

Not Without Consent: Parents' Willingness to Permit Use of NBS Sample for Research & Storage



Beth A. Tarini, MD, MS
Assistant Professor of Pediatrics
Child Health Evaluation and Research (CHEAR) Unit
University of Michigan



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Disclosure Statement



Dr. Tarini has
no conflicts of interest to disclose



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Storage of NBS Sample



- Newborn screening
 - One drop of blood taken from nearly every infant born in U.S.
 - Blood tested for a number of inherited disorders
- Storage
 - 20 states store NBS samples from 1 to 23 years



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Use of NBS Samples



- Stored NBS samples used for...
 - retesting of samples
 - evaluating future NBS tests
 - forensic purposes
 - research



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Research & NBS Sample



- Permit studies not otherwise feasible
 - population-based
 - long periods of follow-up



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Consent & NBS Sample



- Most parents not informed or consented
 - use of NBS samples for research
 - storage of NBS samples
- 2007: Minnesota advocacy group calls for destruction of stored NBS samples
- Public backlash could threaten millions of samples stored over decades



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What do parents think?



- Limited data on parents' attitudes about use of NBS samples for research and storage





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Study Objective



- Parents' willingness to permit use of their child(ren)'s NBS samples for research, with and without consent
- Parents' willingness to allow storage of the NBS sample



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Study Design



- Web-enabled survey
- C.S. Mott Children's Hospital National Poll on Children's Health
 - Measures public opinion on major health care issues for U.S. children



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Study Population



- 3935 adults randomly selected from KnowledgePanelSM (Knowledge Networks)
 - Nationally representative panel of adults
 - Compiled by probability sampling of online & offline members of U.S. population
 - Parents were oversampled (n=3047)

Survey Instrument: NBS sample for **research**

Every infant born in the US is tested for inherited diseases.

These tests are done as part of each state's newborn screening program.

Blood is collected for these tests shortly after birth.

After testing is completed, the DNA from these blood samples can be used for future research studies on various diseases.

Survey Instrument: NBS sample for **research**

Order of questions
randomly assigned

Outcome: NBS sample for **research**

How willing are you to have your child's blood sample (from their newborn screening) used for future research studies, with your permission?

- Very Willing
- Somewhat Willing
- Somewhat Unwilling
- Very Unwilling

Outcome: NBS sample for **research**

How willing are you to have your child's blood sample (from their newborn screening) used for future research studies, without your permission?

- Very Willing
- Somewhat Willing
- Somewhat Unwilling
- Very Unwilling

Survey Instrument: NBS sample for **storage**

States may store newborn screening blood samples for varying amounts of time.

Survey Instrument: NBS sample for **storage**

*How long are you willing to have your
child's blood sample
(from their newborn screening) be stored?*

- I would not allow the blood to be stored
- 1 year
- 2 years
- ...
- 18 years
- As long as the state decides it is necessary



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Independent Variables



- Parent age
- Parent gender
- Parent self-reported race/ethnicity
- Parent education
- Household income
- Health status of the youngest child
- Age of first and second child



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Data Analysis



- Census-based sampling weights provided to obtain representative estimates for the US population
 - All analyses performed on weighted sample



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Data Analysis



- Predictors
 - Parent age
 - Parent gender
 - Parent self-reported race/ethnicity
 - Parent education
 - Household Income
 - Health status of the youngest child
 - Age of first and second child



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Data Analysis



- Multinomial Logistic Regression



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Data Analysis: Outcome



- Responses
 - Very Willing
 - Somewhat Willing
 - Somewhat Unwilling
 - Very Unwilling



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Reference Outcomes



Somewhat
Unwilling

VS.

Very
Willing

Somewhat
Unwilling

VS.

