27	59 (44-73)
Family doctor	19	41 (27-56)
Education		
High school or less	8	17 (6-28)
College	14	30 (17-44)
University or higher	24	52 (38-67)
Employed as health professional	15	33 (19-46)
Primiparous	16	35 (21-49)

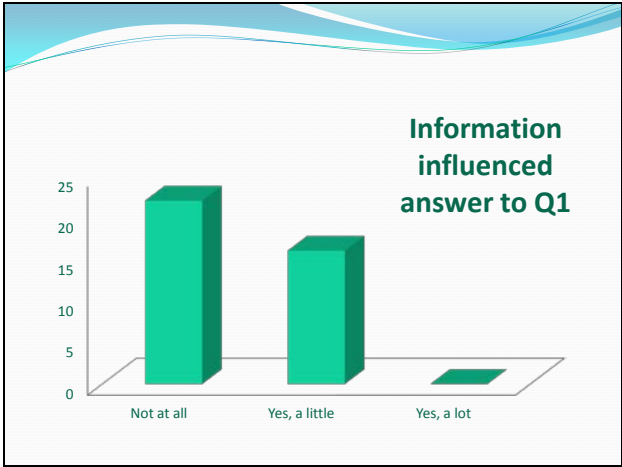
Randomization

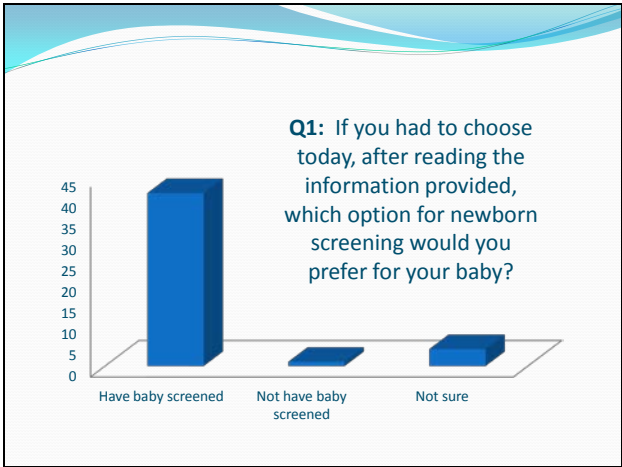
Message	n
"Consent"	24
"choice"	22
"directive"	22
"Pain"	31
"False positive/negative"	23
"Over-diagnosis"	18
"Storage/secondary uses"	25

Prior knowledge of NBS









Knowledge

Statement	Correct % (95% CI)
Newborn screening identifies babies who look healthy but may have a disease (T)	93 (86-100)
A baby's blood sample from newborn screening will be thrown out as soon as it is tested (F)	59 (44-73)
If a baby has a 'screen positive' newborn screening result, it means that the baby has the disease (F)	46 (31-60)
Newborn screening tests for very common diseases (F)	76 (64-88)
Newborn screening is not mandatory for babies in Ontario (T)	91 (83-99)
All babies diagnosed with disease included in newborn screening will develop health problems without treatment (F)	52 (38-67)

Decisional conflict

- Mean score = 25 (95% CI 19-31)
- min = 0, max = 55, median = 25
- 17% respondents scored 0 – high certainty or “satisficing”?

Score	N (%)
<25	23 (50)
25-37.5	10 (22)
>37.5	13 (28)

Messages sent and decisional conflict

	Received?	N	Mean DCS score (95% CI)	p*
Consent	<i>“choice”</i>	24	19 (11-27)	0.02
	<i>“directive”</i>	22	32 (24-40)	
Pain	<i>Yes</i>	31	27 (20-33)	0.49
	<i>No</i>	15	22 (12-33)	
False positive/negative	<i>Yes</i>	23	33 (24-41)	0.006
	<i>No</i>	23	18 (11-25)	
Over-diagnosis	<i>Yes</i>	18	26 (17-36)	0.74
	<i>No</i>	28	24 (17-32)	
Storage/secondary use	<i>Yes</i>	25	24 (17-31)	0.66
	<i>No</i>	21	26 (17-36)	

*Difference between means, t-test

Discussion

- Rigorous method, empirical orientation, messages selected and worded after evidence reviews and expert discussions
- High levels of intention to accept screening, low mean levels of decisional conflict
- Preliminary findings suggest some messages have more impact on quality of decision making than others

Discussion

- On the face of it, higher decisional conflict associated with
 - “directive” version of consent message
 - information on possibility of false positive or false negative test results
- Further analyses deferred until target sample size achieved
- Associations, not causation

Conclusions

Evidence-based parental educational interventions in newborn screening require

- a rationale (**why** is education important?)
 - Clarifies relevant outcomes
- and evidence!
 - The effect of specific content
 - The effectiveness of different methods



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Presenter Disclosures

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“No relationships to disclose”
