This article was downloaded by: [Dierickx, Kris]

On: 14 September 2010

Access details: Access Details: [subscription number 926899351]

Publisher Routledge

Informa Ltd Registered in England and Wales Registered Number: 1072954 Registered office: Mortimer House, 37-41 Mortimer Street, London W1T 3JH, UK

New Genetics and Society

Publication details, including instructions for authors and subscription information:

<http://www.informaworld.com/smpp/title~content=t713439262>

The use of stored tissue samples from minors for genetic research: interviews with professionals

Kristien Hens^a; Kris Dierickx^a

^a Centre for Biomedical Ethics and Law, Katholieke Universiteit Leuven, Leuven, Belgium

Online publication date: 14 September 2010

To cite this Article Hens, Kristien and Dierickx, Kris(2010) 'The use of stored tissue samples from minors for genetic research: interviews with professionals', New Genetics and Society, 29: 3, 329 — 342

To link to this Article: DOI: 10.1080/14636778.2010.507488

URL: <http://dx.doi.org/10.1080/14636778.2010.507488>

PLEASE SCROLL DOWN FOR ARTICLE

Full terms and conditions of use: <http://www.informaworld.com/terms-and-conditions-of-access.pdf>

This article may be used for research, teaching and private study purposes. Any substantial or systematic reproduction, re-distribution, re-selling, loan or sub-licensing, systematic supply or distribution in any form to anyone is expressly forbidden.

The publisher does not give any warranty express or implied or make any representation that the contents will be complete or accurate or up to date. The accuracy of any instructions, formulae and drug doses should be independently verified with primary sources. The publisher shall not be liable for any loss, actions, claims, proceedings, demand or costs or damages whatsoever or howsoever caused arising directly or indirectly in connection with or arising out of the use of this material.

The use of stored tissue samples from minors for genetic research: interviews with professionals

Kristien Hens* and Kris Dierickx

Centre for Biomedical Ethics and Law, Katholieke Universiteit Leuven, Kapucijnenvoer 35/3 Box 7001, 3000 Leuven, Belgium

Stored tissue sample collections from minors are useful for genetic research. The ethics of the use of stored tissue samples for genetic research continues to be a much debated topic of discussion. The use of pediatric samples poses specific ethical questions. Although a substantial corpus of empirical literature exists on the use of samples from adults, not much is known about the opinion of professionals on the use of samples from children. We conducted ten interviews with professionals in the field. Their primary concerns were the fact that consent procedures should not be too bureaucratic, the need for proper research design and privacy protections, the fact that research should not burden children and guidance from ethics committees.

Keywords: bio banks; minors; interviews with professionals; ethics

Introduction

Stored tissue sample collections from minors are useful for genetic research. For example, longitudinal studies such as ALSPAC (Pembrey 2004), which follow children from pregnancy onwards, can shed important light on the relationship between genes and environment, and help to study diseases such as asthma, childhood obesity and allergies. Pediatric tumor banks are crucial in understanding childhood cancer. Other types of collections can take the form of disease-specific collections (Qualman *et al.* 2004). Also, samples that were collected in the context of diagnosis could be useful for genetic epidemiology research. Such collections exist in centers for medical genetics throughout the world. Moreover, blood spot cards, containing blood that was gathered from newborns to screen for certain metabolic diseases, could also be used for this purpose (Klotz *et al.* 2006).

The ethics of the use of stored tissue samples for genetic research continues to be a much debated topic of discussion. A substantial corpus of both theoretical as well as empirical studies exists on the topics of consent, privacy, ownership, commercialization and the return of incidental research findings. The vast majority of these documents, however, deal with tissue samples from competent adults. Meanwhile,

