Public concerns regarding the storage and secondary uses of residual newborn bloodspots: an analysis of print media, legal cases, and public engagement activities

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Abstract Recently, public concerns have been expressed regarding the non-consented storage and secondary research uses of residual newborn bloodspot (RBS) samples. The purpose of this paper is to examine public responses to the storage and secondary uses of RBS that can be identified through analysis of media, legal cases, and documented public engagement activities. Coverage in the examined print media confirmed the importance of RBS to journalists and those people who expressed their concerns to these journalists. Several lawsuits, brought by parents concerned about the storage of newborn bloodspots, placed the practice of storing NBS into the spotlight. This resulted in controversial debates and the mandatory destruction of millions of samples. Analysis of public engagement activities across several jurisdictions indicated that across (inter)national boundaries there are common elements to what is perceived as inappropriate governance of RBS. Public concerns were grouped into five main themes: trust, transparency, confidentiality, ownership, and stigmatization/discrimination. The results of our analysis help to make a compelling case for placing citizens at the center of the debate and developing policy about the storage and secondary uses of newborn bloodspots.

Keywords Residual newborn bloodspots · Storage · Secondary uses · Public concerns · Public engagement activities · Print media

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Introduction

Worldwide, more than 60 countries conduct newborn blood screening (NBS) (Centers for Disease Control and Prevention 2008). The screening involves a quick prick to the baby's heel and a few drops of blood are placed on special pre-printed filter paper (Guthrie cards) that preserves the sample and makes it viable for laboratory use (Maschke 2009). The screening is used for pre-symptomatic detection of rare metabolic, hormonal, functional, and genetic disorders (e.g., phenylketonuria (PKU)), though there is a wide spectrum in the number and specific disorders for which screening is offered by jurisdiction. It is common practice to collect more blood spots on each card than are needed for the screening (i.e., to permit confirmatory diagnosis and retesting if needed), resulting in residual bloodspot (RBS) samples (Government of Michigan Department of Community Health 2010; Maschke 2009).

Currently, the storage of RBS samples varies widely across the jurisdictions that perform NBS (Botkin et al. 2013; Lewis et al. 2012). RBS samples are stored for various reasons (e.g., quality control for the newborn screening program or for improvement of screening tests; secondary medical, clinical, and/or public health research) (Centers for Disease Control and Prevention 2008; Kharaboyan et al. 2004). Some NBS programs strictly monitor the use of RBS and do not allow the samples to be used for purposes other than screening and quality control (Kharaboyan et al. 2004; Therrell et al. 2011). However, many of the NBS programs around the world do not have clear guidance regarding the length of RBS storage and permitted uses of the samples (Burgard et al. 2012; Kharaboyan et al. 2004; Therrell et al. 2011). Clear guidance on this issue is valuable because the storage of RBS samples raises a number of socio-ethical and public policy issues (e.g., concerns about privacy, respect for autonomy, transparency).

Information on newborn screening procedures and policies is usually provided to parents in written form during the prenatal period and/or after their child's birth. In many countries, NBS is conducted without explicit consent from parents because newborn screening is (1) considered part of the routine care for newborns and (2) seen to be in the best interest of the child's health (Drabiak-Syed 2010; Laberge et al. 2004). In many jurisdictions, RBS specimens have been (and are being) stored and used for secondary purposes beyond the original screening without parental knowledge or consent (Kharaboyan et al. 2004). For example, in the 1990s, Michigan researchers used anonymous RBS to find out how many newborns were exposed to HIV (Government of Michigan Department of Community Health 2010). Between 2003 and 2007, the United States Armed Forces DNA Identification Laboratory secretly obtained 800 de-identified RBS samples from the Texas Department of State Health Services (TDSHS) to add to their national mitochondrial-DNA database for forensic identification (Ramshaw 2010; Texas Department of State Health Services 2013).

For many years, there has been scholarly debate in the USA (and other countries) over the issue of informed consent from parents for secondary research involving RBS. The main positions on this topic that have been argued are as follows:

1. The use of RBS for secondary research should be governed by the Common Rule (i.e., when NBS specimens are collected, and when the specimens are matched with identifying information, the infants become human research subjects and parents should be required to provide consent prior to secondary research uses) (Drabiak-Syed 2010, 2011; Suter 2014);
2. Archived RBS samples are similar to community property because everyone benefits from the research; therefore, consent should not be needed to use RBS samples for any research (Drabiak-Syed 2011; Suter 2014);
3. Secondary research using RBS is an extension of NBS and researchers should not need to obtain consent from parents or inform parents about secondary research (Drabiak-Syed 2010, 2011);
4. RBS donors have a right to autonomy. This involves "the notion that individuals may not be treated as merely a means to an end" (Suter 2014, p. 767). There is an expectation on the part of parents who consent to NBS that there is a relationship of trust because involvement in the NBS program involves sharing personal information—a relationship that has an inherent "balance of power" (p. 767). Consequently, researchers have a fiduciary obligation to protect RBS donors and informed consent should be obtained (Drabiak-Syed 2011; Suter 2014);
5. Obtaining informed consent from all RBS donors or their parents is not practical; therefore, it should not be required if the samples are anonymized, de-identified, or the research results using identified RBS samples in no way indicate the identity of the donor and/or the family (Drabiak-Syed 2011; Suter 2014);
6. Although it is believed that obtaining consent from all RBS donors or their parents is not practical, some researchers do this. Therefore, being impractical is not a good enough reason for not obtaining informed consent. Consequently, informed consent from donors or parents should be required for RBS research (Suter 2014);
7. Anonymizing and de-identifying the data may, on the surface, appear to protect the privacy of the donor; however, DNA samples cannot be truly anonymized. That is, RBS samples contain genetic information that are "readily identifiable" (Suter 2014, p. 767) because technology currently exists and is evolving that allows for biological samples to be linked to donors. Consequently, informed consent should be required prior to the use of RBS for any research purposes (Drabiak-Syed 2010, 2011; Suter 2014);
8. There is a distinction between NBS and secondary research utilizing RBS; thus, researchers should be allowed to use RBS for research unless parents opt-out (Drabiak-Syed 2010, 2011; Suter 2014); and
9. "Opting-in" requirements for storage and additional research uses of RBS are more appropriate than "opt-out" requirements because opt-out puts the onus on the parents to take steps to remove their child's RBS from possible future research purposes. Many parents may not understand how to do this and/or the possible consequences for their child's access to future genetic testing if the storage facility does not make a distinction between storage of RBS for future NBS testing and RBS for future additional research (Suter 2014).

