Using newborn screening bloodspots for research: public preferences for policy options

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1 **Using newborn screening bloodspots for research:**
2 **Public preferences for policy options**
3

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37

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39 reasoning
40

41
42 **What's Known on This Subject**

43 Newborn screening programs have traditionally used parental opt-out models to authorize their
44 use of residual newborn screening bloodspots for medical research, but public controversy and
45 recent changes to research policy may challenge current practice.
46

47 **What This Study Adds**

48 The public prefers that permission be sought to use NBS samples for research. This preference
49 was diminished but still retained where challenges associated with this option, as well as other
50 policy options, were presented.
51

Contributors' Statement Page:

FA Miller conceived of and led the study. FA Miller designed the data collection instruments, coordinated and supervised data collection and analysis, critically reviewed the manuscript, and approved the final manuscript as submitted.

RZ Hayeems assisted with study design and the design of the data collection instruments, participated in the oversight of data collection and analysis, drafted and critically reviewed the manuscript, and approved the final manuscript as submitted.

CJ Barg, Y Bombard, C Cressman assisted with study design and the design of the data collection instruments, participated in the oversight of data collection and analysis, critically reviewed the manuscript, and approved the final manuscript as submitted.

M Painter-Main conducted the statistical analysis, critically reviewed the manuscript, and approved the final manuscript as submitted.

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Abstract

Objective: The retention of residual newborn screening (NBS) bloodspots for medical research remains contentious. To inform this debate, we sought to understand public preferences for, and reasons for preferring, alternative policy options.

Methods: We assessed preferences among four policy options for research use of residual bloodspots through a bilingual national internet survey of a representative sample of Canadians. Additionally, 50% of respondents were randomly assigned to select reasons supporting these preferences. Understanding of screening and research concepts, attitudes towards medical research, and demographics were also assessed.

Results: Of 1102 respondents (94% participation rate; 47% completion rate), the overall preference among policy options presented was *ask permission* (67%); this was also the most acceptable option (80%). *Assume permission* was acceptable to 46%, *no permission required* was acceptable to 29%, and *no research allowed* was acceptable to 26%. The acceptability of the *ask permission* option was reduced among participants randomly assigned to the reasoning exercise (84% vs 76%; $p=0.004$). Compared to *assume/no permission required*, ordered logistic regression showed significant reduction in preference for the *ask permission* option with greater understanding of concepts ($OR=0.87$, $p<0.001$), greater confidence in science ($OR=0.16$, $p<0.001$), and a perceived responsibility to contribute to research ($OR=0.39$, $p<0.001$).

Conclusion: Members of the Canadian public prefer that explicit permission be sought for storage and research use of NBS bloodspots. This preference was diminished but retained when reasons supporting and opposing routine storage, and other policy options were presented. Findings warrant consideration as NBS communities devise new strategies to respond to shifting legislative contexts.

Introduction

Repositories of residual newborn screening (NBS) bloodspots exemplify the promise and challenge of big data for translational research. They provide unbiased coverage of populations for medical and public health research but raise questions regarding appropriate governance. Policy and practice related to the storage and retention of residual NBS bloodspots for medical research remain variable and contentious.¹⁻⁵ Lawsuits have challenged NBS programs' right to retain samples for research without explicit parental consent, resulting in the destruction of some population-level bio-specimen collections.⁶⁻¹¹ Yet supporters of the 'opt-out' (i.e. non-explicit consent) model argue that it ensures high participation rates and is simply more feasible than seeking explicit permission from the entire population of pre/post partum parents.¹²⁻¹⁴

When engaged in deliberation about the tensions between individual and collective interests, American and Canadian publics are generally supportive of research use of stored samples.^{1,15,16} Where concrete policy options have been proposed, however, majorities favor active permission-based approaches over passive notification/opt-out processes.^{1,17-21} Even genetics professionals perceive risk to secondary uses of bloodspots without permission, including reduced uptake and loss of public trust.²² Seeking to sustain research capacity while recognizing these concerns, recent guidance emphasizes the importance of transparency regarding the storage and research uses of dried bloodspots but remains ambivalent on the question of consent. Botkin et al²³ state, *"Either an opt-in or an opt-out approach for enabling parental choice can be ethically*

126 *acceptable. In either case, parental education and public transparency are central to the*
127 *ethical conduct of the program...*”

128
129 Despite this apparent flexibility, proposed changes to the Common Rule, prompted by
130 the NBS Saves Lives Reauthorization Act of 2014, suggests that US-federally funded
131 NBS research will require explicit consent for use of bloodspots, eliminating the ability
132 of ethics boards to waive consent.²⁴⁻²⁷ Recent guidance from the American Society of
133 Human Genetics also advocates parental choice regarding the use of bloodspots.²⁸ What
134 constitutes informed consent in this context, and how this legislation and professional
135 guidance will affect other jurisdictions, remains unknown. Whether public preferences
136 for explicit consent are robust in the face of practical and value-based challenges
137 associated with consent also remains unknown. To deepen our understanding of public
138 preferences for this complex policy issue and to inform the development of strategies
139 related to storage and use of NBS bloodspots in a shifting legislative context, we report
140 results from a study that engaged the Canadian public on this policy question.

