Informing the Debate

Stored Blood Spots

Ethical and Policy Challenges

Authors
Leonard M. Fleck, Ph.D.
Ann Mongoven, Ph. D.
and
Sarah Marzec, MA

MICHIGAN STATE UNIVERSITY

Institute for Public Policy
and Social Research

Institute for Public Policy and Social Research at Michigan State University

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Authors
Leonard M. Fleck, Ph.D.
Center for Ethics and Humanities in the Life Sciences
Michigan State University

Ann Mongoven, Ph.D.
Center for Ethics and Humanities in the Life Sciences
Michigan State University

Sarah Marzec, MA
Center for Ethics and Humanities in the Life Sciences
Michigan State University

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Communications Assistant

MICHIGAN STATE UNIVERSITY

The Institute for Public Policy and Social Research is housed in the College of Social Science at Michigan State University.

EXECUTIVE SUMMARY

For the past 35 years or so Michigan has been collecting and storing blood spots taken from newborns shortly after birth. Blood is taken in order to test for a number of inherited metabolic disorders that can be fatal if not treated shortly after birth. Taking this blood was mandated by law as a public health measure. Consequently, parents were not asked to give explicit informed consent. Parents perceived this as a routine blood draw. Sometimes a nurse or technician might offer a brief word of explanation as to why the blood was taken.

Michigan initially felt that storing these blood spots for 21.5 years was reasonable. Other states have stored these blood spots for as little as a few months to just a few years. In the past the blood spots were saved with the thought they might have clinical value to the patient from whom they were drawn. But the completion of the Human Genome mapping project and the subsequent explosion of medical research around genetics has given new value to these blood spots as a medical research resource. In the late 1990s the Michigan legislature permitted the Michigan Department of Community Health to expand the length of time for storage of these spots. It also permitted medical research to be done with these spots as long as they were de-identified (so there was virtually no risk of privacy violations). An obvious ethical and policy question with this decision was that neither the parents of the infant, nor that infant, who could be an adult now, gave informed consent for this research to be done with their blood spots. However, maybe explicit consent was not needed. Maybe consent could be presumed because of the public good that would be served and minimal risks posed by this research to any individual.

The Center for Ethics at Michigan State University with the help of an IPPSR grant [Institute for Public Policy and Social Research, MSU] convened a diverse deliberative jury of citizens from mid-Michigan to explore this issue (and a range of related issues) over three meetings. On some matters there was considerable diversity of judgment, which would suggest a need for broader efforts at community education and engagement on the range of relevant issues. In brief, the major outcomes from the deliberative jury project were:

(1) Jurors desired to discuss current policy for taking bloodspots for newborn screening before they addressed possible guidelines for research use of the bloodspots. At the same time, they suggested the need to separate issues of consent for screening and consent for research use. The jurors were significantly split on whether, contra current policy of required screening, parents should give explicit informed consent for the screening blood draw. The majority thought “no” but a significant minority (31%) thought “yes.”
Certain consensuses underlay this disagreement. Jurors agreed that all babies should be screened for their own benefit; that current education about screening is inadequate; and that there should be an improved education process for expectant parents. The significant minority who preferred an informed consent requirement hoped it would insure that an education process would take place and expected that appropriately educated parents would consent (whereas others feared babies would be lost to screening if consent required).

(2) The vast majority of jurors (87%) supported the generic idea of medical research being done with the blood spots, but something close to that that same majority (82%) wanted parents to have the option to "opt out" of allowing their child's blood spots to be used for any research purposes. They wanted some process of explicit informed consent for research uses of the blood spots.

(3) At present the blood spots are stored on cards in a warehouse in Lansing without climate control. Michigan is considering creating a BioTrust as a repository for the relocation and management of these blood spots. The deliberative jurors felt there ought to be a Community Advisory Board (in addition to the IRB and a Scientific Advisory Board) to assess proposed research projects from the perspective of community values and reasonable public purposes. The Community Advisory Board would be the head of a broad network of more localized or regional boards that would be responsible for community engagement around larger policy issues related to the use of the blood spots in the future and the large policy issues related to the functioning of the BioTrust (such as the degree to which for-profit enterprises could have access to the blood spots). The Community Advisory Board would be broadly representative of the state in terms of race/ ethnicity, economic status, geography, religious orientation, age, professional background, civil liberty concerns. There was virtual unanimity on all these points. We did not have time to discuss in detail the extent to which for-profit enterprises should be seen as being among "good public purposes." Roughly 61% of the group were initially inclined to endorse this idea whereas 33% would disagree with that idea.

Leonard M. Fleck, Ph.D.
Center for Ethics and Humanities in the Life Sciences
Michigan State University

STORED NEWBORN BLOOD SPOTS: ETHICAL AND POLICY ISSUES

In the late 1960s states within the US (and many other countries around the world) began to do what is known as "universal newborn screening," primarily as a result of the work of Dr. Robert Guthrie, a microbiologist. The term "universal" refers to the fact that *every* newborn infant was screened by taking a small sample of blood. Infants were being screened for rare metabolic inherited disorders that were treatable and that needed to be treated immediately in order to avoid devastating health consequences (an early death or profound neurological disabilities). Phenylketonuria (PKU) was the first of these disorders. PKU is an autosomal recessive inborn error of metabolism. This disorder has an approximate incidence of one case in 15,000 births. Infants with this disorder are unable to process phenylalanine, the consequence of which is severe mental retardation, seizures, and a number of other neurological problems. [The reader may go to the following Web site to see a long list of common foods that contain phenylalanine, along with the quantity found in a normal serving: http://www.healthyeatingclub.com/info/books-phds/books/foodfacts/html/data/data2e.html.] If these infants are put on a very special diet shortly after birth, these problems can all be avoided. This is the beginning of the set of ethical and policy issues we will address in this essay.

I. Newborn Screening Tests: Which Ones and Why?

In the US several hundred infants are born each year that would be vulnerable to PKU. What we need to emphasize is that the dietary intervention designed to address PKU is completely effective in preventing the devastating consequences of this disorder. It is difficult to imagine any caring rational parent refusing the "heel stick" that takes the few drops of blood needed to do this screening. Consequently, unlike what is common in the rest of medicine where parents are asked to give informed consent for any medical procedure done to their children, no such consent process has been part of doing this blood draw. Parents are often (not always) told why blood is being drawn, but they are not asked to give permission. From the perspective of public health authorities this seems reasonable since there are no risks to the child from the blood draw and this intervention clearly protects the best interests of each (potentially vulnerable) child.

Over the next several decades a small number of other screening tests were added as medical research identified more of these metabolic disorders that were rare and that needed prompt medical intervention to prevent otherwise devastating health consequences for that infant (if diagnosis were left to the time when clinical symptoms manifested themselves). Cost was an issue when it came to adding more tests to the screening procedure since each test for each disorder was distinct. But in the 1980s a new technology was introduced, tandem mass spectrometry, which permitted doing dozens of these tests all at once. It took a while for this technology to be widely disseminated but by the turn of the century this was accomplished. In the span of just a few years the number of conditions for which we screened jumped from about 10 to about 50. Those inherited conditions now included endocrine and hematologic disorders, along with expanded metabolic disorders. Here in Michigan we screen for 49 disorders at this writing, but this number varies considerably from one state to another. In 2004 some states screened for as few as three disorders while others screened for more than fifty (Arn, 2007; Therrell et al., 2006; Alexander and van Dyck, 2006).