Prior to 2005, only a few countries (e.g., Denmark, New Zealand, UK, Germany) had policies in place to obtain informed consent from parents for RBS storage and secondary research (Drabiak-Syed 2010; Laberge et al. 2004; Ramshaw 2010). Recently, public concerns have been expressed regarding the non-consented storage and secondary research uses of RBS samples (e.g., Armstrong 2010; Cohen 2010; Rothwell et al. 2010). The purpose of this paper is to inform developing policy approaches in this area by analyzing the ways in which public concerns have manifested in three important areas. We focus on public concerns with the storage and secondary uses of RBS that have been expressed in numerous countries via (1) print media (i.e., newspaper/magazine articles), (2) legal actions, and (3) public engagement activities that have been conducted on these issues. We conclude with an observation that public engagement on the issue of storage and secondary uses of RBS is a crucial element for ongoing trust relationships between publics and those governing such resources.

Methods

We employed three approaches to examining public concerns in three domains: print media, litigation, and public engagements. Examining these three domains together allows comparisons to be made between domains and a fuller account of public concerns to be presented.

Print media

The *Factiva* newspaper database was used for the search of English language newspaper/magazine reports. This database contains print media from more than 118 newspapers/magazines from 18 countries. *Factiva* was searched from January 1990 to April 2013 using the following keyword combinations: “newborn bloodspots” and “storage” or “retention,” or “newborn blood spots” and “storage” or “retention,” or “newborn screening” and “storage” or “retention.” The search yielded 743 hits (this included approximately 300 articles that were duplicated and published in more than one source). The selection criteria for an applicable article in this section were that it must: (a) be from a newspaper/magazine accessible to the general public; (b) be specific to the topic of the storage and potential uses of RBS; (c) contain information about parents’ and/or the public’s opinion and/or actions relevant to this topic; and (d) be written in English. Articles were excluded if they were (a) a public announcement from a NBS program or health department or (b) an article in an academic journal. Eighty-four articles were selected. The concerns expressed in each article were listed and then categorized according to themes (Braun and Clarke 2006).

Litigation

The *CanLII* and *Lexis NexisQuicklaw International* databases were used in the search of English language legal cases. The *CanLII* database provides access to court judgements and tribunal decisions from all Canadian jurisdictions. *Lexis NexisQuicklaw International* database was used to search for legal cases at the international level. These databases were searched from January 1990 to April 2013 using the same keyword search terms listed above in the print media section. The searches were conducted for cases in the following countries: Canada, USA, UK, NZ, and Australia. The selection criteria for a relevant lawsuit were that it must (a) be from a judicial Court; (b) be specifically related to the topic of the storage and potential uses of RBS; and (c) be written in English. Lawsuits were excluded if they were (a) not related to NBS or (b) not related to storage. Ten legal cases were found. In the USA, six cases were identified. Among these six cases, three were directly related to our topic and two judgments related to the same legal action (Court of Appeal and Supreme Court). In Canada, two cases were identified, with

only one relevant to the topic. In the UK, two cases were found, but they were not selected for our study because they did not address the topic of NBS. No cases were found for NZ and Australia. The relevant lawsuits are listed in Table 1.

Public engagement scholarly literature

The *Primo Central* index was used to search for online academic English language journal articles from January 2003 to April 2013 using the same keyword search terms used in the two previous sections. The search yielded 1,167 articles. The selection criteria for an applicable journal article were that it must (a) be from a peer-reviewed journal; (b) be specific to the topic of the storage and potential secondary uses of RBS; (c) contain information about parents’ and/or the public’s opinion relevant to this topic; and (d) be written in English. Articles were excluded if they were a review, conference proceeding, dissertation, or newspaper article.