142 **Methods**

143 *Sample and data collection*

144 We conducted a cross-sectional, bilingual (English, French) national internet survey of a
145 representative sample of Canadians in 2013.²⁹ We contracted with Survey Sampling
146 International (SSI), specialists in online data collection for academic and market
147 research. We recruited respondents who met pre-specified criteria for age, sex, and
148 region of residence, in accordance with Statistics Canada data.⁴² We over-sampled

individuals with children <5 to allow sub-group analysis comparing parents of young children with others. Eligible participants were sent an email invitation to participate in the survey by following a link. The survey was comprised of three separate sections to optimize survey completion; respondents were given the option to continue after each section, with incentives (rewards points, prize draws, or cash) dependent on the section(s) completed. University of Toronto's Health Sciences Research Ethics Board approved the study.

The questionnaire

The questionnaire was developed by our multidisciplinary team based on previous work¹⁵ and a literature review.³¹⁻³⁶ It was pretested through three rounds of face-to-face cognitive interviews (n=16) and piloted with members of the internet panel (n=87) to assess comprehension, face and content validity. The questionnaire was designed to educate respondents and measure knowledge and attitudes related to NBS (Sections 1&2), and the use of residual bloodspots for medical research (Section 3). We provided respondents with essential information about this type of research by specifying three minimum conditions: (i) all research would meet strict scientific and ethical standards (i.e. reviewed and approved by an ethics board); (ii) all personal information would be removed to protect privacy (i.e. ID number used for sample identity, linking to personal health records possible but rigorously secured); and (iii) the NBS program would be responsible for deciding what type of research could be done. The survey included professionally-designed images to convey these concepts in a visually appealing way and included interactive quizzes ([Appendix 1](#)) to explain relevant content.

Measures

We solicited preferences for four specific policy options: *no research* (the samples are destroyed after NBS, so that no research is possible), *ask for permission for research* (parents are asked for their permission to store their baby's sample for future research; unless parents agree to research, their baby's sample will *not* be used), *assume permission for research* (parents are *not* asked for their permission to use their baby's sample for future research; permission is assumed but there is a clear process for parents to opt out), and *no permission needed for research* (parents are *not* asked for their permission to use their baby's sample for future research; permission is not considered necessary). We specified that if parents are asked for permission, it would be for all potential medical research rather than for one specific study. We asked respondents to rate the acceptability of each policy option on its own (4-point Likert scale: completely or somewhat unacceptable, somewhat or completely acceptable; Appendix 2a) and we included a forced choice question, asking respondents to identify which of the four policy options they preferred overall (Appendix 2b).

Understanding of bloodspot storage was assessed using true/false quizzes (Appendix 1). After each concept was explained, a quiz assessed understanding, followed by real-time feedback and corrected answers, as appropriate. We also assessed respondents' trust in medical research using two items from the Eurobarometer Social Values on Science and Technology Survey³⁶ (5-point Likert scales from strongly agree, SA to strongly disagree, SD) and one *de novo* item related to research privacy (Table 4). Views about the importance of the public's involvement in governing medical research, the sense of

personal duty to participate in medical research, and views on the responsibility of others to participate (5-point Likert scales, SA to SD) were also solicited.³⁶ We assessed prior awareness that NBS samples could be stored for future research (i.e. samples are currently retained in Canada, albeit with variable consent processes, duration, and research use policies).³⁷

Finally, we drew from Johri et al³¹ to develop a novel approach to the use of value-based reasoning in the elicitation of public preferences. To test the effect of reasoning on policy preferences, half the respondents were randomly assigned to an exercise that presented them with six pro/con reasons, developed by the authors to represent the most salient, countervailing logics driving this debate. Reasons related to the goal of NBS, the challenge of consent, and trust in the research enterprise.³⁸⁻⁴⁰ Three reasons supported routinized bloodspot storage: research is an important opportunity, consent has costs, and medical research can be trusted; three reasons opposed routinized storage: research is not the primary goal of NBS, consent is necessary, and research protections aren't always enough ([Appendix 3a](#)). Participants were then asked to select the most influential reason for each of their judgments about the acceptability of the four policy options ([Appendix 3b](#)) and for their overall policy preference ([Appendix 3c](#)).