Some professional groups have been disturbed by the lack of uniformity across the states because of the potentially devastating differential consequences this can have for infants

depending upon the state in which they happen to be born (Alexander and van Dyck, 2006). What physician would want to say to parents (and what parents would want to hear from a physician), "Your baby would be alive and healthy today if she had been born in Michigan instead of Indiana or Kentucky?" Consequently, the American College of Medical Genetics now urges all states to screen for at least twenty-nine of these disorders and identifies twenty-five other disorders that states might include for universal screening (Arn, 2007). Why, we might ask, should the recommendation not be for all states to screen for all fifty-four of these disorders? It might appear that this was a good way to protect the best interests of all infants. However, the science behind these tests suggests such expansive testing might be more problematic than would first appear (Green at al., 2006; Baily and Murray, 2008). Here, in brief, is the core problem.

From an ethical perspective, making newborn screening *mandatory* for PKU was clearly morally defensible because of both the certainty of the disastrous medical consequences and the availability of an effective therapeutic response that would prevent the worst effects of that vulnerability. However, for some number of the newborn screening tests that have been added to the panel over the past several decades we have neither the certainty of the disastrous medical consequences nor the assuredness that an effective therapeutic response was available and affordable.[6] One of the things we have discovered of late is that (as with many medical disorders that are genetically linked) there are degrees of expression, both in terms of a span of time and degrees of seriousness. Often there are co-factors associated with disease expression that are unknown or very poorly understood. Hence, an infant might test positive for one of these disorders, but then the disorder might not actually manifest itself *for this infant* until the third, fifth or sixth decade of life (or never). If the relevant therapeutic intervention were very cheap and very safe (no matter what the actual disease expression might prove to be), then we could in good conscience provide the therapy no matter what. However, some of these *therapies* will prove deadly for infants not imminently vulnerable to certain disorders.

The other large issue that has become more problematic is the issue of false positives connected to the screening process. Approximately 4,000 infants each year in the US are identified correctly as being afflicted with one of these very rare disorders.[7] However, it is estimated that 12,000 false-positive results are obtained as well.[8] We know that these are *false*

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[6] Moyer et al. (2008) write: "For most other abnormalities identified through tandem mass spectrometry, logic and a close reading of the report [of the American College of Medical Genetics, 2006] reveal much greater uncertainty about the incidence of these abnormalities and their natural history of individuals identified through screening. Given this fact, there is inevitably less direct evidence of benefit and more uncertainty about the health consequences of treatment. Left unscreened, some children might never have known about their conditions because they might never have developed symptoms" (at 37).

[7] This is a very crude number. It is extremely difficult to get a more accurate number because each year some states add more tests and identify more infants with one or another of these disorders. In the period 1995-2005 on average nineteen tests were added by each state to their screening panels (Tarini et al., 2006).

[8] This is again a very crude number since false positives are not routinely reported. These tests have extraordinarily high specificities. However, because four million infants are screened each year by anywhere from twenty to fifty tests, tiny differences in specificity can yield large numbers of false positive results. To illustrate, Tarini et al. (2006) note that if the specificity of each test is 99.995% (very optimistic scenario) then there would be 2575 false positives each year in the US. The medium case scenario with a specificity of 99.95% would yield about 25,644 false positives. The worst case scenario with a specificity of 99.9% would yield 51,059 false positive results. See Gurian et al. (2006) for a discussion of the effects of false positive results on parents.

positives because subsequent alternative testing strategies will establish this fact. Correcting this error may take anywhere from several weeks to several months. Needless to say, this would cause considerable unnecessary anxiety in the minds of these parents. We might be tempted to treat this outcome somewhat casually, perhaps thinking that a little anxiety is a small price to pay to save those other 4,000 infants. But the anxiety is not a trivial matter. Parents in these circumstances are faced with the immediate decision of whether or not to start some treatment needed to save the life of the infant or to prevent irreversible neurologic damage when it might turn out that the treatment itself could be harmful if their infant is not a true positive.[9]

We would like to believe that in an age of scientific medicine all of these proposed newborn screening tests have a strong scientific basis that establishes their reliability and utility. And we would expect to have an equally reliable scientific understanding of the risks and benefits associated with any therapeutic intervention employed in connection with a positive test result. In some cases we have very strong scientific evidence in both these respects. But in other cases the scientific evidence has gaps and uncertainty that might not be readily correctable (Botkin et al., 2006). Much of this deficiency will be related to the very small number of infants who exhibit any one of these disorders. We can often make very reliable, very finely discriminating medical scientific judgments when we have hundreds of thousands of patients per year with this form of cancer or that form of heart disease. That level of refinement is not possible when only 20-50 infants per year might manifest a specific disorder. Perhaps prudence would warrant not including some of these tests in state-mandated panels when evidence of their utility and reliability is somewhat marginal. However, in some cases, advocacy groups have been organized around some of these rare disorders. If such groups have strong and effective leadership, along with monetary resources, then they can generate political pressure for including such tests in these state-mandated panels. A prominent example of this phenomenon would be Krabbe's disease, in which case a star NFL football player had a child who died of this disease, which prompted him to create a foundation to find a cure for this disorder (and develop a test to identify at-risk infants at birth). See: http://www.medicinenet.com/script/main/art.asp?articlekey=54315

This situation raises some ethical and political challenges. We could be morally confident in the 1970s that mandatory screening for PKU without specific informed consent was a morally legitimate option. Today we cannot be as morally confident in that conclusion for the reasons given above. Should we then adjust our moral compass a bit and require specific informed consent for newborn screening? Should state policy also require this, since there are now some identifiable (not insignificant) risks associated with newborn screening as opposed to only clear benefits for all concerned? An adequate informed consent process might be impractically time-consuming, especially if we fairly think about what might have to be conveyed to parents.

If we put that concern aside for the moment, are there other moral costs associated with seeking explicit informed consent for newborn screening? The short answer would seem to be

[9] Botkin et al. (2006) write: "Indeed, some children with benign conditions were seriously harmed from unnecessary restrictions in their diets." The reference is to the PKU diet. See also Baily and Murray, 2008. We should add that for Baily and Murray their other concerns are related to cost-benefit ratios and justice issues at the social level (as opposed to the clinical level). Resources used to achieve very small benefits (as might well be true for some number of these newly introduced screening tests) are not available for other services needed by infants that might be much more beneficial.

that many parents who were just somewhat risk-averse might say, "There is only a very remote chance that our baby would have any of these disorders. Why risk the anxiety of a false positive result? Skip doing the newborn screening." Let us suppose that no more than 1% of parents reacted this way. That represents 40,000 unscreened infants, which would mean (statistically) that about 40 infants with one or another of these disorders would slip through and suffer the irreversible adverse consequences of whichever disorder was missed. Should we (citizens of a responsible and caring society) accept that as part of the social cost of respecting the right of parents to make informed choices or informed refusals regarding newborn screening? Or should screening remain mandatory on the grounds that it is in the best interest of these infants?