Given that *Primo Central* may not identify all the articles relevant to this topic, the reference sections of highly relevant articles were examined and additional articles were located. Further, numerous public engagement activities regarding the storage and secondary uses of RBS were conducted by government departments or private agencies (i.e., gray literature). English language gray literature discussed in the peer-reviewed articles was located and included in this review. Further Internet searches were also conducted using the same search terms. This search yielded 5,060 hits. The selection criteria for an applicable gray literature document were that it must (a) be an official government/agency report; (b) be specific to the topic of the storage and potential secondary uses of RBS; (c) contain information about parents’ and/or the public’s opinion relevant to this topic; and (d) be written in English. Gray literature was excluded if it did not take the form of a report for an official department/agency (e.g., it was a conference or agency PowerPoint presentation), it was the opinion of one person or an advocacy group on a website/blog, it was a proposal for a public engagement activity, or the

Table 1 Lawsuits relevant to storage and secondary uses of RBS

Year(s)	Lawsuits
2009, 2010, and 2011	Bearder vs. State of Minnesota (Minnesota Dept of Health—MDH) Minnesota State District, Appeals and Supreme Courts, USA
2009	Beleno vs. Tex. Dept. of State Health Services San Antonio District Court Texas, USA
2010	Higgins vs. Texas Dept. of Health Services Austin District Court, Texas, USA
2011 and 2012	LD (Guardian ad litem of) vs. Provincial Health Services Authority of British Columbia British Columbia Supreme Court, BC, Canada

public engagement activity was regarding NBS activities and storage of RBS was not included. Examination of all of the possible sources of information regarding public engagement activities relevant to the storage and secondary uses of RBS resulted in 36 articles/documents (three articles are referenced in more than one public engagement activity category).

Results

In 1991, a member of the Danish Parliament was the first to question a government's storage and secondary use of RBS samples (Norgaard-Pedersen and Hougaard 2007). This action sparked a public debate on the issue and the Danish government initiated inspections of its NBS biobank facility by members of parliament and the government's Ethical Council. More recently, genetic technology has "skyrocketed" the research value of RBS (Tarini 2011). For example, researchers successfully amplified DNA and conducted genome-wide gene expression profiles using RBS (Hollegaard et al. 2013; Khoo et al. 2011). Researchers have also successfully extracted and amplified RNA from stored unfrozen samples almost a decade old (Haak et al. 2009). Many public health officials, researchers, ethicists, and policymakers around the world have expressed concern regarding the ethical, legal, and social implications of using RBS specimens for research unrelated to the NBS process (e.g., Norgaard-Pedersen and Hougaard 2007; Rothwell et al. 2010, 2011; United Kingdom Newborn Screening Programme Centre 2005; University of Michigan Health System 2011). Importantly, some parents of children whose RBS are being stored, and in some cases used for purposes beyond the initial screening, have also expressed strong concerns (e.g., Carmichael 2011; Armstrong 2010; Cohen 2010; Botkin et al. 2012; Rothwell et al. 2012; Tarini et al. 2009). This section will summarize public concerns identified in (1) print media, (2) lawsuits, and (3) public engagement activities.

Print media

Public concerns regarding this issue were newsworthy in several English-speaking countries (i.e., Canada, USA, New Zealand (NZ), Australia, UK, and Ireland). The main themes identified in these news reports are as follows: transparency, respect for autonomy, confidentiality, ownership, and stigmatization/discrimination.

- i. **Transparency:** Concerns were expressed from all of the abovementioned countries about the secrecy surrounding the decision by many of the NBS governing bodies to store the samples (e.g., "Why isn't it more publicized? It just seems like they're being awfully sneaky about it" (Roser 2009)). In the USA and the UK, concerns about secrecy

were coupled with quotes from parents explicitly claiming a breach of trust by the NBS governing bodies (e.g., "I'm horrified that they would breach my trust, keep my child's sample for years on end" (Driver 2010)).

- ii. **Respect for autonomy:** In all of the countries, concerns were expressed about the NBS governing bodies making decisions on behalf of parents regarding the type of secondary research purposes that are acceptable. Quotes from parents stress that they want to be asked for their explicit consent before any secondary research using RBS is conducted (e.g., "This is my DNA; it's not yours. Ask me if you want to use me for some project" (Roser 2009)).
- iii. **Confidentiality:** Concerns were expressed in the USA, Australia, NZ, and Canada that stored RBS may not be safe and that breaches in privacy may occur (e.g., "Once we start using DNA for more and more things like regular medical records, somebody could do a cross-check and say whose blood it is" (Stein 2009)). In the USA, NZ, Australia, Ireland, and the UK, concerns were expressed that some NBS facilities are selling and/or giving away RBS specimens to be used in research beyond screening (e.g., "Some people fear a future government might, for example, sell the DNA information on the cards to a drug company" (McLoughlin 2005)).
- iv. **Ownership:** In the USA, NZ, Australia, Ireland, and the UK, some parents were reported to express concern that RBS are considered the property of the NBS governing bodies (e.g., "We do not believe our DNA becomes government property" (Collin 2011)). This concern was coupled with concern about unidentifiable future research uses of RBS (e.g., "My concern is they might not be able to do much with it right now, but 10 years from now? They could do a lot with it the way technology is going" (Roser 2009)).
- v. **Stigmatization/Discrimination:** Concerns were expressed in news reports in Canada, the USA, NZ, and the UK that the genetic information obtainable from RBS may be used by third parties to stigmatize or discriminate against people with certain genetic predispositions or markers (e.g., "I'm worried that findings from research targeting certain geographic areas or other populations might lead to stigmatization or other harms" (Nickel 2004)).

Legal actions

In the USA and Canada, some public concerns on this topic escalated to legal actions (see Table 1). The underlying rationale for these lawsuits was the fact that leftover bloodspots were stored unbeknown to parents. Similar arguments were raised across the lawsuits: violation of privacy, failure to obtain a fully informed consent, and misrepresentation (Knoppers et al. 2012). As a result, in Texas alone, 5 million

dried bloodspots were destroyed (*Beleno v. Tex. Dep't of State Health Servs* 2009).

