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Childhood genetic testing for familial cancer: should adoption make a difference?

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Abstract

Professional guidelines and practice in clinical genetics generally counsel against predictive genetic testing in childhood. A genetic test should not be performed in a child who is too young to choose it for himself unless that test is diagnostic, will lead to an intervention to prevent illness, or enable screening. It is therefore generally considered unacceptable to test young children for adult-onset cancer syndromes. However, these guidelines are challenged when clinical genetics services receive requests from adoption agencies or pre-adoptive parents for predictive genetic tests in children being placed for adoption. Testing will foreclose a pre-adoptive child's future autonomous right to choose, yet those commissioning these tests argue that adoption should form a special case. In this paper, we argue that predictive genetic testing as part of a pre-adoptive 'work-up' should be discouraged when the same test would not generally be carried out in a child who is not being adopted. We present an argument based on a principle of consistency and question those claims that privilege the adoptive process, whilst acknowledging the array of uncertainties faced by pre-adoptive parents. We suggest that if pre-adoptive testing is considered, this should only take place after prospective adoptive parents have had the opportunity to meet the clinical genetics team and fully understand the implications of the testing process.

Keywords

Adoption, Autonomy, Best interests, Cancer, Children, Genetic testing, Parent–child relations

Abbreviations

BAAF: British Association for Adoption and Fostering
BRCA1: Breast cancer 1 gene
FAP: Familial adenomatous polyposis

Introduction

Genetic testing in children frequently provides dilemmas for medical care providers, despite the existence of professional guidelines [1]. An additional issue has recently emerged—the testing of pre-adoptive children as part of the adoption process. In assessing children prior to adoption, part of the standard work-up is to establish family history, insofar as this is possible. Such a work-up will include a consideration of the child's risk of inherited conditions. Anecdotal evidence suggests that

there are increasing cases where a genetics department is being requested by either the adoption team or prospective adoptive parents to carry out genetic testing on children not primarily to determine an imminent healthcare need, but to provide information about the future health of the child. On occasion this has given rise to legally enforceable demands for testing.

In this paper, we consider the ethical issues that arise in light of requests for pre-adoption predictive genetic testing for familial cancer syndromes in children who are too young to engage at all with the decision-making process. We will argue that such testing, which could be commissioned by either an adoption agency or pre-adoptive parents, should in general not go ahead when the same test would not be carried out in a child who is not being adopted; particularly if there is no engagement with clinical genetics services. In making this argument, we support a principle of consistency and query claims based on the exceptionalism of adoption. Our position admittedly privileges the interests of children who are being placed for adoption over those of prospective adoptive parents, a stance we support in part due to the fact that a guarantee of a healthy child is unattainable, irrespective of adoption or genetic testing.

Genetic testing in children and the role of health information in adoption

Genetic tests are often classified into categories, characterised by the purpose of the test. This paper focuses on those genetic tests that are pre-symptomatic or predictive of a future risk of contracting cancer; either in later childhood or early adolescence (such as FAP), or mid-to-late adulthood (such as BRCA1). Clinical practice and guidelines [1] for testing young children suggest that where a child will not require medical intervention prior to the age at which he or she could be involved in the decision-making process, testing should be deferred to allow them to participate in this choice even if they are not yet fully competent. This position draws on the following ethical rationales:

- Predictive genetic testing in young children removes their right to autonomously decide once competent whether to have the particular test. A 'right not to know' one's risk for genetic conditions is recognised and many adults choose not to know their status;
- As the child grows up, the burden of knowing or imparting this information may be significant; both for the child and the parents; and
- Carrying out a predictive genetic test in a very young child may have implications for their future opportunity to obtain life or income protection insurance.

We recognise that these rationales are arguable [2–5] and are not always followed in practice. We have assumed that they provide a valid baseline for clinical practice and that in general it is preferable not to perform predictive genetic testing in young children who cannot participate at all in the decision-making process. Occasionally, young children are tested, after parents and healthcare professionals have together decided that this is the best course of action. The ethical justification for this exceptional testing is usually based on a conception of the best interests of the family as a whole, including allaying significant anxiety in parents. But such decisions are made carefully on a case-by-case basis, following significant engagement with clinical genetics services. The practice of some pre-adoption testing, in which a genetics service will never meet the parents of a child for whom a test is requested, is a significant departure from this practice.