Somewhat
Willing



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Response Rate



- Parent response rate = 49.5% (n=1508)
- Non-respondents more likely to
 - be female
 - report non-white race/ethnicity
 - have a lower education



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Parent Demographics



Age (mean), years	38
Female, %	57
Race/Ethnicity, %	
White, Non-Hispanic	67
Black, Non-Hispanic	11
Hispanic	15
Other, Non-Hispanic	7
Education, %	
< = High school	40
> High school	60



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Parent & Child Demographics



Income, %

< \$15,000	10
\$15,000 - \$<30,000	14
\$30,000 - \$<60,000	28
\$60,000 - \$100,000	30
>\$100,000	18

Number of Children (mean) 2

Age of Child (mean), years

First Child	12
Second Child	9.5

Child Health Status, %

All children in excellent or very good health	88
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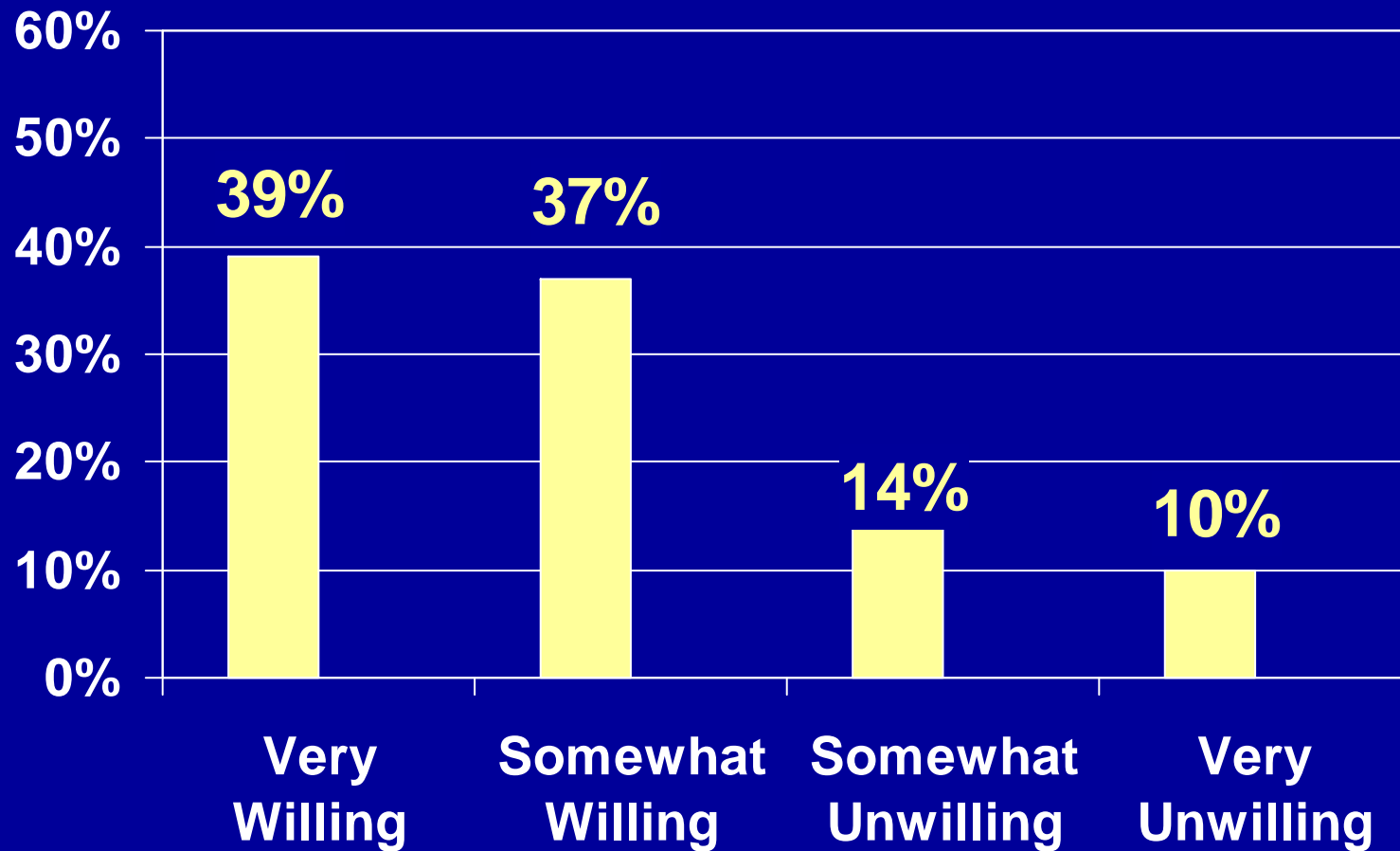