Public engagement activities

Public concerns such as those identified in print media and via lawsuits regarding the storage and secondary research uses of RBS have led some NBS programs and their governing bodies to consider ways in which to respond to public concerns and regain public trust in NBS programs (Camahan 2011). To this end, several public engagement projects have been conducted in the past decade. The term “public engagement” tends to be used to describe different types of activities (Rowe and Frewer 2005). In our paper, we use the term very broadly to denote a solicitation of public opinion and/or feedback via such activities as surveys, focus groups, community forums, and workshops. In this section, (A) lists the public engagement activities relevant to this topic that have occurred in the past decade (see Table 2) and (B) summarizes the concerns expressed during these public engagement activities.

(A) Public engagement activities

Table 2 illustrates that public engagement activities regarding the storage and secondary research uses of RBS have been conducted on local, national, and international levels. The public engagement projects we reviewed were conducted in the following jurisdictions: Canada, USA, Australia, NZ, European Union (EU) Member States, and the Asia Pacific Region (some projects were conducted in these larger jurisdictions; others in smaller jurisdictions within the larger ones). The public engagement projects have taken several different forms, from surveys and focus groups to more involved designs (e.g., a series of advisory committee meetings that produced a white paper that was made available to the general public for comment—public comments were incorporated into the paper (Therrell et al. 2011); a combination of focus groups, surveys, and knowledge network panels (Botkin et al. 2012); a diverse deliberative jury of citizens (Fleck et al. 2008).

(B) Concerns identified by the public engagement activities

It is evident from some of the public engagement activities reviewed that not all of the participants in these engagement activities had concerns about the storage and possible secondary uses of RBS (e.g., Canada (Bombard et al. 2012), USA (Fleck et al. 2008), China (Gong et al. 2012)). That is, there was a segment of the population of these engagement participants that did not express concerns about the storage and secondary uses of RBS. It is therefore important to recognize that public concern in each jurisdiction involved in the public engagement activities is not unanimous. However, within and

Table 2 Public engagement activities regarding RBS storage and secondary uses

Activity	Jurisdiction
Surveys	<ol style="list-style-type: none"> 1. Perth, Western Australia (Davey et al. 2005) 2. Canada (National) (Avard et al. 2006^a) 3. New Zealand (National) (Newborn Metabolic Screening Programme Advisory Group 2008) 4. Chicago, IL, USA (Neidich et al. 2008) 5. United States (National) (Tarini et al. 2009) 6. Michigan, USA (Gehring et al. 2009) 7. Japan (National) (Fujii et al. 2010) 8. Michigan, USA (Duquette et al. 2010) 9. Canada (National) (Richer et al. 2011) 10. Beijing, China (Gong et al. 2012) 11. TX, CO, UT, NM, AZ, MT, and NV, USA (Botkin et al. 2012^a) 12. European Union Member States (Burgard et al. 2012)
Advisory committee meeting	<ol style="list-style-type: none"> 1. USA (National) (Therrell et al. 2009) 2. USA (National) (Benkendorf et al. 2010) 3. USA (National) (Drabiak-Syed 2010) 4. Oregon, USA (Oregon Health Authority 2011) 5. USA (National) (Therrell et al. 2011^a)
Roundtable	<ol style="list-style-type: none"> 1. USA (National) (Olson and Berger 2010) 2. Michigan, USA (Chrysler et al. 2011)
Consultative workshop/ Working group	<ol style="list-style-type: none"> 1. Europe (12 countries) (Godard et al. 2003) 2. England, Northern Ireland, and Wales (United Kingdom Newborn Screening Programme Centre 2005) 3. Canada (National) (Avard et al. 2006^a) 4. Minnesota, USA (Minnesota Genetic Information Work Group 2009) 5. European Union Member States (Cornel et al. 2012) 6. Asia Pacific Region (Padilla and Therrell 2012) 7. USA (National) (Botkin et al. 2013)
Public webinars	<ol style="list-style-type: none"> 1. USA (National) (Therrell et al. 2011^a)
Focus groups/ community forums	<ol style="list-style-type: none"> 1. New Zealand (National) (deBonnaire et al. 2007) 2. Salt Lake City, UT, USA (Rothwell et al. 2010) 3. Mountain States Region (AZ, CO, MT, NM, NV, TX, UT, and WY), USA (Rothwell et al. 2011) 4. Ann Arbor, MI, USA (Kardia 2011) 5. Michigan, USA (Duquette et al. 2012) 6. Toronto, ON and Montreal, PQ, Canada (Bombard et al. 2012) 7. TX, CO, UT, NM, AZ, and OR, USA (Botkin et al. 2012^a; Rothwell et al. 2012) 8. Salt Lake City, UT, USA (Rothwell et al. 2013)
Living room forums	<ol style="list-style-type: none"> 1. Washington State (National) (leRoy et al. 2005)
Reconvened group discussions	<ol style="list-style-type: none"> 1. New South Wales, Australia (Muchamore et al. 2006)
Public deliberative juries	<ol style="list-style-type: none"> 1. Lansing, Michigan, USA (Fleck et al. 2008)

^a These studies were included in more than one public engagement activity category

across all of the different public engagement activities, it is very clear that there is a segment of the population of participants that do have concerns.