The reader should note the use of the plural to refer to "infants." From a collective point of view, we can easily defend the claim that mandatory screening protects the collective best interests of infants. Parents, however, have only their own infant to be concerned about, though the reality is that they have only *aggregate* data regarding screening risks and benefits, which is to say they have insufficient information to judge what might be the *actual best interest of their baby*. Some writers have argued that when it comes to making policy decisions regarding newborn screening some third-party interests ought to be accorded moral weight. Two examples of what these writers have in mind would be the following: (1) Universal newborn screening would alert parents to the risks they would pose to future possible children unless they chose alternative reproductive options. (2) Universal newborn screening saves parents from endless and frustrating medical odysseys when their children would otherwise present with symptoms too non-specific to direct a physician's thought to the very rare disorders with which they are actually afflicted.

The conclusion this is supposed to warrant is that mandatory newborn screening without specific informed consent is reasonable and warranted. Other writers will dissent from that conclusion concluding instead that "these points (are) insufficient to justify mandated public health screening of all newborns" (Moyer et al., 2008, at 35). Some may dissent, not because of a risk/benefit analysis to infants, but rather because of a felt intrinsic importance to informed consent. Alternatively, others might dissent because of skepticism about whether the duty to inform (which may stand independently even when consent is legitimately constrained) will be maintained when formal informed consent is not required.We will return to these issues below as they relate to perceived connections and disconnections between the clinical uses of newborn screening and research use of the bloodpots.

II. Stored Newborn Blood Spots: May They be Used for Research?

The other large issues we need to address are ethical and policy concerns around the use and storage of these blood spots after screening has been done. After the heel stick five drops of blood are deposited on "Guthrie cards" for analysis. Only a tiny punch from one of these spots is needed to do this analysis. The extra spots were available in case additional analysis needed to be done over a period of weeks or months. Some states permitted the destruction of these cards after a few months because their original purpose had been adequately met. Other states preserved these cards for various periods of time ranging from less than a year (30 state programs) to more than twenty years (See Appendix 2 in Therrell et al, 2006 for a complete breakdown by state [attached].). Michigan initially required storage of these blood spots for 21.5 years. But that limit was lifted in 2000 by the state legislature, and authority was given to the Michigan Department of Community Health to determine the length of time for storage be-

yond 21.5 years. What motivated the lifting of that limitation was the rapidly approaching completion of the mapping of the Human Genome. The realization among medical researchers was that three million stored blood spots here in Michigan represented a veritable treasure trove of medical information in the era of genetic medicine.

Research done on these stored blood spots in the latter third of the twentieth century focused on quality control regarding newborn screening, the development of new and better assays, and some epidemiologic studies. This kind of research raises virtually no concerns regarding either violation of individual rights or threats to public interests. However, in the era of genetic medicine all will recognize that those blood spots contain the DNA of individuals. Consequently, depending upon the nature and results of the research, some risk exists that the privacy rights of individuals or their families could be threatened, especially if negative health information about an individual became available to an insurer or an employer. The risk of this happening might be reduced to nearly zero if the blood spots themselves were completely and permanently anonymized. (However, some people raise questions about whether absolute de-idenfication is possible, given the uniqueness of human DNA.) But complete anonymization would greatly reduce the value of the blood spots for a broad range of medical research.

Moreover, in some circumstances, the future possible welfare of some individuals could be compromised. The type of scenario we have in mind might look like this: In the course of doing cancer genetics research with these blood spots it might be discovered that individuals with a very specific genotype were much more vulnerable to some specific cancer later in life than others with a different version of that genotype. Further, specific health measures could be taken to prevent the actual expression of that genotype so long as that cancer had not become clinically manifested. But no way would exist of contacting the individual who was the source of that blood spot if that blood spot had been completely and permanently anonymized. A middle way does exist for dealing with this sort of problem. The blood spot could be linked to a unique numerical identifier which in turn was linked to identifying information for that individual securely locked away with very restrictive access, so that violations of the privacy rights of that individual would be an extremely remote risk. This might be a reasonable approach to the scenario we sketched above. However, variations of the scenario will again create more complications.

We described a situation in which an early and effective intervention was available in response to being identified as the bearer of a specific cancer-prone genotype. We believe that the vast majority of individuals who might be vulnerable in that way would want to know this genetic fact about themselves so that they could take advantage of that intervention. But what if the only available early preventive intervention had only a 50-50 chance of making a difference, or worse, a 30-70 chance of making a difference? In this latter scenario it is far from obvious that there was only one reasonable response to the availability of this information, namely, embracing it. Some individuals might prefer not to know because of the anxiety or other debilitating negative feelings such information might trigger. That is not obviously an irrational response. That choice seems worthy of respect as well, as opposed to foisting the information on an individual.

What it seems we need here is a policy response that is sensitive to the rights and interests at stake. If we believe it is of utmost importance that we be sensitive to the privacy rights and autonomy rights of individuals, then we might say that individuals (or parents) should al-

ways be given the opportunity for informed consent or informed refusal for the use of their blood spot for any kind of medical scientific research. After all, this is (for good reason) the required practice for medical research generally. However, one reason why this practice is not seen as being especially onerous is that the patients involved in research are "right there" to discuss the research and its potential risks and benefits for them. In the case of research with the stored blood spots the "patients" who were the original source of those blood spots could be literally anywhere in the world (given the highly mobile nature of our society), especially if ten or more years have elapsed since the spots were first collected. It could take an enormous amount of labor and economic cost to track down those individuals, so much so that it seems the vast majority of researchers would refuse to undertake research that required this. This would represent the loss of some quantity of potentially socially valuable medical research. So choices about alternative de-identification processes may raise questions about the extent to which the state is or is not obligated to turn research intended for population-level health toward individual health benefit—in addition to questions about the preference of research participants.

This must sound very vague to the reader, but it is not possible to be much more specific than this, especially if researchers faced with this obstacle would not even bother to try to conceptualize some of these research projects. What makes this research so potentially valuable is that tens of thousands or hundreds of thousands of these blood spots could be quickly and efficiently analyzed for their medical knowledge value in a brief period of time. This is not something that could be as readily accomplished if researchers had to deal with tens of thousands or hundreds of thousands of individual persons. The basic moral and political argument that would seem to support this view of the researchers is that the blood spots are anonymous to them, that their work presents no risks to individuals as such.

The rationale at the end of the prior paragraph has been persuasive to a large majority of state legislatures that have explicitly addressed the issue of research uses of these stored blood spots. To be precise, approximately half the states have not addressed research uses of these blood spots. This might be because they store the blood spots for too brief a period of time, six months or less in the case of 24 states (Therrell et al., 2006). About 19 states address in legislation the conditions under which medical research may be done with these stored blood spots. (In a handful of other states policies exist that govern the use of these residual blood spots, but these policies have not been legislatively enacted.) In all cases, the understanding is that these blood spots have been de-identified for research purposes. If the research requires linkage back to identifiable individuals, then the uniform requirement is that explicit informed consent must be obtained from those individuals or their guardians. Three states would seem to be exceptions among the nineteen that address research uses of the blood spots. Pennsylvania and Nebraska require written consent from parents for any medical research use of these blood spots; California allows parents to refuse permission for the use of these blood spots for any medical research (Goldenberg and Martinchek, 2007). Presumably, the main opportunity to express this refusal is at the time the blood is drawn and parents are told of future possible uses of those blood spots during a storage period.