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Use of NBS sample for **research**



With Parental Permission



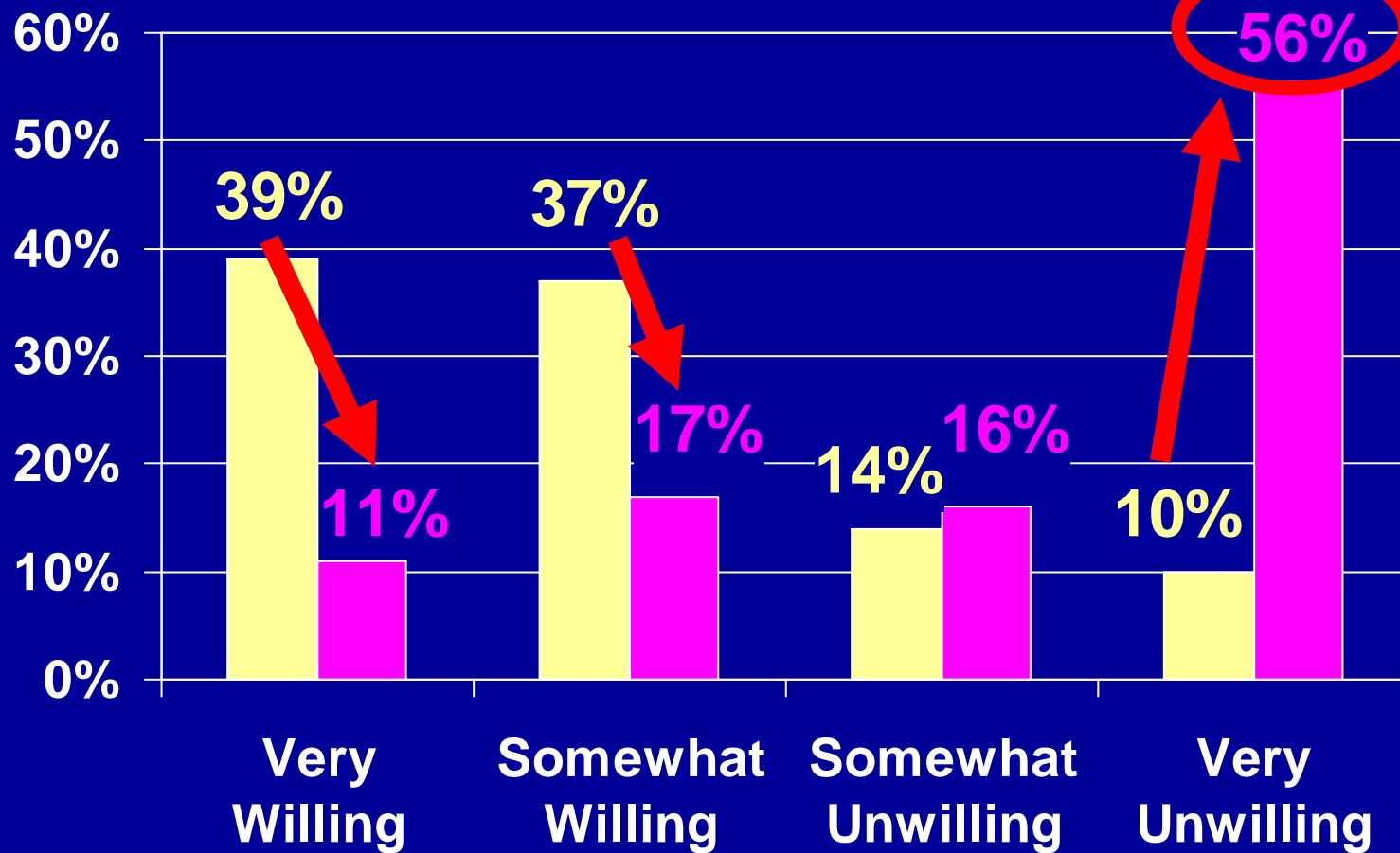


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Use of NBS sample for research