In most of the jurisdictions that conducted quantitative public engagement activities (i.e., surveys), a main finding reported was that participants expressed low support for extended storage and secondary research uses without prior parental consent (e.g., Canada (Richer et al. 2011); Australia (Davey et al. 2005); Japan (Fujii et al. 2010); Beijing, China (Gong et al. 2012)). In contrast, significantly higher support was given for storage and secondary research when questions were prefaced by providing parents with information and obtaining explicit consent. Other areas of concern reported in most of these jurisdictions were possible privacy breaches (e.g., Australia (Davey et al. 2005); Chicago, USA (Neidich et al. 2008)) and research not intended to directly benefit the health of the community (e.g., cloning; Chicago, USA (Neidich et al. 2008)).

Although survey research is technically a form of public engagement, participants are restricted in the type of information they can provide on an issue. In contrast, during qualitative public engagement activities (i.e., all of the public engagement activities except surveys), participants are afforded more freedom to express concerns and provide justifications for their concerns. The outcomes from qualitative public engagement activities provide richer data. The remainder of this section will focus on describing the main qualitative findings.

Initially, we summarized the main concerns presented in each qualitative public engagement reviewed. We then identified, across the public engagement articles, six key topics discussed by the authors: transparency, respect for autonomy, confidentiality, ownership, equity, and stigmatization/discrimination. Although we have divided the concerns into discreet categories, there is overlap between them. Due to space restrictions, we will provide only a few examples of the jurisdictions where participants discussed these concerns. Although most of the public engagement activities we reviewed discussed to some degree elements of each of the six key topics, some projects went into more detail than others.

Transparency A common finding reported in the public engagement studies we reviewed was that participants who were parents did not remember anyone asking them about doing NBS, or they remember being asked (and even signing a form), but no one explained that the RBS would be stored and used beyond the original purpose (e.g., Canada (Bombard et al. 2012); NZ (deBonnaire et al. 2007); Australia (Muchamore et al. 2006); Washington, USA (LeRoy et al. 2005)). In these studies, the general lack of knowledge about the possible practices related to NBS was attributed to insufficient

transparency of the process. In some public engagements, participants cited media stories in which NBS governing bodies were discovered to secretly possess, have sold, or given access to RBS without the knowledge of parents or donors (e.g., Australia (Muchamore et al. 2006)). Suspicions of conspiracy theories were discussed in a few studies in which participants suggested that the governing bodies were doing something wrong that required them to hide their actions from parents and the general public (e.g., Canada (Bombard et al. 2012); Australia (Muchamore et al. 2006); NZ (deBonnaire et al. 2007)).

Insufficient transparency in the NBS process was associated with lack of trust in the NBS governing bodies during some public engagements (e.g., Canada (Bombard et al. 2012); EU Member States (Cornel et al. 2012); Mountain State Regions, USA (Rothwell et al. 2011)). Lack of trust was primarily discussed in relation to a belief that research conducted using RBS may not always be in the best interest of the child and/or the health of the community. Rather, the research may be conducted for the sake of profit and/or to advance government or private agendas (e.g., law enforcement efforts, profit).

Respect for autonomy A main concern regarding respect for autonomy identified in many public engagements was the consent processes or lack thereof (e.g., Canada (Bombard et al. 2012); NZ (deBonnaire et al. 2007); EU Member States (Cornel et al. 2012); Michigan, USA (Chrysler et al. 2011)). In many jurisdictions (e.g., Canada, China, California), consent is not considered necessary for the collection, storage, and possible uses of newborn blood samples (Carnahan 2011; Gong et al. 2012; Avarde et al. 2006). A concern that emerged in many of the public engagements we reviewed was that the lack of obtaining explicit consent made respect for autonomy impossible.

Participants in some jurisdictions expressed concern that the opt-out consent model, currently used by many NBS programs, hindered the possibility of respect for autonomy in two ways. First, the opt-out consent model was believed to impede informed consent efforts in that there is no incentive for NBS programs to ensure that parents are informed of RBS storage and possible uses (e.g., USA (Botkin et al. 2013); Michigan, USA (Fleck et al. 2008)). Further, the opt-out consent model was viewed in some jurisdictions as coercive (e.g., Canada (Bombard et al. 2012); EU Member States (Cornel et al. 2012); Michigan, USA (Fleck et al. 2008)). That is, NBS screening programs in many jurisdictions (e.g., Canada, most states in the USA, most countries in the EU) do not differentiate between opting out of storage/secondary uses relevant to clinical screening research (i.e., screening for various genetic conditions) and general research (Carnahan 2011; Bombard et al. 2012; Cornel et al. 2012; Chrysler et al. 2011).

This can be seen as a flaw in the use of this consent model, in that if parents opt-out of having their child's RBS stored, this may be taken as stored for any reason. Specifically, in some jurisdictions parents may not be provided with the option of having their child's RBS stored for future clinical reasons (e.g., new tests for genetic conditions) while opting out of storage for general research purposes (e.g., pharmaceutical or health research unrelated to NBS). Participants in some of the public engagement activities reviewed believed that parents may agree to storage because they do not want their child to be possibly disadvantaged should new genetic testing be developed (e.g., Canada (Bombard et al. 2012)).

Second, participants in some of the public engagements reviewed expressed concern that parents who do not opt out of storage for their child's RBS may be considered by NBS programs to have consented to allow whatever uses deemed appropriate by NBS governing bodies (e.g., Canada (Bombard et al. 2012); Michigan, USA (Fleck et al. 2008); NZ (deBonnaire et al. 2007)). According to Camahan (2011), this policy has been implemented in Texas subsequent to the court order to destroy stored RBS. Some participants in the public engagements we reviewed believed this logic to be flawed in that not opting out does not equate to giving consent. It was suggested that if NBS programs operate under the assumption of parental consent through not opting out, they may be less likely to be accountable for the research choices they subject RBS to in the future.