We might imagine that a reasonable and relatively easy resolution to the ethical and policy issues associated with research uses of these dried blood spots would be to solicit informed consent from parents for de-identified uses of the stored blood spots. But there are two problems with this apparently easy resolution. First, in the case of Michigan and comparable states

that have stored these blood spots for ten or more years, getting consent from the current crop of parents of newborns does nothing to legitimate the use of the blood spots that have been stored for all these years (whose original sources would require substantial costs and effort to track down).[10] Second, even for the current crop of parents of newborns we would have great difficulty justifiably speaking of "informed" consent being obtained. What precisely could those parents be *informed* about? No one would have a clue as to what future medical research projects might be proposed in connection with those blood spots. Nor could anyone say what the risks and benefits might be of those future possible research projects, either for the population at large or any sub-type of the population of patients. Nor could anyone say whether any of this future possible research might be offensive to the core values of any religious or cultural or ethnic group. So it seems like it would be disingenuous to refer to such a process as seeking *informed* consent.

No doubt a large number of Americans today are generally supportive of medical research and see such research as generally improving the well being of all in our society. Such individuals could then *consent* to having the stored blood spots of their children be used for medical research *in general*. However, we ought to wonder about the moral or legal status of *consent* that is this open-ended (usually called "blanket consent"). What would we think of parents who told a baby sitter that they should feel free to discipline their child in "whatever way they judged necessary" should the child misbehave? Parents who gave that sort of open-ended permission for discipline would generally be judged to be irresponsible, or at least thoughtless. The California approach might have some relevance here. Parents could be offered the option of refusing permission for the use of the stored blood spots for any sort of medical research. That makes moral sense since parents have no moral obligation to make these blood spots available for research purposes, no matter how noble or benign.

What this approach also means is that parents who have not "opted out" have not in any way affirmatively consented to any or all medical research uses of these blood spots. They have simply "not objected in the present" to future possible research uses of those blood spots, in effect, reserving the right to voice those objections as relevant to specific future research projects. Further, it is not morally or legally necessary that only the individual persons attached in some way to those blood spots would have the right to voice those objections. That right can be transferred to some collective body that might have responsibility for assessing the moral or social legitimacy of specific research uses of those stored blood spots in the future when the relevant information would be available for making an intelligent assessment of the research from both moral and scientific perspectives. This approach has the virtue of avoiding what might otherwise be a meaningless "consent" process at the time of birth for future research uses of these blood spots.

There is another perspective that requires some critical comment regarding consent for research uses of these blood spots. Some states say explicitly in legislation that these blood spots become the property of the state once they have been obtained in the course of newborn screening. This assertion would seem to obviate any need for informed consent for the medical or scientific research uses of these blood spots. The argument might be that the blood is drawn for a reasonable and legitimate public purpose; and consequently, parental consent is irrelevant.

[10] There are eight states that store these blood spots for an indefinite period of time and six more states that store these spots for more than twenty years (Therrell et al., 2006).

However, the clear public purpose for which the blood is drawn is the newborn screening. Very few parents have any awareness at all that the blood has been stored or that it is now valuable material for medical research. [11] Such research might fulfill a legitimate public purpose, but such research might also be morally or socially controversial. The political philosopher John Rawls has called attention in his writing (1971; 1993) to a central source of legitimacy in a democratic society, what he refers to as the "publicity condition." Policies and practices that are intended to be justified as fulfilling a public interest must themselves be publicly visible, transparent and available for public critical assessment. That condition might not be adequately satisfied if the existence and potential uses of these blood spots are effectively hidden from the public. This will be especially true if there are multiple reasonable values in conflict with one another in connection with some specific bit of medical research with these blood spots.

The state might attempt to justify legally its claim that these blood spots are public property by invoking the outcome of the *Moore* case in California (Rao, 2007). Three states [California, Maine, and Washington] explicitly declare through law or regulation that the blood spots are property of the state (Therrell et al., 2006). Mr. Moore was a patient with a genetically distinctive form of cancer of the spleen. He needed surgery to remove the spleen to address the cancer issue. His physician was also a cancer researcher who was able to cultivate a medically and scientifically valuable cell line from Moore's cancerous spleen cells. He did this without informing Mr. Moore. Moore eventually discovered that this was occurring and demanded a share of the profits from the sale of what had become both economically and scientifically valuable. He claimed that he had not consented to this use of his spleen, which was his property. However, the California Supreme Court eventually ruled that Mr. Moore no longer had any property rights in his spleen because it had been alienated from him through the surgery, to which he had freely agreed. The same is legally true for items any of us might throw out in the garbage. The spleen that was removed was now regarded as medical waste which his surgeon was free to appropriate. This is the legal analogy the state might invoke to justify its claim that these blood spots are now public property. However, there are limits to the legitimacy of this analogy. We can illustrate the point with the following example.

I might throw into the garbage my credit card receipts with my credit card numbers intact. Someone going through that garbage might thereby "obtain" my credit card numbers. But no one believes that they now have a right to my credit card, that they can justifiably use that information to purchase goods on the Internet for which I would be charged. That information remains private; that is information that I alone may legitimately control. The same might be said with regard to the blood spots. What makes them medically valuable is the DNA information they contain. [12] But that information is private; I have not given anyone else the right to use

[11] If parents are told that these blood spots are stored for prolonged periods of time and they might be available for research of various kinds, few parents at the time of birth would be motivated to "take in" or reflect on that information. The relevance of the information to their lives and any practical decisions would be too remote. The result of this is that the existence of these stored blood spots (and their use) is effectively hidden from the general public. But this is the sort of situation that can be exploited by someone who discovers this fact and believes the state is deliberately trying to hide something from the public (a secret eugenics agenda). And this is precisely what happened in Minnesota where Twila Brase organized a vocal group of privacy advocates who became strong critics of the entire newborn screening program as "involuntary genetic testing" (Yee, 2007).

[12] Therrell et al. (2006) note that the DNA in these blood samples is very stable over long periods of time under a range of storage conditions. The biological material that was the direct focus of the original screening tests is much less stable, so it is much less likely this would be reliably available for research much beyond a year after collection had occurred.

genetic information about me simply because they might physically possess something that at one time was biologically part of me and contains my DNA. My DNA would be very much like my credit card information. If we take the implications of this analogy in the strictest possible way, then that would mean there would be no access to these blood spots without explicit informed consent. As we noted already, however, the practical implication of this would be that virtually no one would be willing to invest the time or energy or money necessary to get these permissions; and consequently, there would be a loss of what might have been socially and medically valuable information, especially in connection with those blood spots that have been stored for years. This outcome seems neither reasonable nor desirable. What should we do?

In addition to issues regarding individual autonomy, privacy, and vulnerability, research use of the bloodspots may raise issues regarding group vulnerabilities. Could certain well-intended research questions, or certain research results, harm or stigmatize certain subgroups of the population? What groups might be, or might perceive themselves, to be vulnerable? Racial, ethnic, or socioeconomic groups? Or others? Vulnerable to what harms? Even what counts as "harm" may be greatly influenced by cultural location. For example, in several historical cases Native American tribes have expressed concern that proposed research with genetic material might: challenge their origin myths (if genetic tracing of migration patterns suggested different origins) and thereby undermine their land claims; stigmatize them with disease associations; or might undermine tribal authority (Mayo Clinic, 2007, p. 6).