Analysis

We used descriptive and multivariate statistics, using chi² tests to assess proportional differences where applicable; one-sided p-values of <0.05 indicated statistical significance. We produced two composite measures: correct responses to the quizzes

were summed to produce the “understanding of screening and sample storage” measure, scored from 0 to 24 with higher scores indicating greater understanding of these concepts. The “trust in medical research” index was scored from 1-15 and normalized on a 0-1 scale indicating less or more trust in research, respectively. We assessed the reliability of these measures; high Cronbach’s alphas (0.70 each) supported their inclusion in the regression model.

To identify the prevailing policy preference and understand the predictors of this preference, including the role of reasoning, we categorized policy preferences as “acceptable,” (completely or somewhat acceptable) or “unacceptable,” (completely or somewhat unacceptable). The proportional odds assumption was met (i.e. OR was the same irrespective of the cut-point between “no permission required”, “assume permission”, and “ask for permission”), so we used ordinal logistic regression to assess the association between socio-demographic characteristics, knowledge, and attitudes and the most popular policy option. Data were managed and analyzed using Stata (v10.1, StataCorp LP., Station College, Texas).

Results

Survey Respondents

Response rates are not typically measured for internet surveys, where recruitment is designed to achieve quotas.⁴¹ The participation rate was 94%, reflecting the proportion of respondents who agreed to participate (n=2345) out of the number of unique individuals who visited the first survey page (n=2499). The survey completion rate was

47%, reflecting the proportion of respondents who completed all three sections of the survey and met pre-determined response quality criteria from those who agreed to participate (i.e., we removed respondents who completed sections in less than the minimum pre-specified time or ‘straight-lined’ through the same response column for blocked items). Of the 1102 respondents who completed sections 1-3, 547 were randomized to receive the reasoning exercise.

Compared with the general population, the sample was better educated and had higher median income (Table 1; Census 2013⁴²). Compared with those who completed only sections 1 and 2 (n=780), respondents who completed sections 1-3 (n = 1102) were more likely to be female ($p < .001$), older ($p < .01$), and score better on the understanding questions ($p < .01$). There were no differences between groups according to the other demographic variables, nor demographic differences between those randomized or not to the reasoning exercise.

The prevailing policy preference

The rating exercise for each policy option indicated that *ask for permission* was acceptable (completely or somewhat) to 80% of respondents, *assume permission* was acceptable to 46%, *no permission needed* was acceptable to 29%, and *no research* was acceptable to 26%. When forced to choose among policy options, *ask for permission* was selected by the largest proportion (67%) and *no research* was selected by the fewest (1%; Table 2).

Reasons associated with policy preferences for those randomized

Table 3 summarizes the relationships between the ratings for each of the four policy options (i.e. individually and forced choice), and the reasons available to participants to justify their choices. Across all policy options, the most frequently selected reason endorsed routinized storage (i.e. *important research opportunity*; up to 85%), whereas the next most frequently selected reason opposed routinized storage (i.e. *consent is necessary*; up to 73%); small minorities selected *consent has costs* (2-10%), *medical research can be trusted* (7-9%), *research is not the goal of NBS* (2-4%), *research protections aren't always enough* (4-11%).

Effect of reasoning exercise

Those randomized to the reasoning exercise found the *ask permission* policy option less acceptable than those not randomized: 76% vs. 84% ($p=0.004$). Exposure to reasoning was not significantly associated with attitudes toward the acceptability of the other three policy options.

Attitudes, understanding, and awareness regarding medical research

The majority demonstrated a strong understanding of screening and storage concepts once introduced to them (mean=22 /24 correct, SD=2.39), though most were unaware that NBS blood samples could be stored for medical research (83%). Most expressed trust in medical research (81-87%) and a majority positively valued public involvement in the governance of medical research (65%). With respect to perceived duties and the responsibilities of others, a majority were disposed to donate their own residual

biological material to medical research (88%) but fewer believed that others were obliged to do so (33%; [Table 4](#)).

Ordinal regression

Respondents with lower knowledge and trust scores were more likely to have selected the *ask permission* policy option (OR=0.88, 95% CI 0.82-0.95; OR=0.14, 95% CI 0.04-0.45, respectively). This option was also more likely to be selected by those who did not believe it a personal duty or a responsibility of others to participate in research (OR=0.34, 95% CI 0.14-0.83; OR=0.40, 95% CI 0.28-0.57; respectively), as well as by those who positively valued public involvement in research governance (OR=1.59, 95% CI 1.36-1.86; $p<0.001$). Demographic factors (including having children), awareness of sample storage, and participating in the reasoning exercise were not associated with this policy preference ([Table 5](#)).