*Corresponding author. Email: kristien.hens@med.kuleuven.be

the use of stored tissue samples from minors poses specific ethical problems that are different from those posed by adults. Two recently published reviews of the empirical and theoretical literature and of existing guidelines have identified these differences (Hens *et al.* 2009a, 2009b). Specific ethical questions raised are for example, whether parents can consent for their children, the scope of the parental consent (generic/broad), the right to assent for the child, the requirement that research should not pose more than minimal risk to the child, the question whether results should be returned to the parents or to the children. With regard to consent, different issues arise. What is the scope of parental consent? Are parents allowed to give consent to the indefinite storage of stored tissue samples from their children for genetic research or should there be a time limit? With regard to the content of the consent, are parents allowed to give broad consent for any future genetic research, or only for specific research (Hens *et al.* 2009b)? Should children be allowed to assent to research and should they be recontacted for consent when they become legally competent? It is universally acknowledged that asking young adults to recontact is good practice and that they should be allowed to withdraw their consent. It is unclear, however, to what extent researchers should try to contact them or what the specific modes of withdrawal should be (Helgesson 2005, Burke and Diekema 2006). A recent article by Gurwitz *et al.* published as a policy forum in *Science* stresses the need for extra guidelines for pediatric biobanks and longitudinal studies involving children (Gurwitz *et al.* 2009). As children, unlike adults, cannot consent to storage and use of DNA themselves, restrictions should be put in place. Longitudinal studies should either invest in in-house processing facilities or await the consent of participants once they are adults. The authors refer to possible risks associated with stored DNA, risks which are primarily linked to discrimination by insurers and employers. Since DNA is stable throughout the lifetime of a human being, they argue that children are in need of extra protection and that parents should not be allowed to consent to all types of research using their children's DNA. Indeed, the most frequently discussed issue with regard to biobanks and risks is privacy and confidentiality related. As research collections for biological samples contain genetic information, this is potentially interesting for third parties such as employers and insurers. Genetic information is thought to contain a "future diary" of the individual (Annas 1993). This is enhanced by the fact that such information may be stored for longer periods of time. If such third parties gain access to this information, research participants, who altruistically donate their samples to research, may be confronted by the adverse effects of their participation later in life. With children this is further complicated by the fact that they themselves have not chosen to incur this risk. Other risks that are quoted in literature are the potential physical and emotional burdens that the gathering of biological samples and subsequent medical examination can cause children (Goodenough *et al.* 2003) or the fact that children's opinions may change or differ from their parents' and their viewpoint may be disregarded (Williamson *et al.* 2004).

Despite the continuing debate, only limited empirical work has been published regarding the opinions of clinicians and researchers working with stored tissue samples from minors. Solely one paper directly addressing some of these issues was found; it reported findings of a survey that investigated the views of healthcare professionals working in childhood cancer about seeking consent for tissue banking from potential donors (Jackson *et al.* 2009). We also found a study reporting on eight interviews with pharmacogeneticists on the implications of pharmacogenomic research with children (Avard *et al.* 2009), and one study querying the opinions of researchers in Spain and the USA on the issue of stored tissue samples in general (Ruiz-Canela *et al.* 2009). Ries *et al.* conducted six semi-structured telephone interviews with investigators of six birth cohort studies (Ries *et al.* 2010). They examined how these studies handled ethical, legal and social issues with regard to recruitment, parental consent and assent/consent from the child, withdrawal, confidentiality and sample/data protection, handling sensitive information and disclosure of results (Ries *et al.* 2010a).

It is still largely unknown what professionals consider ethically sound and realistic guidelines. Therefore, we considered it a necessity to interview such professionals in order to find out their thoughts and opinions regarding the main ethical issues involved in the storage and use of tissue samples from minors. Herein, this article presents the principal findings of these interviews. We shall use the term “minor” and “child” in the rest of this paper, denoting persons unable to fully consent to research because of their age.

Methodology

As not much is known about the opinions of professionals on genetic research on tissue samples from children, we chose to perform in-depth interviews with professionals involved in such research. The qualitative method of analysis allowed us to detect themes of importance. This is a preliminary study of the issues at stake, and we think our findings could serve as a basis for further quantitative research in the form of surveys. Interviewees were selected through the snowballing technique, which resulted in 10 interviews. These interviews took place during the period of January 2009 to August 2009. The interviews were conducted with healthcare professionals or researchers from Belgium (three persons), the UK (six persons) and from Saudi-Arabia (one person). Six interviews were conducted face to face, while for practical reasons, the four remaining interviews were accomplished by telephone. The interviewees' profiles and main area of specialty varied (Table 1) but they are/were all involved in the procurement and/or management of tissues for research or in the research itself.