To consider another example, imagine that the bloodspots were used to analyze the presence of a certain environmental toxin in the population and it thereby became clear that infants from a small geographic area of the state had unusually high concentrations in their blood. That might be useful public health information, but it also might make property values in that region plummet. (Of course, whether such a scenario is possible depends on whether zip code data is stripped in the "de-identifying" process. In this example, both pros and cons of more or less rigorous de-identification are apparent.) Policy-making processes thus may require mechanisms to "hear" group concerns as well as concerns to respect individual research participants.

III. Policymaking and the Public:

Controversial Policy Issues and Democratic Deliberation

An increasingly common phenomenon in our society will be policy debates that have as a central element conflicting value perspectives. We are not referring to values that might be regarded as being relatively superficial. Rather, we have in mind values that are deep and central to differing ways in which different social groups choose to live good lives. The debate about embryonic stem cells and whether there ought to be public funding to support research aimed at maximizing the therapeutic potential of those cells would be perfectly illustrative of this point. Many of us would greatly value medical research aimed at curing or alleviating life-threatening medical problems. Research with embryonic stem cells suggests enormous therapeutic potential. But those cells must be derived from embryos that have been grown to the 100 - or 200-cell size. Others in our society are profoundly disturbed by what they regard as casual disregard of these earliest stages of human life. They are especially distressed (morally speaking) that their dollars (tax dollars) would be used to fund research that would violate their deep moral commitments. How does this happen? What should we do?

Political scientists would explain that these conflicts come about because we are a liberal, pluralistic, tolerant democratic society. That is, our core political principles commit us to

trying to maximize the liberty of each individual (or social group) to choose a life plan they find satisfying and reasonable so long as that choice does not violate the equal rights of others to make such choices and so long as no public interests are threatened by those choices. In being politically respectful of a broad diversity of value commitments and priorities among those commitments we will necessarily generate value conflicts when it comes to framing public policies with which we all must live. Further, the state is supposed to remain "neutral" among these differing value perspectives. So what should we do?

We should certainly *not* seek to use majoritarian power to impose public policies on minority groups that would be deeply morally distressed by such choices. That would show a lack of basic moral respect. On the other hand, simply shouting at one another and generating destructive rancor (as we have seen regarding the abortion issue) is hardly a better alternative. If the abortion issue were correctly regarded as being an isolated social aberration, then there might be a way of politically managing the issue that minimized threats to the integrity of our democratic practices and our social fabric. But the kinds of value conflicts reflected in the abortion issue are rapidly becoming ubiquitous, very often because of advancing medical technologies in all areas of medicine. Certainly this is something that is true with regard to newborn screening and the storage of these blood spots. Tandem mass spectrometry has made possible a very rapid expansion of the number of newborn screening tests that can be done. So-called "chip technology" will permit doing literally thousands of tests for genes or gene variants in an infant all at once (if we were to choose,as a society, to pursue that extraordinarily expansive form of newborn testing). Likewise, the explosion in genetic knowledge and genetic technologies has given enormous scientific value to research with those stored blood spots that was non-existent prior to 2000.

One thing we have noticed so far is that there is considerable variation among the states with regard to the details of newborn screening, including the number of screening tests, what parents are told, what is done with the blood spots after testing, who has access to these stored blood spots and for what purposes. This diversity is not a phenomenon peculiar to the US. We would find a comparable degree of diversity among the nations of Europe and the provinces of Canada (Therrell and Adams, 2007). What this suggests is that these differences in policy and practice are not obviously unreasonable, misguided, or driven by ideological zeal. We also see significant differences in the recommendations that come from various professional groups that have taken it upon themselves to weigh in on the ethics and policy issues related to newborn screening. This suggests two things: First, there are reasonable differences of expert opinion on a range of issues related to newborn screening among physicians, research scientists, and public health officials. We might reasonably conclude from this that no specific expertise is likely to yield a single "best" answer regarding what specific policies and practices would yield the most in terms of scientific knowledge or medical/ public health good for society as a whole. Second, the same will be true with regard to the range of value trade-offs that might be at stake in connection with a range of policy options and practices. How important is it, relatively speaking, to maximize medical and scientific knowledge of various kinds from research on the stored blood spots, as opposed to being careful to respect the genetic privacy of patients, or to avoid inadvertently inflicting harm on these newborns (or their future possible selves), or to respect the autonomy rights of those who were the sources of the blood spots (who may or may not want those spots used for research for reasons that might be personal or religious or cultural)? Quite obviously, judgments have to be made. Mo matter the option all are value-laden beliefs

or morals. Choosing, for example, to let the blood spots just stay stored and remain unused means the loss of potentially valuable medical knowledge with broad public health consequences. So, if choices have to be made (and are made), then who should have the moral or political authority to make such value-laden choices? Individuals with various professional backgrounds and kinds of expertise certainly are more "knowledgeable" than the average citizen, but it is not obvious that their expertise gives them special authority to make these value-laden decisions that affect many others who would make very different decisions were they given the option.

The argument we have made elsewhere (Fleck, 2008) to address situations such as this (where we must have a policy decision, where there are these value conflicts among multiple reasonable values, where no one "best" policy option is rationally identifiable and where expertise cannot yield a reasonable policy proposal all ought to embrace) is that we ought to appeal to fair processes of rational democratic deliberation that include all who might be affected by the choice of some policy. We have spelled out in another essay the norms that ought to govern such democratic deliberative processes to maximize the likelihood of a deliberative process that is fair and reasonable in form and in fact (Fleck, 2006). In the next portion of this essay we describe, in detail, one version of democratic deliberation in connection with the ethics and policy issues related to these stored blood spots. We describe both the process itself and its outcomes.

We have noted already that considerable expert disagreement exists regarding reasonable policies for newborn screening and research uses of these stored blood spots. One thing, however, does recur frequently across the various professional reports and appointed commissions regarding these stored blood spots, namely, that there ought to be a substantive process of public engagement and public deliberation regarding the range of policy options available. What must be emphasized is that such engagement is conceived by its advocates as a sustained active constructive process of deliberation. It is typically contrasted with various types of polling or focus group techniques, all of which are seen as simply aggregating opinions from members of the public, opinions that could be either well-formed or ill-formed, opinions that might have some stability or opinions that were held only for the few seconds for which an answer was required. Those methods of gathering public opinion fail to yield the stable, thoughtful, informed judgments that need to be the foundation for reasonable and fair public policies. They tend to allow majoritarian tyranny to determine outcomes.

The defining feature of democratic deliberation is that it is a socially constructive process. The citizen deliberators first come to understand that they are faced with a social problem requiring a socially agreed upon resolution. They also understand that a significant aspect of the problem is that there are these value conflicts among multiple reasonable values, and that ideally we want a resolution to the social problem that gives due weight to each of these values. We want a resolution that reflects the mutual respect we owe one another as citizens of a common society. Consequently, we will abjure the use of majoritarian power to impose a resolution on a minority group. Instead, we will talk through and think through options with one another in order to construct a policy that represents a reasonable and creative compromise among the values in conflict. These public conversations will be informed by the relevant expertise, but the information provided by assorted experts will not be determinative by itself of the policy option that ought to be chosen. The hope that such conversations might be successful is not utopian because, unlike the situation with the abortion issue, few individuals have rigid long-standing ideological views that constrain the policy options they would be willing to accept.

Rather, with regard to something like the research uses of these stored blood spots, the vast majority of individuals in our society can appreciate the reasonableness of the value conflicts and the need to find policy options and practices that are sufficiently respectful of the full range of relevant values. Further, there is recognition that one or more *public interests* are at stake; and consequently, we need a *policy* response, as opposed to just allowing everyone to choose their own course of action. In the next section of this essay we describe in detail the processes and outcomes of our own efforts at such a deliberative conversation.