The goal of adoption is to successfully place children with suitable families and vice versa. All children being placed for adoption are therefore referred to a paediatrician for an in-depth health assessment of their medical, developmental and emotional needs. This assessment will also consider the child's family history and his or her antenatal and birth records. However, the depth of this information may often be limited, for example due to an absent father or lack of information about risk exposure in utero. Adoption is therefore inherently uncertain and there will always be a degree of speculation about the risks a child will face during childhood and beyond. Often a range of possible outcomes is described and the professionals involved provide as much information as they reasonably can within the constraints they are working under.

The British Association for Adoption and Fostering (BAAF) has a Practice Note on genetic testing and adoption. This stipulates:

> *All children … need protection from the potentially negative effects of genetic testing. Therefore, whenever possible, unless there are convincing indications to the contrary, looked after children should have the same rights as children who are living with their birth families. The threshold for testing should be the same. Testing should never be undertaken to make a child more adoptable* [6, p. 4, emphasis added].

This position is supported by the Joint Committee of Medical Genetics 1994 recommendations on genetic testing in children [1] and the American Society for Human Genetics [6].

The cases of George and Lisa

To examine the issues that arise when a pre-adoption request for genetic testing is made by an adoption agency or a pre-adoptive couple, we shall refer to two hypothetical cases:

Case 1: George

George is a 10 month-old whose mother had FAP, a hereditary cancer syndrome which can lead to cancer in childhood, but with onset rare before the age of 12. His social circumstances are such that he is now in foster care and being prepared for adoption. The adoption agency has requested genetic testing, on the grounds that this will allow them to place him with the most suitable family.

Case 2: Lisa

Lisa is a 10 month-old whose mother had BRCA1. Her mother died in an accident shortly after she was born, and no relatives can look after her, so she is in foster care and being prepared for adoption. The adoption agency has requested genetic testing so that they can provide all available information to prospective parents.

We have chosen these cases to illustrate the different thresholds for predictive genetic testing. Testing for FAP will normally take place later in childhood, as the disease may manifest prior to George being able to autonomously decide about testing himself. If George carries the FAP mutation, his adoptive family will need to support him through regular screening tests and eventually prophylactic surgery to remove some of his bowel. But it would be unusual to test for FAP in a child as young as George. If he was not being adopted, the decision about whether or not to do a predictive test would be likely to be left until he was able to participate, at least to some extent, in the discussions; a point we discuss further below.

Testing for BRCA1 is not generally offered until adulthood and so testing Lisa would constitute a significant departure from standard clinical practice. In her case, no medical intervention will be required prior to the age at which she could reasonably be expected to make an autonomous

decision about whether or not to go ahead with predictive testing. The only claim an adoption agency or pre-adoptive couple could reasonably make in favour of testing is the adoptive family's interest in not having a child who is likely to develop a serious medical condition in adulthood.

Why adoption should not make a difference

Neither the adoption process, nor their status as 'looked after,' should justify treating George or Lisa differently from other children with the same risk status who are not being placed for adoption. One initial counter-argument to this position is that as it is legitimate for adoption agencies to provide medical information to prospective adoptive parents, why shouldn't this include genetic information as well? To argue against this may constitute unjustifiable genetic exceptionalism and a failure to recognise that adoption should set children like George and Lisa apart.

However, providing full information about George or Lisa's current health (and their risk of future illness) is different to performing a genetic test to determine this risk. It is not the genetic character of the test which is exceptional, but its predictive nature and the context of adoption per se is not enough to justify a departure from clinical guidelines. George and Lisa have the same interests as other at-risk children with respect to future autonomy and protection from stigma and discrimination. Demands for testing from adoption agencies or pre-adoptive parents (particularly where not coupled with in-depth discussion) could impact on George and Lisa's developing self-image and self-esteem and also their relationship with and expectations from their adoptive parents post-adoption. They are also already vulnerable and at risk of stigmatisation, both before and after adoption. This vulnerability deserves protection, including preserving their at-risk status so that post-adoptive parents can have full engagement with clinical genetics services, placing them in the same position as any other couple with an at-risk child.