Discussion

In light of ongoing debate related to storage and research uses of newborn bloodspots, our findings provide timely insight. Seeking explicit permission is highly valued, even when respondents are probed, and some of the practical and value-based challenges associated with this approach are exposed. When asked to rate the acceptability of the *ask permission* policy option on its own, the vast majority of respondents (80%) were very supportive. Support for this option remain high (76%) but was significantly reduced among those offered credible reasons for supporting or opposing routinized storage, relative to those not offered such reasons. As well, when forced to choose

among four different policy options, a smaller majority (67%) selected *ask permission* than the 80% who deemed it an acceptable option. Further, the ordered logistic regression suggests that the preference for the *ask permission* policy option was sensitive to respondents' knowledge about and attitudes towards research more generally. Specifically, a greater understanding of screening and research concepts, trust in research, and a perceived responsibility to participate in research were associated with *reduced* support for explicit permission.

These findings endorse that the public prefers explicit consent, extend related research by identifying the robustness of this preference in the face of reasoning, and highlight public attitudes to which this preference may be sensitive. Arguably, these findings leave the NBS community with two policy options. One option would be to align with the apparent majority preference and develop an explicit permission-based model. While the precise method of consent mandated by NBS Saves Lives Reauthorization Act has not been articulated, an explicit consent approach would presumably meet its bar. However, claims related to lack of feasibility and concerns about reduced uptake persist,^{12,14} and suggest that optimizing voluntariness *and* collective public health remains a delicate balancing act. There is evidence to suggest that it is feasible to implement permission-based models.^{17,20,43-45} Of note, Charles et al²⁰ tested this approach using a two-stage written consent protocol. They allowed parents to provide separate consent for (i) their baby to be screened and (ii) secondary use of the sample for research. NBS participation remained above 99% and only 6.5% of parents opted out of research use. However, other consent-based bio-repository models (e.g. Michigan

BioTrust) achieve more moderate uptake (i.e. 60%), raising questions about whether explicit consent compromises the ability of programs to conduct generalizable research.⁴³

A second option might be informed by the results of the logistic regression and the evidence of somewhat diminished enthusiasm for explicit consent among those exposed to the reasoning exercise. Such an approach would mobilize efforts to optimize educational strategies and build trust in order to support a more passive opt-out model. In response to parents' stated preferences for receiving information prenatally about NBS and the use bloodspots for research,^{1,46-47} and the Division of Health and Human Services Secretary's Advisory Committee on Heritable Disorders in Newborns and Children's recommendations regarding these goals,⁴⁸ efforts to develop educational tools are underway. For example, a recent study¹⁴ identified seven things parents want to know about residual bloodspots (i.e. details about storage, potential uses, risks and burdens, safeguards, anonymity, return of results, and parental choice), and suggested that implementing this knowledge base would improve understanding, trust, and acceptance towards retaining and using stored bloodspots. Whether an opt-out model supported by a robust educational process would comply with the notion of choice recommended by the American Society of Human Genetics,²⁸ meet the requirements of NBS Saves Lives Reauthorization Act,²⁷ and be feasible to implement remains to be seen.

We acknowledge several limitations. First, in presenting the policy options, we did not include parental ‘notification’ as a specific policy option or as an element of any of the policy options as we could not guarantee that such would occur. *Ask permission* was, arguably, the only mechanism where parents were involved in an explicit decision. Second, we cannot determine from our findings if respondents’ conceptualized *permission* as equivalent to the notion of consent, itself having a legal connotation. While an explicit link between permission and consent is made for those randomized to the reasoning exercise, this applies to only half of our respondent group. Third, because of the structure of the questionnaire, which considered screening before issues of storage and research, we cannot shed light on the impact storage policy preferences might have on screening uptake. Fourth, the reasoning exercise had a significant impact on the public’s preferences in only one instance; this may be explained by the stability of participant opinion, incomplete ascertainment of considered judgments, the inconsistency of some reasoning, or participant disinterest. A final limitation concerns the structure of our public engagement exercise, which was consultative, rather than deliberative. Recent research suggests the value of deliberative engagements in the context of population screening⁴⁹⁻⁵² but our study suggests that carefully composed internet surveys can also be used to elicit considered judgments, and thereby provide more quantitative data on the strength of preferences and the structure of trade-offs. Our efforts to increase the intensity with which participants considered the issue - using an explicit reasoning exercise - were novel, and our findings make an important contribution to the NBS dialogue and to public engagement methodology.