IV. Use of Deliberative Juries for Public Engagement

Rationale and Conceptual Model

Public engagement is critical to a more informed approach for the ethical use of biorepositories, places where tissue specimens can be stored for future medical research. Michigan is considering the creation of such a repository for the stored blood spots. It will be referred to as a Biotrust, a name that best expresses public expectations regarding the use of these blood spots. Through public engagement we would hope to address more fairly and thoughtfully what may be intrinsic ethical trade-offs. This could include trade-offs between different kinds of consent processes (for the use of these blood spots), or between policies that maximize protection of donor identity and those that would allow re-identification under some circumstances, or between different kind of processes to keep donors informed of the research undertaken through the proposed Biotrust in Michigan. There may be relevant individual or group vulnerabilities that would remain unrealized without the input of diverse community groups. Moreover, when biobanks are established for public health purposes using public resources, questions are raised regarding what criteria should determine legitimate public goals of research. How the informed public judges such trade-offs, ethically and politically, rightly informs policy guidelines that will protect citizen interests and maintain public trust. Likewise, potential public concerns, public judgment about legitimate goals, and relative consensuses or splits within the public, all these things should also inform policy guidelines that will protect public interests and maintain public trust.

We employed a "deliberative jury" model to engage citizens on a range of issues raised by the Michigan Biotrust Proposal. We modified a deliberative jury model of public engagement that has been pioneered in Great Britain.[13] In order to attain the goals of inclusivity, deliberateness, and reciprocity, the deliberative jury model brings small groups of diverse citizens together in a face-to-face process in which the citizen "jury" is allowed to question "witnesses" (people with special technical expertise or special vested interest in the proposal at hand) and then to discuss policy options and concerns, ultimately articulating both consensuses and ranges of differences. While deliberative juries are not "statistically significant" sample sizes, the qualitative depth of their deliberation can alert policy-makers to a range of issues to guide further public engagement as well as options for policy discussions. Our jury was financially sponsored by IPSSR. It is hoped the jury results will contribute to a wider planned proc-

[13] Graham Smith and Corinne Wales, "Citizens Juries and Deliberative Democracy," *Political Studies* 48:51-65, 2000.

ess of public engagement that includes regional focus groups financially sponsored by the State of Michigan.

Twenty citizens from the greater Lansing area served as jurors in our deliberative process. The jury process was structured as a 3-part process with sessions on 3 consecutive weekends: (1) a half-day educational session (2) a half-day session of short presentations by guest witnesses followed by extensive time for juror questioning and (3) a full day session of jury deliberation on policy recommendations. Jurors were sent briefing materials and a pre-survey before the first session. An audience response system with individual keypads and group tabulating capability was used during deliberations. The virtue of that system is to allow jurors an immediate visual picture of the array of responses to a given question. The system also allows for question addition/modification "on the spot" in response to juror comments or requests. In order to insure juror comfort, their deliberations were not tape-recorded. Rather, a note-taker took detailed notes of the sessions and then provided them to participants.

The jury recruitment aimed to include both citizens associated with organizations that have a special commitment to public health (or specifically to the kinds of health issues addressed by newborn screening and potentially by Biotrust research) and "lay" citizens with no such association. A list of the jurors and the community organizations through which they were recruited is included in the Acknowledgments. Colleagues from Ingham County Health Department and the greater Lansing neighborhood centers provided much-appreciated guidance into our recruiting strategy. Mr. Doak Bloss, an Ingham County Community Health employee with extensive previous experience in community engagement, met with us to consider recruiting strategies.

We specifically aimed to have a higher percentage of racial and ethnic minority participants compared to local demographics, for two reasons. In the first place, literature suggests racial and ethnic minorities may be more suspicious of research with genetic materials than others. In addition, we wanted to minimize the chance that any juror would feel either isolated or pressured to try to "represent" a group. Although we never asked jurors to definitively self-identify race/ethnicity, we estimate that about a quarter of our jury consisted of African-American, Hispanic, or Native-Americans, compared to about 17% in the local community. There was also a South Asian-American among the jurors.

The deliberative jury included physicians, nurses, and several citizens who worked either with disabled or poor clients in health care or advocacy venues. It also included citizens with no special health background. While jurors were recruited through various community organizations, they were instructed that their role was to present their own considered views and questions, not to try to "represent" the organization through which they were recruited.

Guest experts were chosen as jury witnesses to address an array of scientific, ethical, and policy questions regarding the Biotrust. While State of Michigan Department of Community Health officials were not present in person, they provided a detailed slideshow introducing the newborn screening program and the Biotrust proposal for research use of stored bloodspots, and provided written responses to juror questions.[14] Nigel Paneth, M.D., Ph.D., a pediatric epi-

[14] It should be noted that Michigan Department of Community Health officials offered to brief the jury in person, but that scheduling constraints of the moderators' side precluded arranging the educational session at a time those officials could attend. Ms. Janice Bach, Genomics Director, and Ms. Mary Teachout, Genomics Educator, both in the Division of Genomics, Perinatal Health, and Chronic Disease Epidemiology, provided extensive materials and access for questioning by email to the jurors.

demiologist at Michigan State University's College of Human Medicine, addressed the diverse research potential of the stored bloodspots. Mr. Aaron Goldenberg, a Ph.D. candidate in bioethics at Case Western Reserve, who has researched public health biobanking issues and whose dissertation is on the Michigan Biotrust proposal, addressed ethical and policy issues in public health biobanking. One citizen juror, Ms. Rosalyn Beene-Harris, was given dedicated time as a witness as well, given her extensive experience working with the State to improve education on the newborn screening program, especially to minority communities.

Process Challenges

A fundamental challenge was to strike a balance between adequate educational briefing and formats that allowed the jurors' to formulate questions and concerns in their own words. On one hand, it could be hard for jurors to get started without some understanding of the newborn screening program and its clinical/public health purposes, scientific rationale for the Biotrust, and an initial sense of the kinds of concerns generally addressed by research ethics and associated oversight. On the other hand, the process had to be open-ended enough that jurors' could express hopes, concerns, or questions unanticipated by the moderators or the developers of the Biotrust. On one hand, certain procedural questions had to be addressed by the jurists if they were to have practical influence, given that the Biotrust development plan is moving forward (for example, what kind of informed consent process, if any, is ethically warranted for research on stored bloodspots?). On the other hand, the process needed to allow that jurors might see additional policy procedures at stake, or might reject the framework for considering policy options envisioned by the Biotrust developers, or might altogether reject the idea of using the bloodspots for research.

We struggled to strike this balance in several ways. First, the educational materials sent to jurors included both general materials orienting the ethical terrain and more specific materials. The general materials included a description of newborn screening, a description of biobanking, and a description of general axes of ethical concern regarding biobanking—including "individual" issues such as informed consent and privacy, "group" concerns such as potential stigmatization of population sub-groups, and "public" issues such as criteria for publicly valuable research. Some of those materials were borrowed, with generous permission, from colleagues at the Mayo Clinic in Minnesota and the University of British Columbia in Canada (Mayo Clinic, 2007). Those materials were developed for a citizens' engagement process designed to elicit general hopes and concerns of citizens regarding public health biobanking, not to address a more specific policy proposal such as the Biotrust. The more specific materials were a preliminary set of questions geared more explicitly to the Biotrust proposal, probing jurors' preliminary thoughts on some policy choices regarding the use of newborn bloodspots for research, and comparing juror attitudes toward the clinical functioning of the newborn screening program and the proposed research uses of bloodspots. The same questions could then be polled with an audience response system during deliberative sessions, with any difference between pre-deliberative and deliberative polling potentially demonstrating influence of group discussion processes. The full list of questions is included in the Appendices. (Not all questions on the list were addressed in the communal discussions. Of those that were, some generated much more discussion than others.)