Interviewees were initially contacted via email and face-to-face or telephone meetings were then arranged. Each interview took between 40 to 90 minutes. The language of the interviews was English or Dutch.

We developed an interview guide based on a literature review (Hens *et al.* 2009b). The interview guide contained the themes consent, assent, usefulness of

Table 1. Information about the interviewees.

		#
Profession of interviewees	Clinical geneticist	4
	Gynecologist	1
	Epidemiologist	2
	Oncologist	1
	Research team leader	1
	Internist	1
Gender of interviewees	Female	4
	Male	6
Location of interviewees	Belgium	3
	United Kingdom	6
	Saudi Arabia	1
Type of collection	Diagnostics collection in a center for medical genetics	4
	Longitudinal study	3
	Tumor bank	1
	Research-only collection	2
Type of interview	Live	6
	Telephone	4

research, genetic information, return of result and minimal risk. Our approach was a loosely structured interview rather than a structured one as we wanted to elaborate interesting remarks and findings during the interview. Kristien Hens (KH) was the interviewer in all 10 cases; Kris Dierickx (KD) was assistant interviewer at the first interview.

Audiotapes of the sessions were transcribed but not corrected for grammar, in order to capture the oral nature of the discussion. We used NVivo 8 to code the transcripts (Bazeley 2008). First, the texts were coded according to the 10 cases, reflecting the 10 interviews, allowing us to check distribution of certain themes. Then we did a detailed topical coding. We subsequently combined the codes into broader thematic categories. As a last pass, we did a fresh, analytic coding to grasp common themes across the different topics. We selected quotes that were especially illustrative of these themes and subthemes. We have specified the profession of the interviewee and the type of interview with each quote. These common themes and the associated extracts are reported in the “results” section. An overview of the themes is given in Table 2. Given the limited number of interviewees and to protect their anonymity, we shall use the pronoun “she” when referring to the interviewee.

Results

Bureaucracy, good governance and ethics committees

The word *bureaucracy* or a derivative was mentioned 11 times by seven different sources. Official requirements regarding consent and assent for stored tissue

Table 2. Overview of themes and findings.

Themes	Findings
Bureaucracy	Long written consent forms for children and their parents is often considered unnecessary burdensome for participants.
Governance	
Ethics committees	Need for proper privacy and data protection. Ethics committees should review whether reconsent is needed for when research protocols change. Ethics committees should review burden of research for children.
Process of decision-making	People are willing to contribute and consent to research on stored tissue samples from their children. Good communication between researcher, parents and children is important. Children should be given information at their level. Whether a child should be asked to assent should be decided on a case-by-case basis rather than on a fixed age threshold.
Burden/benefit	If possible, samples and information should be reused for research purposes to decrease burden for children. If possible, anesthetic cream should be used for venepunctures. Research on children should be for pediatric conditions or for conditions that develop during childhood.
Genetic information	Genetic information considered to be similar to other information. The difference lies in the familial nature of the information.

samples and minors were often seen as bureaucratic. Interviewees from the UK and one from Belgium stated that they thought requirements of having children sign consent forms were superfluous. As a seven-year-old, or even an older child, is never asked to sign forms in everyday life, these interviewees considered asking children to do so for research was useless. They also complained of the long consent forms the ethics committee asked them to use. Interviewees stated that the longer the information sheet and the more boxes parents and children had to tick, the less confidence participants had in the research and the less likely they were to consent. Written consent was seen by these professionals as a legal requirement to avoid prosecution, rather than as added value to the researcher–participant relationship. The UK interviewees referred to the stringent requirements they had to fulfill after the Alder Hey scandal, which involved the use of post-mortem tissue samples without consent. They thought that this was not applicable to tissue from children that were still alive, as people were overall willing to contribute to research. Researchers did think that good governance of samples with adequate data protection and scientifically sound research design was essential. If these requirements were fulfilled, they thought research was ethical, and not too many restrictions should be put in place. This is well illustrated by this quote from a professional involved in a cohort study with children:

I think transparency is the key, and having good systems, if people do retract their consent, which people do, we are able to destroy samples, giving people assurance

that we are treating it well and, ehm, being very robust about anonymization of samples. We had consent about the transfer within between countries, there may be concern about that, I think the key for us is we have a wonderful resource in terms of performing future ehm health policies, and that is what people have signed up for, and we should not over bureaucratize it, I think. (Epidemiologist, telephone interview)

This person clearly summarizes what was also quoted by others as good research with good governance. Projects should be transparent, people should be able to withdraw and samples should not be identifiable to the researchers working with them. Note the use of the word “anonymization” here, which was used by some interviewees, but upon further questioning this in all cases meant “coded.” Although ethics committees were sometimes seen as being too stringent and requiring too long and bureaucratic consent, interviewees also saw a great advantage in having research ethics committees review protocols. Such committees could help decide about the acceptable level of involvement that researchers could ask from children. They could also help in deciding whether reconsent for certain research proposals was necessary. Indeed, some of our interviewees thought that parents could be asked to give broad consent for research on stored tissue samples from their children. However, after further elaborating this point, all of them agreed that broad consent did not necessarily mean “any and every” research for them. For example, they all thought genome-wide association studies might be too far from the original research protocol: in such cases they would seek advice from an ethics committee to check whether reconsent was necessary. Two interviewees also used the term “fishing expedition,” a colloquial expression to denote an attempt to extract more, or other – in this case genetic – information than was specified in the research protocols. Hence, although they supported a more general consent for research, they suggested that a research protocol should specify the type of information that is sought.

Process of decision-making

Overall, interviewees stated that people were happy to contribute and to give consent for research on stored tissue samples of children, especially when these children had a medical condition themselves. Also interviewees thought that many parents were willing to enroll during pregnancy for population studies, because they saw the spread of certain childhood diseases in their environment, and participating made them feel part of something important. They thought it would be unacceptable to not use tissue samples from children for research and were convinced parents would not be pleased if existing information and samples could not be used to the benefit of medical science.

They did not think, however, that this positive attitude towards scientific research among parents should be taken for granted, as parents and children also have the right to proper information. Therefore, they thought good communication and a good relation between researchers and families was indispensable. When research

samples are gathered in a diagnostics circuit, this is obvious. However, also the interviewees associated with research-only projects stated the need for proper communication, for time spent with parents and children to explain the procedures. They also thought that parents were the primary decision-makers when the child was small, but that it was important that children are given explanations on their level and would be gradually more involved in the decision making:

Even with the little babies, before you give them an injection the majority would still say it is going to be a sharp scratch. The baby has no clue what you are saying, but you still tell them what you are doing. And it just gets more detailed what you are doing when they get older. So the guidelines say at seven you should start giving information, you are doing that from eight weeks old, you are just doing that in a completely different way. (Team leader, oral interview)

The above quote shows the difficulty of setting a definite age threshold from which point onwards a child can understand information or should be given the opportunity to consent. In this context, interviewees quoted the fact that some children have mental retardation due to genetic illnesses, and that some children are more knowledgeable than others at a given age. Hence, they thought the amount of information given to children and whether or not these children could be asked to assent should be evaluated on a case-by-case basis. Related to this, the fact whether small children should be allowed to dissent to certain procedures was discussed with some professionals. Many interviewees claimed that they would not force a screaming child to undergo a venepuncture, and would hence give priority to the dissent of the child over their own research goals. However, one interviewee stated that she thought parents should be the ones to decide whether to continue the procedure or not, but that she thought there was also a difference between healthy and sick children:

... and you had a four-year-old with cancer and a healthy four-year-old, and they would both say no, I don't want the needle. I think I would accept the view of the four-year-old with cancer because she or he would have had hundreds of needles before and if she is saying no I don't want it, she knows what it is about. A normal four-year-old is saying no, they would just say no ... So I think they just have a different experience and therefore should be dealt with differently. I am not saying you should just pin every four-year-old down ... (Team leader, oral interview)

This interviewee links the ability to dissent to the experiences children have with the procedures. In this respect, children with a medical condition such as cancer, who had to undergo the procedure frequently would be more able to make a judgment about whether they wanted to participate or not. But healthy children could be stimulated to participate if their parents agree to it, as they might say no without actually knowing what they say no to. In this respect, she considers the procedure (venepuncture) as relatively harmless: participating in research involving such

procedure may be considered as part of normal activities children just have to do, despite their wishes.