Without Parental Permission





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Use of NBS sample for research

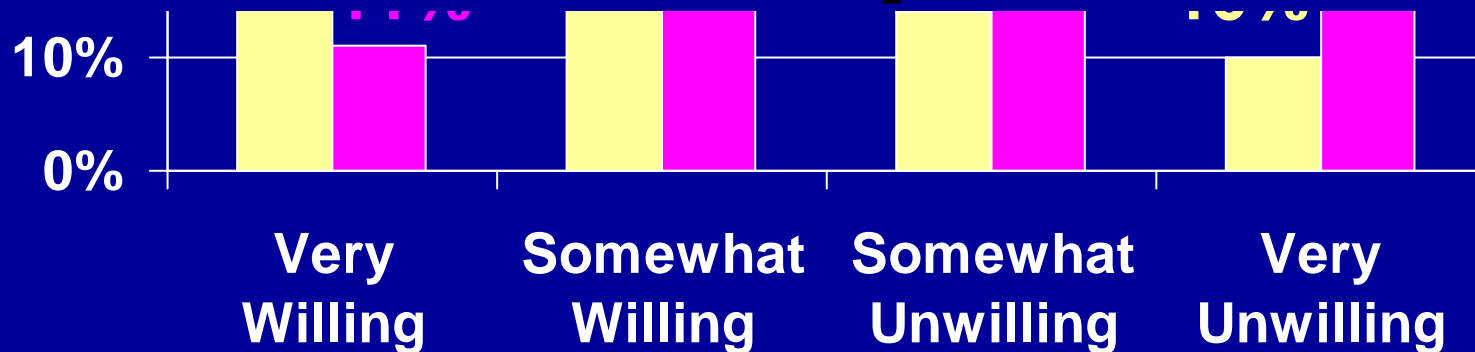


Without Parental Permission

60%

56%

**Order of questions
did not significantly
effect responses**



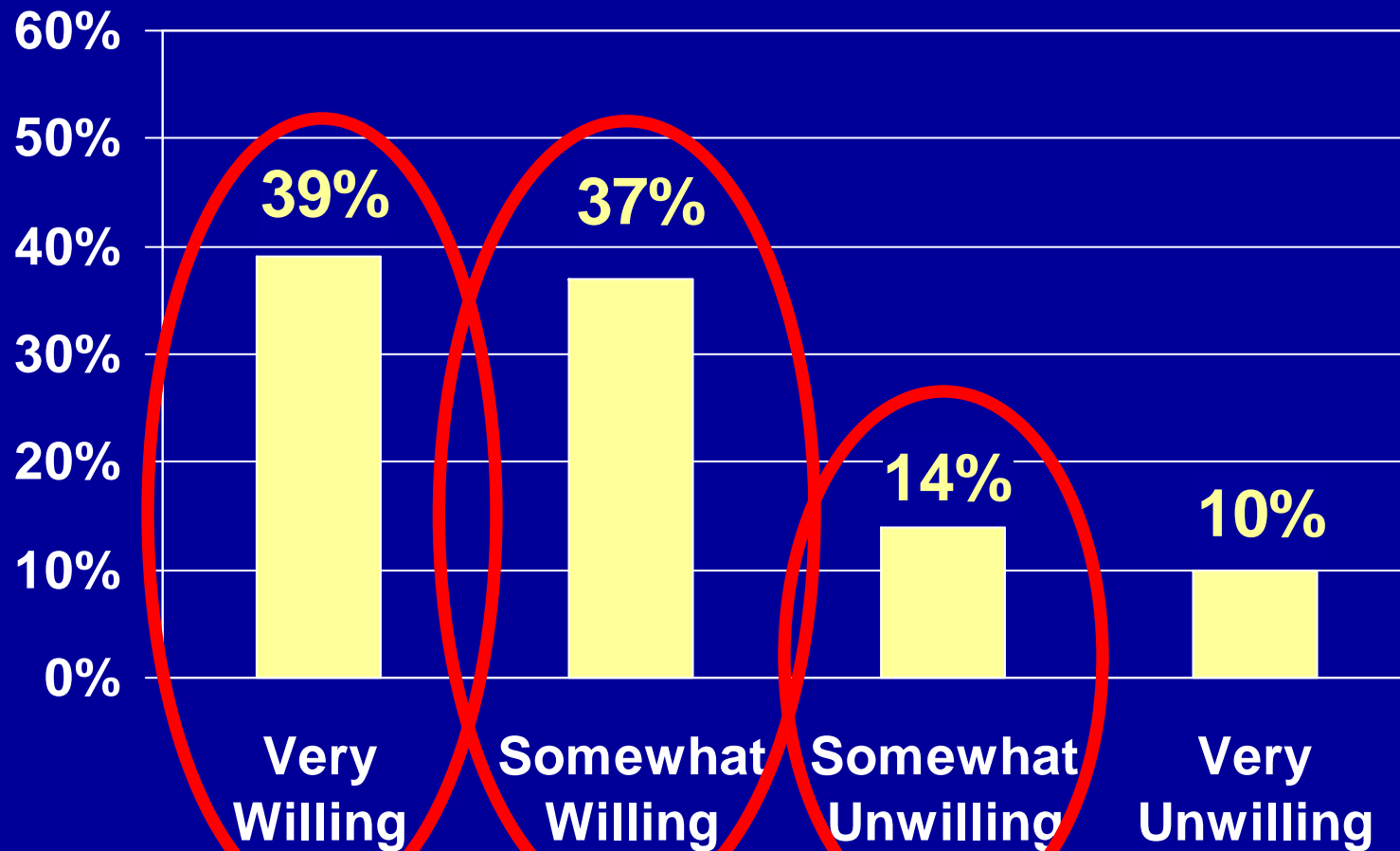


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Use of NBS sample for **research**



With Parental Permission





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Reference Outcomes



Somewhat
Unwilling

VS.

Very
Willing

Somewhat
Unwilling

VS.

Somewhat
Willing



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Significant Finding



Somewhat *Unwilling* vs. Very *Unwilling*

- More likely to report all children in excellent or very good health

Odds Ratio 2.1

(95% CI: 1.2 – 3.9)



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Reference Outcomes



No significant association with

Somewhat parent age vs. Very
 Unwilling parent gender Willing
 parent education

Somewhat household income vs. Somewhat
 Unwilling number of children Willing
 age of first or second child

Outcome: Storage of NBS sample

How long are you willing to have your child's blood sample (from their newborn screening) be stored?

- I would not allow the blood to be stored
- 1 year
- 2 years
- ...
- 18 years
- As long as the state decides it is necessary

Outcome: Storage of NBS sample

Allow state to store NBS samples
for at least some length of time

78% of parents

Outcome: Storage of NBS sample

Allow state to store NBS samples
indefinitely

39% of parents



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Storage of NBS sample



Parents who would
prohibit storage of NBS samples...

More likely to \leq high school education

OR 1.6

(95% CI 1.07-2.5)

More likely to report all children in
excellent or very good health

OR 2.1

(95% CI 1.3-3.6)



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Is there a relationship between
willingness to permit research
and
willingness to allow storage?