We also tried to strike a balance in the actual structure of the group sessions. Even during educational or expert briefings, considerable time (more than half) was devoted to juror

questioning. During the group deliberative session, the jurors sat in a square to address each other, while the "moderators" sat behind that square to flag their presence as resources for conceptual or historical clarification without pretending to be the group "leaders." The principle moderator (Dr. Fleck) facilitated the use of the audience response system.

The challenge of this balance was evident in evaluations. For example, about ½ of respondents were unsure whether the use of the pre-survey had been helpful, while the other ½ found it helpful or very helpful. The difference suggests some may have thought the pre-survey overly directive, while others felt it had helped them connect general ethical hopes and concerns to specific policy choices. At the least, striking the appropriate mean between possible excesses of inadequate briefing and overdirectiveness seems to demand significant time for open-ended questioning and comment, and specifically invited opportunities to raise concerns or hopes not otherwise addressed. It may be impossible to strike a mean that is ideal for each juror. The reasonable goal may be to strive for an adequate balance for the group as a whole.

Another process challenge was time. Despite 16 hours of face-to-face time and additional time demanded to read and prepare materials, many jurors indicated that they felt the issues at stake demanded more time for discussion. Others wished the deliberation session, one long workday, could have been broken up into shorter sessions over more days. (However, increasing the number of days of availability required decreasing the potential juror pool, given schedule conflicts.) In general, the time necessary for structured deliberative processes stands as a challenge to "deliberative democracy." At the same time, the extensive time devoted to this deliberative jury resulted in rich qualitative data that is a special resource for the wider state public engagement process. Hopefully the state can use parameters of discussion from the deliberative jury to structure its necessarily shorter and more streamlined regional focus groups, so that they vicariously benefit from the more extended time-frame of the jury project.

A general challenge of deliberative democracy is to insure that robust citizen deliberation concretely informs actual policy-making processes. The perceived magnitude of that challenge was reflected by significant juror skepticism about the expected influence of their deliberation on policy-making. While the jurors almost universally reported enjoying and learning from the deliberative process, more than half expressed, at best, lack of confidence or, at worst, outright doubt that their deliberations would affect actual policy-formation on the Biotrust. At the same time, the group
expressed general optimism that citizen deliberation offers the potential to guide ethically sensitive policies for research ethics. The jury's good work thus provides an important opportunity to actualize linkages between citizen deliberation and policy formation.

Tenor of the Deliberative Process

The deliberative jurors who participated in this process were paragons of citizenly virtue. They not only read and reflected upon all pre-meeting materials, but they asked for more detail than we had provided in our background overview, particularly about the history of the Biotrust proposal, the current administration of research on stored bloodspots, and planned future administration of the Biotrust proposal. By their accounts in evaluations as well as by our account, they treated each other with great respect, found the expression of views from community members with different backgrounds and interests enlightening, and took directive control of the deliberations. Both because of the bonding that occurred among members and because the jurists found reasons given by citizens with opposing views thought-provoking, the jurists

became vested in insuring their full range of views was reported. When there were either close splits between "majority" and "minority" positions, or when a smaller minority's views on the issue at hand were deeply-felt, the majority as well as the minority expressed concern that the minority view be represented. At several points jurors expressed concern that differences among them would be minimized in an effort to report some vague consensus. At one point a juror threatened to leave the project if assurance to the contrary was not received. (It was.)

Jurors' Deliberative Conclusions

The following reporting of results is in summary form. It reports conclusions that in fact derived from evolving deliberation. During the deliberations, citizen-jurors amended views on relevant axes of discussion and on policy-tradeoffs in response to each other. Thus the summary flags axes or perceived tradeoffs that came to organize deliberation. The summary also flags reports, consensuses, and disagreements at the close of deliberation.

Juror consensuses.

There was strong support (87%) among jurors for the general idea of using bloodspots in medical research. There were other guidelines or issues upon which there was near-universal agreement among the jurors that became increasingly solidified through conversation. These issues centered on the perceived linkage and disjunction between the newborn screening program and the Biotrust, the purposes of the Biotrust, and the administration of the Biotrust.

Jurors felt the clinically-focused newborn screening program and the research goals of the Biotrust were linked by the common blood draw and by an educational process, though consent issues for the two should be treated distinctly. Most felt that current education on newborn screening is inadequate, and that improving education on newborn screening is critical to creating the possibility for consent to Biotrust research, as well as to public trust. Jurors unanimously urged greater education of prospective parents and the public on the newborn screening program as well as on the Biotrust. They advocated educational programs be devised so that prospective parents understand newborn screening before the physically and emotionally stressed time of birth-giving. Several jurors expressed concern that the mandatory nature of newborn screening could result in parents being forced into providing research samples without their awareness.

That fear led jurors to emphasize a distinction between participation in newborn screening and consent for research use of the bloodspots. A significant majority of jurors felt an explicit informed consent process should be required for research use of the bloodspots, though there was disagreement about whether the process should culminate in an opt-in model (specific documented consent required) or an opt-out model (research use acceptable unless donor indicates not acceptable at a specifically flagged point). A minority seemed to feel that explicit opt-out provisions beyond current policy were not necessary, or that research participation might be a civic obligation if the bloodpots were de-identified and the research was oriented to public health.[15] Thus a considerable challenge to current policy was expressed. In accord with the minority rationale, current policy presumes consent unless parents self-initiate objection. The policy presumes parents in general do not mind the use of bloodspots for de-identified research, although newborn screening informational

[15] The size of the minority is hard to quantify given some overlapping questions, but a possible range of 18-40% of participants.

materials in Michigan include a phone number to call if parents do not want samples used in research.

Jurors objected to the fact that when parents who do not want samples used for research call the given phone number, their child's bloodspots will be "destroyed" (according to current informational materials). They felt that parents should have the option to have bloodspots saved for later potential clinical use without having to make them available for research. They also worried that the total destruction of spots for research nonparticipants might serve as a subtly coercive pressure to donate one's bloodspots for research. Here, jurors may have flagged an issue that is unintentionally ambiguous in current policy. Despite the wording on the informational materials, it is not clear whether all the spots are destroyed for parents who self-initiate an "opt-out" from research—since state law requires the saving of a child's bloodspot. Moreover, clarifying an option to save spots for clinical use without participating in research would avoid coercive potential while remaining consistent with the initial reason for providing a phone number to parents discomfited by research participation.