378 Despite these limitations, we engaged a demographically diverse and representative
379 public in a complex public policy issue using a detailed online training module and a
380 range of strategies to deepen participant involvement. Finally, we reiterate the value of
381 engaging lay publics rather than invested consumers, parents, and professionals since the
382 research at stake is considered to be a public good.

384 **Conclusions**

385 Members of the Canadian public prefer that explicit permission be sought for storage
386 and research use of NBS bloodspots. While enthusiasm for this option was not without
387 qualification, the majority remained committed to an explicit permission model. In the
388 context of a shifting legislative context, these findings warrant consideration.

Table 1: Survey Respondents*

		N(%)
Gender (n=1102)	Female	586 (53.2)
	Male	516 (46.8)
Age	18-35	283 (25.7)
	36-54	446 (40.5)
	55+	373 (33.8)
Marital Status	Married/Common Law	688 (62.4)
	Single/separated/divorced/widowed	414 (37.6)
Geographic Region	Eastern Canada	84 (7.6)
	Quebec	256 (23.2)
	Ontario	420 (38.1)
	Western Canada	342 (31.0)
Community Size	Less than 30,000	282 (25.6)
	30,000-499,999	445 (40.4)
	500,000+	375 (34.0)
Income	Less than \$40,000	358 (32.5)
	\$40,000 - \$79,999	375 (34.0)
	\$80,000 +	287 (26.0)
	Preferred not to say	82 (7.4)
Highest Education Completed (n=1090)	High school or less	347 (31.8)
	College/other post-secondary	400 (36.7)
	University (undergrad/grad)	343 (31.5)
Children	Have children	735 (66.8)
	Planning or pregnant w/o children	111 (10.1)
	No plans to have children	108 (9.8)
	Other**	147 (13.4)
Children under 5	Yes	188 (25.6)
	No	547 (74.4)
Family history of genetic disease	Yes	188 (17.0)
	No	851 (77.2)
	Other (i.e. prefer not to say)	63 (5.7)

*Respondents who completed Section 3 of the survey

**These individuals were ≥ 50 years of age and reported not having children; they were, therefore, not asked whether they were planning to have children

398 **Table 2: Results of policy options rating exercise (N=1102)**
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Policy Options	Acceptability of each policy option N(%)				When forced to choose among policy options
	Completely Unacceptable	Somewhat Unacceptable	Somewhat Acceptable	Completely Acceptable	
No research	418 (37.9)	399 (36.2)	185 (16.8)	100 (9.1)	15 (1.4)
Ask permission for research ^a	59 (5.4)	161 (14.6)	297 (27.0)	585 (53.1)	742 (67.3)
Assume permission for research ^b	282 (25.6)	305 (27.7)	329 (29.9)	186 (16.9)	207 (18.8)
No permission needed for research	524 (47.5)	258 (23.4)	190 (17.2)	130 (11.8)	138 (12.5)

400 ^a Ask permission vs. assume permission $p < .001$

401 ^b Assume permission vs. no permission needed for research $p < .001$

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Table 3: Policy option ratings by reasons selected¹

¹ Sub-sample randomized to reasoning exercise only (n=547)

Policy Option Rating ²	Reason Selected N(%)					
	Reasons that favour routinized storage			Reasons that do not favour routinized storage		
	Important research opportunity	Consent has costs	Medical research can be trusted	Research is not the goal	Consent is necessary	Research protections aren't always enough
No research allowed						
Acceptable n=133 (24)	30 (22.6)	9 (6.8)	15 (11.3)	15 (11.3)	48 (36.1)	16 (12.0)
Not acceptable n=414 (76)	333 (80.4)	1 (0.002)	35 (8.4)	9 (2.2)	33 (8.0)	3 (0.007)
Ask permission for research						
Acceptable n=414 (76)	93 (22.5)	12 (2.9)	19 (4.6)	6 (1.4)	259 (62.6)	25 (6.0)
Not acceptable n=133 (24)	61 (45.9)	21 (15.8)	21 (15.8)	7 (5.3)	20 (15.0)	3 (2.2)
Assume permission for research						
Acceptable n=254 (46)	99 (39.0)	44 (17.3)	39 (15.4)	5 (2.0)	49 (19.3)	18 (7.1)
Unacceptable n=293 (54)	32 (10.9)	11 (3.8)	9 (3.1)	11 (3.8)	198 (67.6)	32 (10.9)
No permission required for research						
Acceptable n=167 (30)	98 (58.7)	16 (9.6)	30 (18.0)	1 (0.006)	15 (9.0)	7 (4.2)
Unacceptable n=380 (70)	19 (5.0)	11 (2.9)	8 (2.1)	14 (3.7)	277 (72.9)	51 (13.4)
Forced Choice						
No research allowed n=4 (0.7)	0 (0.0)	0 (0.0)	0 (0.0)	2 (50.0)	1 (25.0)	1 (25.0)
Ask permission for research n=368 (67.3)	99 (26.9)	5 (1.4)	13 (3.5)	5 (1.4)	224 (60.9)	22 (6.0)
Assume permission for research n=103 (18.8)	65 (63.1)	16 (15.5)	11 (10.7)	2 (1.9)	7 (6.8)	2 (1.9)
No permission required n=72 (13.2)	61 (84.7)	1 (1.4)	10 (13.9)	0 (0.0)	0 (0.0)	0 (0.0)