Burden and benefit

None of our interviewees stated that research on stored tissue samples from minors would not be useful. On the contrary, two of them stated explicitly that they thought too many restrictions would be unethical, as children would be disadvantaged.

In several guidelines on stored tissue samples and biobanks, the section on minors states that research on children should be for children (Hens *et al.* 2009a). We asked some interviewees whether they thought this restriction made sense. They agreed, although they thought this should be interpreted rather broadly. For example, some diseases may be not typically “child specific” but may develop during childhood. Other diseases may also occur in adulthood, but the adult variant may not be entirely analogous with the childhood one. One interviewee stated that in her line of research, it is easier to work with tissue from children than from adults, because the genetic condition she studied caused severe mental retardation. It is easier to get parental consent for research when the child has not yet reached the age of majority than it is to get consent for an incompetent adult.

Although interviewees thought that there was not much risk associated with genetic research on stored tissue samples from minors, they agreed that children should not be burdened with research. On the one hand, the use of anesthetic cream was mentioned nine different times by four different interviewees. They thought the use of this cream, possibly in combination with a video and proper professional assistance was enough to satisfy the non-burden requirement, as is illustrated by the following quote:

I suppose it is not actively beneficial, but I think the risk is pretty minimal. And I think taking blood from a child who is not struggling hard is also minimal, I do not see that as a major problem. Particularly at least now, in the past, before anesthetic creams are used, there were some children that really became quite distressed, but that shouldn't be a problem now. (Clinical geneticist, oral interview)

On the other hand, the reuse of information and samples gathered in a diagnostics context was seen as good practice. One interviewee mentioned the risk of thrombosis through venepuncture, and thought that, although this risk is low, drawing of blood should be restricted to the minimum and samples should be reused as much as possible. Along the same lines, cooperation with schools and the possibility to collect medical data through the healthcare system was applauded as a good way of reducing the burden on children. The following quote states clearly that researchers have an ethical responsibility towards research participants to be as least intrusive as possible:

Researchers duplicate and we are collecting, we do have to collect additional information sometimes, but we have got clinicians doing all these measures and

assessments on one side, and in parallel we have got researchers doing the same thing I think there is a responsibility vis-à-vis the parents to say we'll be slick, we won't disturb you where possible. (Epidemiologist, phone interview)

This interviewee thought that redoing measurements and collecting duplicate information because of stringent consent requirements was unethical. The need for informed consent to reuse data and samples from a diagnostics collection is sometimes hard to obtain. It may be easier to gather again samples and data and consent from research participants. However, this causes extra burden as procedures would have to be done twice. Especially when children are concerned, the issue of burden becomes important and the need to reduce this burden may be more important than the need to gain consent for each new use of samples and information.

Specialty of genetic information

Most of our interviewees did not consider genetic research as different from other medical research, provided that proper data protection measures are put in place. Some referred to the difference between the European and the US context, and the fact that many assumptions are based on the latter. They thought that in the context of a public healthcare system, where health insurance is strictly regulated, the risk of participating in genetic research is minimal. As such, they did not see a big difference between the risks for children as for adults. They also thought that their research participants did not consider genetic research as particularly different or potentially risky, as is illustrated by the following quote:

People are not so concerned about the genetics. Especially not in Belgium, in a safe situation. It is illegal to abuse such information, and as a doctor you are not obliged to give that information. The genetic aspect is not an issue for patients. (Internist, oral interview)

This interviewee thought that in Belgium, as it has an extensive public health system and legal restrictions on the use of genetic information, people were not concerned about privacy issues related to this type of information. Hence, her viewpoint on the risks of this information is related to potential misuses of this information through "malevolent" third parties such as insurers and employers.