For most jurors on the panel, it mattered greatly *WHAT* research was going to be done with the bloodspots—not only its scientific rigor, but its public health-related goal. Their iconic example of an indefensible pole to be avoided was cosmetic research with little medical benefit that greatly financially benefited a private company. Since jurors understood, or came to understand through the conversation, that new research potentials are continuously evolving and that bloodspots can be used to examine a variety of biomarkers and variables, they struggled with what they perceived as a "chicken-and-egg" question regarding which issue comes first: issues of public purpose or issues of consent for research use of the bloodspots. At several points, they explicitly discussed and changed their minds about in what order to take up the two issues. On one hand, in one juror's words, "I have to know what I am consenting to before I decide whether to consent." On the other hand, others focused on how consent processes could serve as a check on public purposes, given the open-ended research potentials. If consent processes were devised that offered conditional provisos, those jurors might be more comfortable that, as a bloodspot donor, they would have influence on determining public goals of research. A majority of jurors (about 80%) said that parents should have the option of limiting the kinds of research for which their child's bloodspot can be used. The jurors' back-and-forth between issues of public purpose and consent processes is instructive for presentation of related issues in other public forums.

Most jurors felt that the potential for state economic development, especially job creation, is one valid criterion for determining good public purposes of research (assuming the research was otherwise ethically acceptable). However, a significant minority (about 1/3) were uncertain of that. The jurors' discussion of potentially valid and potentially distorting financial incentives progressed from considering differences between potential research institutions (academic, public health, nonprofit, private pharmaceutical) to considering the existence of potential public contributions and potential conflicts of interests within each of those venues. Here again jurors returned to the importance they placed on questions of what actual research would be done.

These axes of concern led to the jurors' unanimous support for a Community Advisory Board [CAB] to oversee research of the Biotrust. They felt there ought to be a Community Advisory Board, in addition to the IRB and a Scientific Advisory Board, to assess proposed research projects from the perspective of community values and reasonable public purposes.

The CAB could also address larger policy issues related to the Biotrust, such as the degree to which private for-profit companies could have access to the bloodspots for research. The Community Advisory Board would be broadly representative of the state in terms of race/ethnicity, economic status, geography, religious orientation, age, professional background, and civil liberty concerns. It could be a single state-wide entity or it could be the head of a network of regional boards. (Perceived tradeoffs between maximizing local voice and creating practicable bureaucratic structures would affect choice of particular structure.)

Areas of Non-consensus

Deliberative jurors disagreed about whether, contra current policy, explicitly documented consent should be required for the newborn screening program. The majority thought "no" but a significant minority (31%) thought "yes." Two consensuses underlay that disagreement: agreement that all babies should be screened for their own benefit, and agreement that current education about screening is inadequate. While both groups favored increased education, the majority feared babies would be lost to screening if documented consent was required. (That fear underlies current policy of mandatory participation and presumed consent.) The minority who preferred documented informed consent hoped it would insure that an education process took place, and expected that appropriately educated parents would consent

Jurors also disagreed about whether an informed consent process for research use of bloodspots should operate on an opt-out or opt-in basis, with considerable support for both models. Underlying discussion of the trade-offs was consensus on the importance of flagging potential research use to parents, differentiating that from the clinical goals of newborn screening, and having some explicit informed consent process for research use.

Jurors were split on whether the bloodspots should be viewed as a public resource, rather than as private property. Two-thirds agreed; one-third did not. Reasons for viewing the bloodspots as a public resource included a wide conception of stakeholders (all citizens of the state, not just individual parents) and realism about social mobility (people move in and out of state). Reasons for declining to view the bloodspots primarily as a public resource included a conception of the bloodspot as private property or a desire for donor voice in use of the bloodspot ("I donated the bloodspot. I should have a say in it").

(c) **Areas of Juror Ambivalence**

Jurors' individually and collectively expressed great ambivalence on several issues related to de-identification. They were uncertain whether they believed bloodspots could really be completely de-identified (given unique genetic information or the potential for identification by combining known variables). Many went "back and forth" about whether they would prefer a system in which bloodspots made available for research are permanently "de-linked" to protect donor privacy (thus not under any circumstances able to be re-linked to the identification of the donor) or one in which information was provided to researchers in a de-linked form, but someone in the health department had a code to re-link if information clinically helpful to the donor was discovered through research. Because questions of de-identification were fraught with ambivalence, deliberative jurors asked detailed questions about how current de-linking processes operate and how they might operate in the future. Briefing materials that address the administrative complexity might be helpful to continued public engagement efforts.

Considerations for Future Uses of the Public Deliberative Jury Model

The proceedings of this deliberative citizens' jury suggest both the value of and challenges for the model as a venue of public engagement. The carefully considered deliberations of

the jurors clearly excavated important axes of concern and clearly articulated tensions and trade-offs to be considered in wider public engagement and policy-making on the Biotrust. While the jury is not a statistically significant sample, it is nonetheless highly significant that the some of its deliberations challenged current policy for bloodspot use (such as presumed consent for research use of the bloodspots). In other cases, deliberation suggests that even where clear majorities support longstanding policies there may be significant discontent among minorities (such as on whether documented informed consent should be required for newborn screening). Beyond any specific topical findings, the jurors' expressed appreciation for alternate views of diverse participants in a face-to-face format suggest that the public jury model can play a helpful role articulating the good-faith reasoning behind different policy options.

Our process also suggests challenges—not insurmountable—to the use of public deliberative juries. Inclusive jury recruitment inevitably poses challenges. While we made great efforts to insure adequate inclusion of minorities and "lay" people without any obvious vested interest, jurors expressed regret that we did not have more representation from the business community. (One juror was recruited through a business and economic development organization).

Construction of the guest witness panel also poses challenges. While we used the language of "guest expert" to denote those with special knowledge whom jurors' were able to question, some believed in retrospect the language of "guest witness" was preferable. The implication of the jury process is that citizens themselves are the "experts" on primary value judgments at stake in policy questions.

Since that is the case, some would argue that it would be preferable to take into consideration juror input on what kinds of guest experts/ witnesses might be important, as well as the moderators' choices of guests. However, that would extend the time necessary for the process, as jurors could not be expected to form ideas about desired guests until after some initial educational briefing. Whether community organizations could play an intermediate role in suggesting important experts/ witnesses may be deserving of exploration.

Time is a challenge in many ways. Just given the need for availability throughout the process (or on a certain day, for the guest experts/ witnesses), schedule constraints may limit the potential pool of jurors or guests. Moreover, the perception of luxurious time in the jury process may be a dangerous illusion. While the much greater time, compared to traditional focus groups, allows for greater levels of complexity to be addressed, those levels of complexity demand greater time to address. Thus it may be challenging to direct deliberation to definite conclusions or clear articulations of ranges of views, though the articulation of levels of complexity is itself a great accomplishment. One possible modification for future juries might be to have an explicit process to ask what life experiences brought people to the table, rather than simply allowing that to come out sporadically in conversation. That might efficiently catalyze discussion of connections between viewpoint and view that the jurors seemed to value greatly.

A final challenge is to insure that the effort-filled and careful deliberations of citizens in a jury process receive due attention from policy-makers. We hope the State will tap the deliberative jury's expressed concerns and spectrums of views as it designs its process of regional focus groups on the Biotrust. We also hope that legislators and other policy-makers will consider the implications of the jury's deliberations for policy-formation.

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MICHIGAN STATE UNIVERSITY

Institute for Public Policy and Social Research

College of Social Science
Michigan State University
321 Berkey Hall
East Lansing, MI 48824-1111
Phone:517-355-6672 Fax:517-432-1544
Web: www.ippsr.msu.edu
Email: ippsr-action@ssc.msu.edu