In most of the public engagement projects reviewed (e.g., Canada (Bombard et al. 2012); Michigan, USA (Fleck et al. 2008); EU Member States (Cornel et al. 2012)), participants were asked if they thought that NBS programs should begin obtaining written informed consent from parents for storage and secondary research uses. Across the public engagements, we identified three main concerns regarding obtaining written consent.

Are parents and donors who were not given the option to consent any less entitled to give consent than parents today? According to participants in some jurisdictions, starting to obtain consent from parents from the present point forward ignores the millions of parents who were not given the option to consent or withdraw from RBS storage and secondary research (e.g., Michigan, USA (Chrysler et al. 2011; Fleck et al. 2008); EU Member States (Cornel et al. 2012)). This was seen as an ethical and equality issue. Further, in some jurisdictions, RBS have been stored long enough that donors are now adults; adults who are probably unaware that their genetic material is being stored and possibly used in various research projects (Michigan, USA (Chrysler et al. 2011); EU Member States (Cornel et al. 2012)). This raises the question:

What is "informed" consent? Participants in several of the reviewed public engagement activities made the point that

passing parents a brochure and/or having them sign a consent form shortly after their child's birth (when they are occupied with the care of their child) may be ineffective and inadequate (e.g., Canada (Avard et al. 2006); USA (Therrell et al. 2011); EU Member States (Cornel et al. 2012)). This point raises the question if (or when) NBS programs obtain consent from parents post-natal for storage and possible secondary uses of RBS, is it "informed" consent? "Uninformed" consent can leave NBS programs open to the possibility that, at a later date, parents may claim that they did not understand the information, or they were unaware giving consent included storage and secondary research (e.g., Mountain States Region, USA (Rothwell et al. 2011); Australia (Muchamore et al. 2006)).

What are parents being informed of with regard to possible future research? In several jurisdictions, it was pointed out that the exact nature of future medical and/or environmental research is unknown (e.g., Canada (Bombard et al. 2012); Michigan, USA (Fleck et al. 2008); NZ (deBonnaire et al. 2007)). Further, currently, no one is certain of all of the risks and benefits of future research for individuals or part or all of the populations who have RBS stored in various jurisdictions. Nor could anyone truly determine the possibility of offending cultural, religious, and/or personal values of donors and/or their families (e.g., USA (Chrysler et al. 2011); NZ (deBonnaire et al. 2007)).

Confidentiality Important concerns regarding confidentiality to emerge in the public engagement activities we reviewed were security issues, misuse of information, and breaches in privacy. Concerns regarding security measures in place to protect RBS specimens were discussed in several of the reviewed public engagements (e.g., Canada (Bombard et al. 2012); USA (Therrell et al. 2011); EU Member States (Cornel et al. 2012); Australia (Muchamore et al. 2006)). Participants involved in these public engagements argued that even if the persons conducting the research using RBS do not misuse the information that can be obtained from DNA samples, the samples and associated information may still be misused by others. Across these public engagements, the common point made was that no system of storage for the physical RBS specimens and the electronic and hardcopy information pertaining to the specimens is impenetrable. This makes donors' genetic information vulnerable to being accessed without donors' permission or knowledge. Security breaches open RBS donors and their families to possible breaches in privacy. It is possible that unauthorized access to personal donor and family information pertaining to RBS could be used to facilitate such crimes as identity theft (Australia (Muchamore et al. 2006)). Thus far, there have not been any reported

cases of security breaches in NBS facilities (Kilakkathi 2012). However, there have been some recent breaches in security and privacy in other types of biobank facilities. As such, according to Kilakkathi (2012), investigations of the security policies and procedures at several NBS storing facilities demonstrated a need for more rigorous controls. Another security issue presented in some of the public engagement jurisdictions was that even if parents requested to have their child's RBS samples returned or destroyed after screening, not every NBS facility guarantees that all of the information pertaining to RBS specimens will also be destroyed (e.g., hardcopy and electronic copies of results from testing, samples distributed to third party researchers, and their subsequent findings) (e.g., Australia (Muchamore et al. 2006)).

Ownership In many NBS jurisdictions, RBS are considered property of the NBS governing body (or local government) (e.g., European countries (Godard et al. 2003); most American states (Fleck et al. 2008; Minnesota Genetic Information Work Group 2009); Australia (Muchamore et al. 2006)). In some of the reviewed public engagement activities, participants expressed concern that the current ownership policy gives preference to the rights of the general public over those of the individual donor (Fleck et al. 2008). Specifically, participants were concerned that because NBS is mandatory in the USA, individuals forfeit any rights to and control of the RBS sample of her/his genetic material once it is collected during the screening process. According to Fleck et al. (2008), some deliberative jurors argued that this violated their rights regarding the ownership of their own DNA. Across the public engagement activities we reviewed, numerous participants stated that parents have a right to retain ownership of their child's blood samples and control over who has access to the specimens (e.g., Canada (Bombard et al. 2012); USA (Rothwell et al. 2010, 2012); NZ (deBonnaire et al. 2007)).