Three interviewees mentioned that familial nature of genetic information sets it apart from other information. Two of them considered this to be positive: genetic family trees can explain and provide a reason for certain conditions, and hence provide a way to handle these conditions. One interviewee saw a potential danger in genetic information that can be acquired from longitudinal studies:

Take the example of Huntington, if you do a study where you are looking in a population for the Huntington's gene, and you find a child of 11 or 12 or so with the gene, which we do not have at the moment, and if you are looking in the parents and you find it in the parents and they've got a young child, even when the child is not in the study, ehm, what are the implications for the children about that, knowing about the Huntington condition where there is no remedy . . . (Gynecologist, oral interview)

On the one hand, this interviewee considers the fact that people do not understand genetic information, and might be influenced by the media to consider it as potentially hazardous. On the other hand, the fact that through genetic research on biological samples researchers could detect certain genes leading to untreatable diseases poses additional difficulties. The question whether and how to return such incidental findings requires much reflection beforehand.

Discussion

We interviewed 10 professionals working in the field of stored tissue samples on the topic of research on pediatric biological samples. We found that researchers were concerned about the bureaucratic restrictions of consent procedures on research. But we also found the need for guidance from ethics committees with regard to the scope and content of consent. Interviewees thought that children should not be burdened with research and should be told, in their own language, about the research. There was some disagreement on the issue of dissent of small children. Interviewees did not think genetic research was too different from other research; the difference was situated in the familial nature of genetic information.

Our interviewees thought that genetic research on stored tissue samples was useful and should not be complicated by bureaucratic consent procedures. Especially the fact that young children should also sign consent forms was often seen as a useless requirement, with no relation to the actual reality of children's lives. The interviewees thought the researcher, in conjunction with the parents, would be in the right position to assess what and when to tell a child about research. As some of our interviewees have also argued, long information sheets would frighten participants. Proper communication and building a relationship of trust with both parents and children was seen as essential. This is consistent with two other studies in the field. Avard *et al.* did eight interviews with professionals in the field of pharmacogenomics, where the researchers highlighted the need to communicate with children in a clear and concise manner, and to appropriately involve children (Avard *et al.* 2009). A survey by Jackson *et al.* among health professionals in childhood cancer also stresses the need for conversations with families, next to the written information (Jackson *et al.* 2009).

Although our interviewees denounced complex formal rules on children's assent and consent, they also thought that research should be ethically sound. For many of our interviewees, this meant that research protocols were reviewed by ethics committees and that research was transparent. However, there is a limit to what transparency can accomplish. As Onora O'Neill has argued, transparency and a culture of accountability is thought to replace the need for (naïve) trust in biomedical research (O'Neill 2002). But it is now also acknowledged that transparency cannot completely replace trust. De Vries has made a similar remark: there is more to trust than simple transparency. Disclosure can alert to potential problems but does not fix them as such (De Vries and Elliott 2006). Our interviewees

seem to implicitly acknowledge that: a good policy that protects the confidentiality of the genetic information in samples should be laid down in research protocols and implemented before any sample gathering takes place. And participants or their parents should be properly informed about these measures.

One of the difficulties of formal consent and assent requirements when children and youngsters are concerned is the near impossibility of setting a fixed age threshold. The study of Avard *et al.* suggested that those minors aged 12 and older should be consulted about their willingness to participate in research (Avard *et al.* 2009). Also in the survey by Jackson *et al.* the age of 12 was the mean age quoted for consent, although some respondents in that study gave an age as low as four. Our interviewees acknowledged that besides age, there are other factors to be considered: for example, a child with a chronic illness may be more knowledgeable about medical procedures. Related to this topic is the question if and under which circumstances the dissent of a child should be taken into account. The Convention on the Rights of the Child asserts that children have a right to say what they think should happen when adults make decisions that affect them (Article 12 of the United Nations Convention on the Rights of the Child 1998). Does this mean that researchers would have to respect the wishes of a four-year-old regarding the taking of blood for research purposes, regardless of the wishes of his or her parents? Can such a child be forced to donate blood for research? We think that anyone involved in pediatric research should be trained to assess these situations. Ethics committees should also double-check whether this issue is sufficiently thought through when they review research protocol.