² “Acceptable” = completely acceptable + somewhat acceptable; “Unacceptable” = completely unacceptable + somewhat unacceptable

Table 4: Respondents' knowledge, awareness, and attitudes related to newborn screening and medical research

Construct	Item	Total (n=1102) n (%)	
KNOWLEDGE^	Index of understanding composite score (max=24), mean (SD)	21.72 (2.39)	
AWARENESS	Aware that blood samples collected for NBS could be stored & made available for (before completing questionnaire)	Yes	191 (17.3)
		No	911 (82.7)
ATTITUDES			
Trust in medical research*	Medical research will help to cure illnesses such as AIDS, cancer	Strongly agree/agree	956 (86.8)
		Neutral	119 (10.8)
		Strongly disagree/disagree	27 (2.4)
	Most medical researchers want to work on things that will make life better for the average person	Strongly agree/agree	918 (83.3)
		Neutral	143 (13.0)
		Strongly disagree/disagree	41 (3.7)
	The privacy and confidentiality of people who participate in medical research will be protected	Strongly agree/agree	891 (80.9)
		Neutral	176 (16.0)
		Strongly disagree/disagree	35 (3.2)
Public involvement in research governance	To direct medical research in the right way, it would be better to take more account of what the public thinks, in other words people like you and me	Strongly agree/agree	718 (65.2)
		Neutral	281 (25.5)
		Strongly disagree/disagree	103 (9.3)
Willingness to participate in research	After my doctor uses my blood sample for my care, I would let my leftover sample be used for medical research	Strongly agree/agree	965 (87.6)
		Neutral	84 (7.6)
		Strongly disagree/disagree	53 (4.8)
Perceived obligation of others to participate in research	After a doctor uses a blood sample for someone’s health care, I think it would be irresponsible for them to refuse to let their leftover sample be used for medical research	Strongly agree/agree	362 (32.8)
		Neutral	367 (33.3)
		Strongly disagree/disagree	373 (33.8)

[^]Index sums together the number of correct responses given to 24 questions (Cronbach's alpha =0.70)*Trust in medical research" index (Cronbach's alpha= .70; 0-1 scale, mean= .80, SD=0.16)

421 **Table 5. Predictors of “Ask for permission” Policy Preference†: Ordered Logistic**
422 **Regression**