One justification for parental ownership of RBS samples offered in some of the public engagement projects was respect for family/donor cultural and personal values (e.g., Canada (Bombard et al. 2012); Michigan, USA (Chrysler et al. 2011); New Zealand (deBonnaire et al. 2007)). For example, New Zealand Aboriginal peoples consider that parts of a person that have been removed from the body (e.g., blood, tissues) are still part of that person and remain her/his property (deBonnaire et al. 2007)). Therefore, the individual needs to ensure that whatever purposes the body parts are used for are consistent with her/his personal values. According to this perspective, it would be disrespectful and inappropriate for someone other than the donor or a direct family member to make a decision regarding the use of the donor's body parts (including blood samples).

Another argument evident in the examined studies is for the right of individuals to have ownership of personal information about themselves. For example, during the public deliberative juries in Michigan, USA (Fleck et al. 2008), DNA was likened to information such as credit card numbers. That is, given that each person's DNA is unique, it is a personal identifier just like a credit card number. Participants argued that just because you throw away a receipt with your credit card number on it does not mean that anyone finding that number is allowed to use it for their own personal use. RBS are valuable to researchers because they contain DNA. Even if samples are anonymized, the fact that they contain DNA makes the samples identifiable to those who know how to access the genetic information and who have genetic samples that are identified to compare to RBS specimens.

Across the public engagements we reviewed, most authors reported that participants considered some types of research uses acceptable for RBS (e.g., quality control purposes, research that benefits the health of children and more generally the community) (e.g., Canada (Avard et al. 2006); NZ (deBonnaire et al. 2007)). Other research uses were considered to be unacceptable. For example, human cloning (e.g., EU Member States (Cornel et al. 2012); NZ (deBonnaire et al. 2007)), eugenics, genetic research in which human and animal DNA are combined (NZ (deBonnaire et al. 2007)), any research that may result in discrimination by third parties such as insurance companies and employers (e.g., Canada (Bombard et al. 2012); USA (Therrell et al. 2011); NZ (deBonnaire et al. 2007); EU Member States (Cornel et al. 2012)), for profit research by pharmaceutical and biotechnology companies, commercially oriented research, government or third party DNA databases that may be used for secret research purposes (e.g., Canada (Avard et al. 2006); Australia (Muchamore et al. 2006)), and paternity tests (unless ordered by a court; Australia (Muchamore et al. 2006)). There was also concern expressed in some jurisdictions about the use of RBS in unknown future research (e.g., Canada (Bombard et al. 2012); NZ (deBonnaire et al. 2007); Australia (Muchamore et al. 2006)).

Equity In some public engagements, concerns were expressed about the differences in specimen retention periods across different jurisdictions and what this might mean for equitable treatment of RBS donors (e.g., Australia (Muchamore et al. 2006); EU Member States (Cornel et al. 2012)). For example, at the reconvened group discussion held in Australia (Muchamore et al. 2006), participants asked whether some families are getting more services than others (e.g., benefit of additional NBS research), and are some being exploited more than others (e.g., being involved in more unsanctioned research) in the NBS facilities where RBS are stored for lengthy periods of time or indefinitely? Further, during some public

focus groups held in New Zealand (deBonnaire et al. 2007), participants asked whether some donors/families were disadvantaged because their RBS specimens were given out to third parties for secondary research purposes unrelated to NBS screening, leaving fewer samples available for future screening and personal uses (e.g., identification of missing person, personal requests for NBS retesting)?

Stigmatization and discrimination In several public engagements, participants expressed concerns that donors and/or their families may be at risk of being stigmatized or discriminated against due to the type of secondary research for which RBS may be used (e.g., Canada (Bombard et al. 2012); Michigan, USA (Fleck et al. 2008); NZ (deBonnaire et al. 2007); Minnesota, USA (Minnesota Genetic Information Work Group 2009)). For example, if it was determined that a particular racial or ethnic group had a higher predisposition for a genetic condition that was costly to treat, individuals from that group might not be able to obtain health, life, or disability insurance (or they may pay higher rates than people from other groups) (e.g., Canada (Bombard et al. 2012); NZ (deBonnaire et al. 2007); Washington, USA (LeRoy et al. 2005)). Another concern expressed was that some employers may not hire individuals from this racial or ethnic group for possible health-related reasons (Utah, USA (Rothwell et al. 2010)). In one jurisdiction, participants proposed that if it is found that babies born in a particular geographical area have a higher concentration of an environmental toxin in their blood, although this would be useful public health information, it may also adversely affect the housing prices in that area (Michigan, USA (Fleck et al. 2008)).

Discussion

The practice of storing newborn bloodspots specimens has existed in some form or other for over 50 years. Historically, this was perceived as a routine course of action to facilitate confirmation of screening results, to monitor and assure quality assurance of the newborn screening program and, in some cases, to confirm a postmortem diagnosis (Bombard et al. 2012). Although RBS have been used for purposes other than the initial screening, more recently an increasing ability of researchers to use RBS for genome-wide gene expression profiling has resulted in a greater demand and appreciation of RBS as a valuable resource for health research. This demand has made salient a range of ethical challenges associated with secondary uses of RBS, which have also found expression in public concern against storage and unconsented secondary uses of RBS. While these challenges have been

recognized, it has been argued that discussion surrounding RBS has been too focused on legal and ethical issues (Tarini 2011), at the expense of ignoring the public policy issue of whether parents have been given an adequate voice in whether and the ways in which samples should be used for secondary purposes. Any solution to this argument requires, in part, a deeper understanding of the concerns that have already been expressed by members of the public in different contexts. The main purpose of this paper has therefore been to examine public responses to the storage and secondary uses of RBS that can be identified through analysis of the media, legal cases, and documented public engagement activities.