In a survey study by Ruiz-Canella *et al.* among geneticists in Spain and the US about the use of stored tissue samples for research (Ruiz-Canella *et al.* 2009), and in the survey study by Jackson *et al.* it is shown that many researchers would prefer broad consent for future research. This is also the preference that some of our interviewees expressed. However, when we investigated what they understood by “broad consent,” and whether this would include genome-wide association studies, we found that for them, broad consent was not analogous to “any possible future research.” Broad consent was seen as a convenience to allow research to proceed without too many impediments. However, interviewees still thought that for research that would cross certain milestones, such as a study that would require a full genome sequence, or research protocols that would be completely different from the original context, ethics committees should review the consent and decide whether recontacting the donors was necessary. We think that it would be extremely useful in the debate about specific versus broad context to investigate further the content of these concepts, as “broad” might mean something different to both parties. The question about the desirability of broad consent is complicated by the tension between the traditional right to personal autonomy of research participants and the need for solidarity (Knoppers and Chadwick 2005). Some authors have argued that, as research on stored tissue samples does not require much effort from the participants and carries potentially huge benefits for

future generations, the duty to participate in such research is higher than the need to emphasize personal autonomy (Chadwick and Berg 2001). In this context, it is acceptable to ask for broad consent. However, children do not easily fit into the solidarity paradigm (Harris 2005). In some contexts, they are exempt from certain duties that adults have, such as paying taxes and being employed, although in other contexts, civilized behavior is expected from them. Although we agree that to ask broad consent from adults may be a good balance between the right to personal autonomy and the duty to promote solidarity in medical research, we also think that for children this may be different, as the decision to give broad consent for research is made by the parents and for small children their wishes are largely unknown. We acknowledge that asking parents or older children to re-consent to any change in the research protocols for research on their samples may be burdensome as well. A compromise would be to allow parents and older children to re-consent or assent to major changes in types of research carried out on their samples and to ask for final broad consent when the young adult reaches the age of competence.

Our interviewees thought the risks associated with research on stored tissue samples from children were fairly minimal. They first and foremost referred to physical discomfort that children might feel and thought this could be minimized by using properly trained personnel and anesthetic cream. This is also found in the study by Avard *et al.* where researchers mentioned buccal swaps, spitting and the reuse of blood taken in a diagnostics context. With regard to the genetic nature of the information this was seen as similar to other medical information in most aspects. The interviewees thought the familial nature of the information was the most important aspect that sets aside genetic information. This has repercussions not so much for the research per se but for the discussion about whether and how individual genetic results should be communicated to the participants, and whether family members also have the right to this information. Interviewees did not spontaneously mention the use of genetic information by third parties as problematic, an issue that is discussed in great depth in the literature. This is analogous with the findings by Avard *et al.* who found that some researchers thought that genetic privacy concerns about third party access should not hold back research (Avard *et al.* 2009).

Our study has shown that the issues related to the use of pediatric tissue for genetic research described in theoretical literature do not always correspond to the opinions and experiences of professionals in the field. Specifically, formal and elaborate consent and assent requirements are sometimes considered superfluous and even an impediment to a proper relationship between researchers and participants. They put much emphasis on good individual relations between researchers, parents and their children, which is more important than fixed guidelines on the age of assent or the need to re-consent for certain changes in research protocols. They thought ethics committees could fulfill an important task in aiding researchers in developing this relationship and making decisions about desirability and burden of research.

Many also thought the risks of genetic privacy breaches and third party access to genetic information were exaggerated and not applicable to countries with a public health system. Considerations of this kind should in any case not prevent research on children's tissue from proceeding. They did acknowledge that measures to protect confidentiality should be put in place and were part of proper research design, and that research should not unnecessarily burden children.

We acknowledge that our study has several limitations. First, we used the snowballing technique for selecting participants. This is not a systematic way of selecting interviewees, and we have probably missed out interesting interviewees. The geographical and cultural diversity of our sample was limited (three countries), which would explain the conformity of answers to certain questions, especially those about third party access and risk. We also used both telephone with live interviews, two different types that do not allow for one-to-one comparison. However, we believed that our data were interesting enough to allow for further analysis and reporting, especially because this is an area largely unexplored, and because the opinions of the professionals in our samples did not necessarily agree with the issues that are considered important in the theoretical literature on this topic. Further research could include in-depth interviews in countries with a less elaborate public health system. Also, a quantitative survey of professionals on the themes discovered in this study could assess the prevalence of similar opinions in a larger dataset.