	“Ask Permission” Policy Preference Odds Ratio (SE)
Demographics	
<i>Gender (Reference: Male)</i>	
Female (CI 0.74-1.38)	1.01 (.16)
<i>Age (Reference: 55+)</i>	
18-35 (CI 0.81-2.30)	1.36 (.36)
36-54 (CI 0.78-1.64)	1.14 (.21)
<i>Geographic Region (Reference: Quebec)</i>	
Eastern Canada (CI 0.48-1.59)	.88 (.27)
Ontario (CI 0.72-1.57)	1.07 (.21)
Western Canada (CI 0.89-2.04)	1.35 (.28)
<i>Income (Reference: \$80,000+)</i>	
Less than \$40,000 (CI 0.90-2.12)	1.39 (.30)
\$40,000-\$79,999 (CI 0.78-1.64)	1.13 (.21)
<i>Education (Reference: Post-Sec.)</i>	
High-School or less (CI 0.66-1.50)	1.00(.21)
College/CEGEP (CI 0.76-1.61)	1.11 (.21)
<i>City Size (Reference: 500,000+)</i>	
Less than 30,000 (CI 0.75-1.72)	1.13 (.24)
30,000-499,999 (CI 0.52-1.05)	.74 (.13)+
<i>Relationship Status (Reference: Not married/not common law)</i>	
Married/Common law (CI 0.84-1.66)	1.18 (.21)
<i>Children (Reference: Kids Under 5)</i>	
Kids Over 4 (CI 0.49-1.32)	.80 (.20)
Plan/Pregnant, No Kids (CI 0.66-2.33)	1.24 (.40)
No Plan/Pregnant, No Kids (CI 0.38-1.18)	.67 (.19)
Knowledge/Awareness	
Index of understanding ^a (CI 0.82-0.95)	.88 (.03)***
Aware of sample storage for medical research (CI 0.70-1.51) (<i>Reference: not aware</i>)	1.03 (.20)
Attitudes Towards Research	
Trust in medical research ^b (CI 0.04-.45) (<i>Reference: Not agree on trust items</i>)	.14 (.08)***
Public involvement important (CI 1.36-1.86) (<i>Reference: Not important</i>)	1.59 (.12)***
Personal duty to participate (<i>Reference: Neutral</i>)	
Disagree personal duty (CI 0.13-2.07)	.51 (.36)
Agree personal duty (CI .14-.83)	.34 (.16)*
Perceived responsibility of others to participate (<i>Reference: Neutral</i>)	
Disagree irresponsible of others not to donate (CI .92-2.01)	1.37 (.27)
Agree irresponsible of others not to donate (CI 0.28-0.57)	.40 (.07)***
Reasoning exercise (CI 0.67-1.22)	.91 (.14)
Pseudo R-squared	.12
N	914
Test of Proportionality of Odds	24.24 (p>.05)
Brant Test of Parallel Regression Assumption	29.54 (p>.05)

423 † DV is 0=“No permission required”; 1= “assume permission” 2=“ask for permission”

424 +p<.10 *p<.05 **<p.01 ***p<.001

425 ^a Index sums the number of correct responses given to 24 questions (Alpha =0.70)

426 ^b Selected items from the Eurobarometer's Social Values on Science and Technology Survey.³¹ “Trust in Medical Research” index
427 created (Alpha= .70; 0-1 scale, mean= .80, SD=0.16); ~CI – 95% confidence interval

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TEST Mode

9

What do you think about research with newborn screening samples?

➡ **How would research with newborn screening samples work?**

- What are the policy options?
- Reasons to consider
- What do you think?

As you know, a few drops of blood are taken from babies’ heels for newborn screening. After blood samples are tested for disease, leftover samples are kept so that the newborn screening program can use them to make sure the program is working properly. For example, the laboratory can re-test old samples to make sure that they have found the diseases they should have found.

These leftover samples could also be made available for future medical research. Many different kinds of medical research could be done with these samples because they provide information about almost all the babies in a province or country. So far, not a lot of research has been done with these samples in Canada.

Conditions of Research

If research with newborn screening samples is allowed, the following conditions would have to be met.

All research would have to meet strict scientific and ethical standards:

For any research to happen, it would have to be reviewed and approved by a “Research Ethics Board”. Research Ethics Boards are responsible for all research conducted in universities and hospitals in Canada; they usually include physicians, researchers, lawyers, ethicists and members of the public

All personal information would be removed to protect privacy:

Information about the baby (e.g., name, address, etc.) would be removed and a number would be used to identify the sample. It would be possible to link the sample to personal health records using this number, but the linking procedure would be rigorously secured. Data would only be analyzed at group level and nobody who heard about the study would be able to identify who participated.

The newborn screening program would be responsible:

In each province, the newborn screening program would have to decide what type of research could be done.

Next, please answer the following questions about what you have just learned.

	True	False
Leftover samples collected for newborn screening can be used to make sure that the program is working properly.	☐	☐
Leftover samples collected for newborn screening could be useful for many different kinds of medical research.	☐	☐
Researchers working with leftover samples would have access to personal information about babies (e.g., names and addresses).	☐	☐

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TEST Mode

Let's see how you did on those questions.

X 1. Leftover samples collected for newborn screening can be used to make sure that the program is working properly. You said this answer was False.

Actually, this statement is true. Leftover samples collected for newborn screening can be used to make sure that the program is working properly. For example, samples can be re-tested to make sure the results are accurate.

X 2. Leftover samples collected for newborn screening could be useful for many different kinds of medical research. You said this answer was False.

Actually, this statement is true. Because they provide information about almost all the babies in a province or country, leftover samples could be useful for many different kinds of medical research.

X 3. Researchers working with leftover samples would have access to personal information about babies (e.g., names and addresses). You said this answer was True.

Actually, this statement is false. Researchers would not have access to babies' personal information.

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Appendix 2a: Policy option presented to non-reasoning arm: An example

9

What do you think about research with newborn screening samples?