The results of our analysis help to make a compelling case for placing citizens at the center of the debate and developing policy about the storage and secondary uses of newborn bloodspots. First, coverage in the print media that was examined confirmed the importance of RBS to journalists and those people who expressed their concerns to these journalists. While obviously mediated, these concerns could be grouped into five themes: trust, transparency, confidentiality, ownership, and stigmatization/discrimination. With some variations, these themes *also* emerged from analysis of public engagement activities across several English-speaking jurisdictions, which indicate that across (inter)national boundaries there are common elements to what is perceived as inappropriate governance of RBS.¹ The public engagement activities reviewed acknowledged that not all participants had concerns regarding the storage and additional uses of RBS; however, we were not able to clearly ascertain jurisdictional differences regarding which issues were of no concern in the different jurisdictions.

Arguably, custodians of NBS programs and, in particular, of RBS collections need to address the specific issues in their revision of policies and governance structures if they are to gain and maintain public trust. Second, several lawsuits, brought by parents concerned about the storage of newborn bloodspots, placed the practice of storing NBS into the spotlight. This has resulted in controversial debates and the mandatory destruction of millions of samples. Irrespective of one's stance on privacy, this destruction presents the loss of a unique resource for health research. Because the bloodspots are collected from the vast majority of newborns in a given jurisdiction (sometimes over several decades), these RBS represent an unmatched resource for research and public health. For

¹ This study was limited to English-speaking jurisdictions. We would like to acknowledge the suggestion of a reviewer to compare our findings from analysis of this issue in English-speaking media, legal cases, and documented public engagement activities to those available in other common languages (e.g., French, Spanish). Although this research would be a valuable addition to the literature on the topic, it is outside the scope of this paper. Consequently, our analysis is limited in this regard and our analytical claims extend only to English language jurisdictions. However, a comparison of perceptions on this topic across different language jurisdiction is a possible avenue of future empirical study.

example, they are an invaluable resource for epigenomic research (Willyard 2012) and could be used in the future to predict epigenetic effects on common diseases (Royal College of Physicians of Ireland 2013). The potential research uses of RBS are limited only by the amount of residual dried blood on the card (Institute of Medicine of the National Academies 2010). Further losses might be prevented if the relationship between RBS custodians and their communities can be improved. Third, public engagement activities from five jurisdictions, which included surveys, focus groups, and other deliberative strategies, in general showed that respondents/participants expressed low support for extended storage without parental consent. However, the findings also suggest that many would support secondary research use of NBS if they had been informed that leftover bloodspots were stored and possibly used for such activities as health research.

To the best of our knowledge, few countries have integrated public engagement into policy regarding the storage of newborn bloodspots. However, efforts to bring the “public voice” into the policy development process are increasing (Oklahoma State Senate 2010; Provincial Health Services Authority (British Columbia-Canada) 2010; Swedish Ministry of Health and Social Affairs 2010; Sénécal et al. 2013). For example, in the UK, the use of extensive public consultation has been endorsed. In 2009, the Michigan Department of Community Health founded the Michigan BioTrust for Health to ensure the preservation of RBS for biomedical research (Langbo et al. 2013). Findings from the Lansing, Michigan deliberative jury (Fleck et al. 2008) informed the development of the BioTrust’s ethical guidelines for the storage and additional use of RBS. Research has determined that the BioTrust has managed to maintain public support and integrity of the NBS program, while avoiding an increase in refusals of NBS (even with the increased negative publicity related to storage and additional research uses of RBS) (Langbo et al. 2013). The BioTrust is a clear example of the benefit of incorporating the “public voice” into the development of policy for the storage and use of RBS.

Nevertheless, most studies, as well as the public concern evident in media and lawsuits, still demonstrate a perceived lack of transparency on the part of parents and the broader public, and a lack of understanding of parental fears as well as a lack of respect for parental decision making on the part of policy makers. It is critical that public policies addressing the storage and use of NBS receive as broad a public endorsement as possible. At the very least, this requires transparency and public communication with detailed information about the storage of RBS; the valuing of individual autonomy by involving parents in decisions about the storage of NBS and their use for research; and public education about the value of newborn bloodspots for research. In addition, it is important to recognize the heterogeneity of individual and community values in formulating policy on RBS. Exemptions may need

to be considered for individuals or communities whose values are different from those of the majority in any given jurisdiction. Complications will inevitably also arise in cases where RBS are used for research across jurisdictional boundaries. In such cases, meaningful public engagement processes will be even more important to avoid public concern and ongoing controversy.

Adopting the principles of the *Universal Declaration on Bioethics and Human Rights* (United Nations Educational, Scientific and Cultural Organization 2005), or other international instruments about the retention and use of newborn bloodspots, forms a critical component of the development of any policies in this area. In addition, to avoid these instruments becoming paternalistic measures that implicitly position publics as passive subjects requiring protection, meaningful public engagement processes are required. Our hope is that by involving publics more directly in decisions about NBS retention and storage, and by developing policies that are sensitive to public concerns, there will be benefit both on the side of increased public trust and greater facilitation of health research.

Compliance with ethics guidelines The manuscript complies with the Canadian research ethics guidelines as set out by the 2nd edition of Tri-Council Policy Statement. No human subjects were involved. Only publicly available documents were analyzed.

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