Acknowledgements

This work was supported by FWO Flanders, project number G029107. We would like to thank our interviewees for their time and insights. We would also like to thank Heidi Howard and the two anonymous reviewers for their valuable comments.

References

- Annas, G.J., 1993. Privacy rules for DNA databanks: protecting coded "future diaries." *Journal of the American Medical Association*, 270 (19), 2346–2350.
- Avard, D., *et al.*, 2009. Researchers' perceptions of the ethical implications of pharmacogenomics research with children. *Public Health Genomics*, 12, 191–201.
- Bazeley, P., 2008. *Qualitative data analysis with NVivo*. London: Sage.
- Burke, W. and Diekema, D.S., 2006. Ethical issues arising from the participation of children in genetic research. *Journal of Pediatrics*, 149 (Suppl. 1), S34–S38.
- Chadwick, R. and Berg, K., 2001. Solidarity and equity: new ethical frameworks for genetic databases. *Nature Reviews Genetics*, 2 (4), 318–321.
- De Vries, R. and Elliott, C., 2006. Why disclosure? *Journal of General Internal Medicine*, 21 (9), 1003–1004.
- Goodenough, T., *et al.*, 2003. "What did you think about that?" Researching children's perceptions of participation in a longitudinal genetic epidemiological study. *Children & Society*, 17, 113–125.
- Gurwitz, D., *et al.*, 2009. Research ethics: children and population biobanks. *Science*, 325 (5942), 818–819.
- Harris, J., 2005. Scientific research is a moral duty. *Journal of Medical Ethics*, 31 (4), 242–248.

- Helgesson, G., 2005. Children, longitudinal studies, and informed consent. *Medicine, Health Care and Philosophy*, 8 (3), 307–313.
- Hens, K., *et al.*, 2009a. Biological sample collections from minors for genetic research: a systematic review of guidelines and position papers. *European Journal of Human Genetics*, 17, 979–990.
- Hens, K., *et al.*, 2009b. Genetic research on stored tissue samples from minors: a systematic review of the ethical literature. *American Journal of Medical Genetics A*, 149A (10), 2346–2358.
- Jackson, C., *et al.*, 2009. Seeking consent to tissue banking: a survey of health professionals in childhood cancer. *European Journal of Cancer Care (Engl)*, 18 (4), 391–400.
- Klotz, J., *et al.*, 2006. Population-based retrieval of newborn dried blood spots for researching paediatric cancer susceptibility genes. *Pediatric and Perinatal Epidemiology*, 20 (5), 449–452.
- Knoppers, B.M. and Chadwick, R., 2005. Human genetic research: emerging trends in ethics. *Nature Reviews Genetics*, 6 (1), 75–79.
- O'Neill, O., 2002. *Autonomy and trust in bioethics*. Cambridge: Cambridge University Press.
- Pembrey, M., 2004. The Avon Longitudinal Study of Parents and Children (ALSPAC): a resource for genetic epidemiology. *European Journal of Endocrinology*, 151 (Suppl. 3), 125–129.
- Qualman, S.J., *et al.*, 2004. Establishing a tumour bank: banking, informatics and ethics. *British Journal of Cancer*, 90 (6), 1115–1119.
- Ries, N.M., Legrandeur, J., and Caulfield, T., 2010. Handling ethical, legal and social issues in birth cohort studies involving genetic research: responses from studies in six countries. *BMC Medical Ethics*, 11 (1), 4.
- Ruiz-Canela, M., Valle-Mansilla, J.I., and Sulmasy, D.P., 2009. Researchers' preferences and attitudes on ethical aspects of genomics research: a comparative study between the USA and Spain. *Journal of Medical Ethics*, 35 (4), 251–257.
- Williamson, E., *et al.*, 2004. Children's participation in genetic epidemiology. In: R. Tutton and O. Corrigan, eds. *Genetic databases: socio-ethical issues in the collection and use of DNA*. London and New York: Routledge, 139–160.