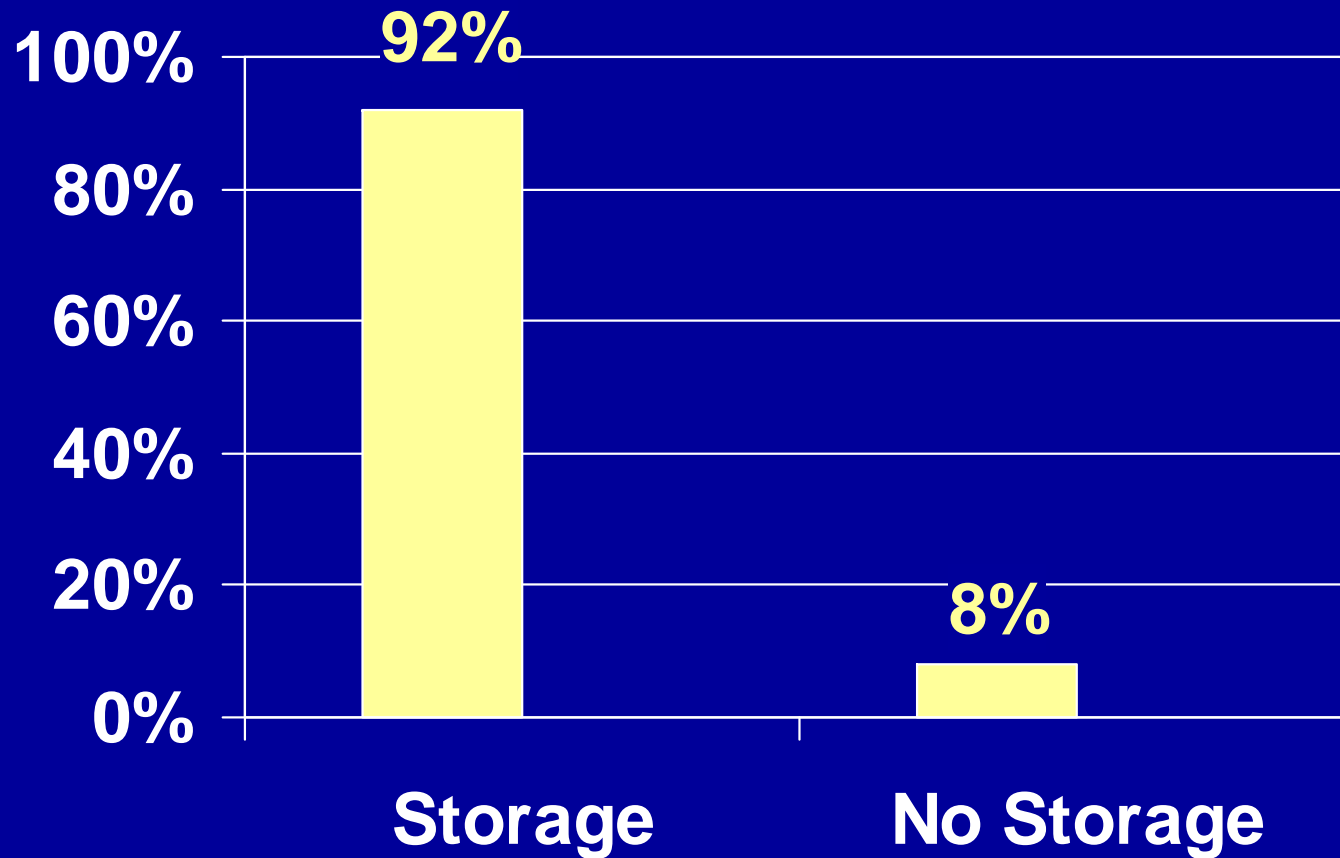


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Research and Storage



Willing to Permit Research



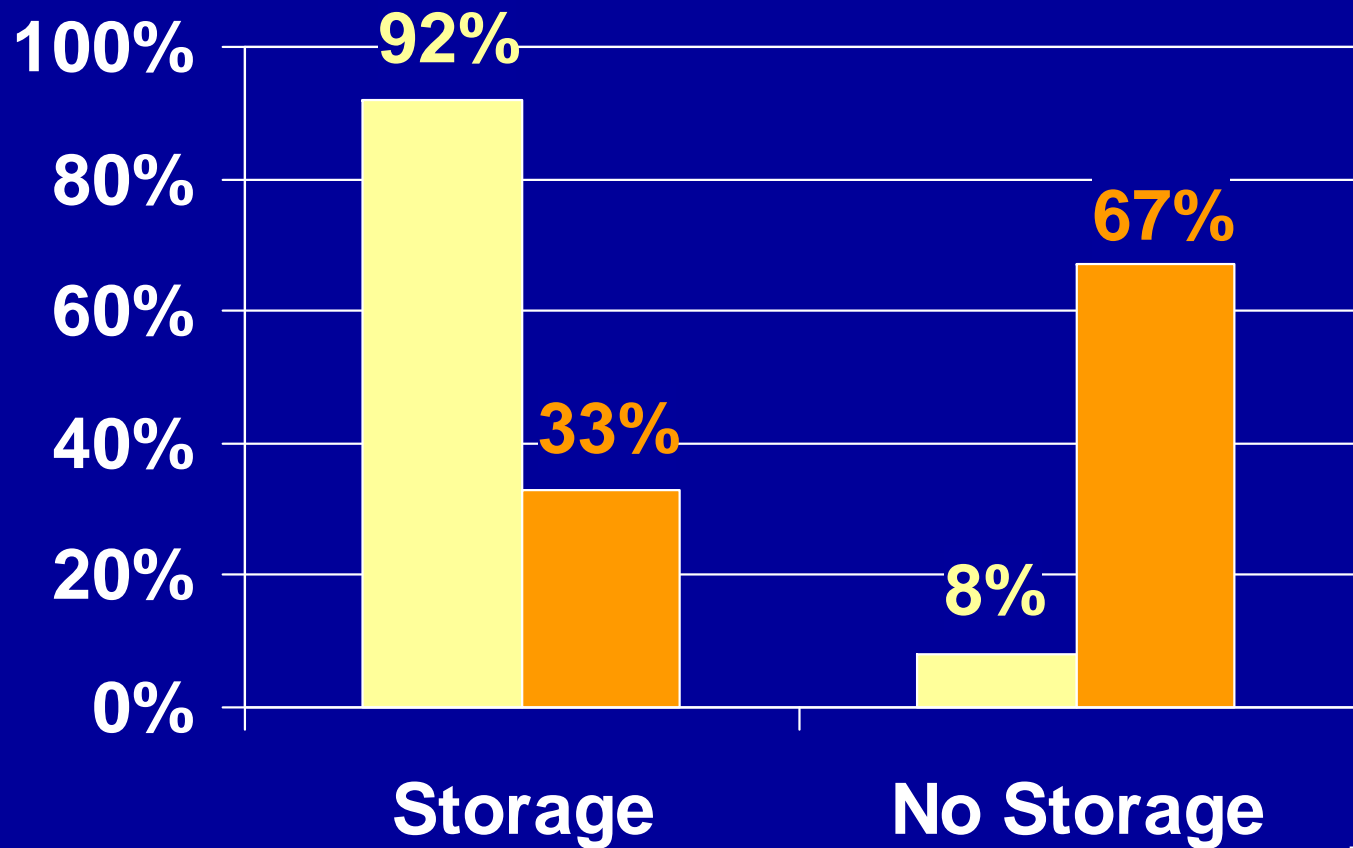


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Research and Storage



Unwilling to Permit Research



$p < .001$



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Conclusions



- A majority of parents are willing to have their children's NBS samples for research—*if their permission is obtained*
- Many parents not supportive of using NBS samples for research
also opposed storage of NBS samples



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Limitations



- Possible response bias
 - May *underestimate* proportion of parents not permitting NBS sample storage
- Differing parental concept of “permission”
 - Formal signed consent vs. public awareness
 - Identifiability of specimens may influence permission



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Strengths



- National sample of parents
- Evaluate relationship between **permission** and parental willingness to permit **use of NBS sample for research**
- Examine parental **support of NBS sample storage** and its relationship to **support of research** with NBS samples



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Implications



- Using NBS samples for research without obtaining permission not palatable to parents
- Educating parents about goals and privacy safeguards of a NBS research program may help alleviate parental concerns



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Somewhat *Unwilling*



- More likely to report all children in good or excellent health
 - vs. very willing: OR 2.1, 95% CI: 1.2 – 3.9;
 - vs. somewhat willing [not significant]: OR 1.8, 95% CI: 0.9-3.5)