- How would research with newborn screening samples work?
- What are the policy options?
- Reasons to consider
- ➔ **What do you think?**

Ask for permission: Parents are asked for their permission to store their baby’s sample for future research. Unless parents agree to research, their baby’s sample will *not* be used. Is this an acceptable or unacceptable policy option?

(Please check only one.)

Completely unacceptable option	Somewhat unacceptable option	Somewhat acceptable option	Completely acceptable option
☐	☐	☐	☐

Appendix 2b: Forced choice question presented to non-reasoning arm

9

What do you think about research with newborn screening samples?

- How would research with newborn screening samples work?
- What are the policy options?
- Reasons to consider
- ➔ **What do you think?**

Of these four policy options, which do you think is the best?

(Please check only one.)

- ☐ **No research:** The samples are destroyed after newborn screening, so that no research is possible.
- ☐ **Ask for permission:** Parents are asked for their permission to store their baby’s sample for future research. Unless parents agree to research, their baby’s sample will *not* be used.
- ☐ **Assume permission:** Parents are *not* asked for their permission to use their baby’s sample for future research; permission is assumed but there is a clear process for parents to opt out.
- ☐ **No permission needed:** Parents are *not* asked for their permission to use their baby’s sample for future research; permission is not considered necessary.

Appendix 3a: Description of reasons presented to those randomized to reasoning arm

TEST Mode

9

What do you think about research with newborn screening samples?

- How would research with newborn screening samples work?
- What are the policy options?
- ➔ **Reasons to consider**
- What do you think?

People disagree about the different policy options. The following are some important reasons to consider when thinking about policy options for medical research with leftover newborn screening samples:

Important research opportunity

We should take advantage of the research opportunity provided by a program that collects samples from almost all babies born in a province or country. This allows researchers to understand patterns of health and disease in the population.

Research

Research is not the goal

We should not lose focus on the primary purpose of newborn screening, which is to ensure that babies with treatable diseases are identified early. Medical research may be important, but it doesn't have to happen through newborn screening.

Consent has costs

Obtaining consent is a complicated process that takes a great deal of time and money.

Consent

Consent is necessary

Individuals should have a choice about whether or not they want to participate in research, regardless of the costs.

Medical research can be trusted

We have good systems in place to ensure that only high quality research is done, and to protect the privacy and confidentiality of research participants.

Trust

Research protections aren't always enough

Despite all of the protections to ensure that research is high quality, and that the privacy and confidentiality of research participants is maintained, things can go wrong.

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Appendix 3b: Policy option presented to reasoning arm: An example

TEST Mode

9

What do you think about research with newborn screening samples?

- How would research with newborn screening samples work?
- What are the policy options?
- Reasons to consider
 - ➔ **What do you think?**

Ask for permission: Parents are asked for their permission to store their baby’s sample for future research. Unless parents agree to research, their baby’s sample will *not* be used. Is this an acceptable or unacceptable policy option?

(Please check only one.)

Completely unacceptable option	Somewhat unacceptable option	Somewhat acceptable option	Completely acceptable option
☐	☐	☐	☐

What reason had the strongest influence on your answer?

(Please check only one.)

☐ Important research opportunity

☐ Research is not the goal

☐ Consent has costs

☐ Consent is necessary

☐ Medical research can be trusted

☐ Research protections aren’t always enough

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Appendix 3c: Forced choice question presented to reasoning arm

TEST Mode

9

What do you think about research with newborn screening samples?

- How would research with newborn screening samples work?
- What are the policy options?
- Reasons to consider
- ➔ **What do you think?**

Of these four policy options, which do you think is the best?

(Please check only one.)

☐ **No research:** The samples are destroyed after newborn screening, so that no research is possible.

☐ **Ask for permission:** Parents are asked for their permission to store their baby's sample for future research. Unless parents agree to research, their baby's sample will *not* be used.

☐ **Assume permission:** Parents are *not* asked for their permission to use their baby's sample for future research; permission is assumed but there is a clear process for parents to opt out.

☐ **No permission needed:** Parents are *not* asked for their permission to use their baby's sample for future research; permission is not considered necessary.

What reason had the strongest influence on your answer?

(Please check only one.)

☐ Important research opportunity ☐ Research is not the goal

☐ Consent has costs ☐ Consent is necessary

☐ Medical research can be trusted ☐ Research protections aren't always enough

Before you answered this questionnaire, did you know that the blood samples collected through newborn screening could be stored and made available for medical research?

(Please check only one.)

☐ Yes, I knew.

☐ No, I had never heard of it /I only came to know of it through this questionnaire.

← →

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