Our argument does not imply that adoption agencies (and local authorities) should leave pre-adoptive parents in a state of ignorance. They should, of course, provide information about George and Lisa's family history (including genetic risks), but this should not automatically lead to tests that would not be offered for a child not being placed for adoption. To do so would be to act inconsistently. Timing of genetic testing should instead be determined by George or Lisa's medical need or their family context post-adoption. Additional exceptional arguments would be required on an individual case basis to justify pre-adoptive testing. In George's case therefore, this is not to argue that he should never have an FAP test in childhood. He will need to commence regular screening from around age 12 and at this point it may be more appropriate to have him tested. But this would be a matter for individual discussion and deliberation between George's adoptive parents and the clinical genetics service.

We can also query whether genetic testing will in fact make a difference. Proponents of testing may argue that without testing, many adoptions would not go ahead as pre-adoptive parents are not prepared to take on this kind of uncertainty. This could mean that otherwise healthy children may not be adopted. Yet there is no research to support this view [7]. An alternate empirical claim may be just as valid, that is: testing may actually exacerbate the problem. If George or Lisa were to receive a mutation-positive result, their chances of adoption could be lessened. There is a pressing need for empirical research to examine these competing claims in more detail. In Lisa's case, it would be very difficult to justify pre-adoptive testing, given that childhood testing (or even adolescent testing) for BRCA1 is rare and that screening may not be offered before age 30. Nevertheless, a prospective adoptive couple who were thinking of adopting Lisa may still benefit from a discussion with a clinical genetics service. But for George, on the other hand, we recognise that as children like him are tested in childhood, a particular prospective adoptive couple may have concerns about his FAP status and wish to have him tested. Were this to be the case, rather than

acquiescing immediately to this request, the prospective adoptive parents should attend the relevant clinical genetics service for in-depth discussion. They should be encouraged to engage in discussions regarding the reasons why testing is generally discouraged and to consider their rationale for testing and their ability to cope with any illness in their adoptive child.

It is, of course, in George and Lisa's interests to be placed with families who feel they could live with a risk of cancer onset (particularly in George's case) and who will be able to offer them appropriate support. If their risk status was clarified prior to adoption, prospective parents who felt they would not be able to cope with this type of medical need could exclude themselves from the process. But even if George or Lisa do not carry their respective mutations and are placed with a family who 'could not have coped' otherwise, what will happen if they then develop a different illness? Genetic testing to rule out familial cancer may unreasonably raise expectations of a healthy child, which if unfulfilled may create more difficulty for the adoptive family. Pre-adoption testing, whatever its impact on chances of adoption or parental anxiety, could come at a significant cost to the child who is tested. For this reason any discussions with pre-adoptive parents about testing pre-adoption should also canvass the possibility that testing may rule out adopting George and the acceptability of this situation.

In advancing the principle of consistency, it is also important to recognise that the nature of gene testing for FAP in young children is itself subject to debate. There is no set age at which FAP gene testing is recommended to take place; instead a case-by-case analysis is carried out with each family. We have argued that, consistent with other children at-risk of FAP, George should not be tested until he is old enough to participate (at least to an extent) in these discussions and that he should not be tested as a matter of course before being placed for adoption. But in advocating this view we recognise that George's understanding will be difficult to determine and that younger children's engagement in and understanding of testing decisions remains uncertain. An in-depth analysis of when it is acceptable to provide FAP testing in children is beyond the scope of this paper, but this does not affect our endorsement of a principle of consistency to treat George and Lisa the same as children not being placed for adoption. That is, any testing (whether pre or post-adoption) should not take place unless adoptive parents enter into dialogue with clinical genetics services, just as any other at-risk family would.

The matching argument

The principle of consistency is vulnerable to a powerful counter-argument, the 'matching argument,' in which a claim is made for special ethical responsibilities in the pre-adoption context that can justify pre-adoption requests for genetic testing. Proponents of this argument recognise that adoption is not just about children like George and Lisa and that the goal of matching should override presumptions against predictive genetic testing in children. The concerns of pre-adoptive parents must also be accounted for. This argument can be summarised as follows [8]:

- Premise 1: Every adoptive child has an interest in being placed with suitable parents;
- Premise 2: The interests of biological and adoptive children are significantly different because adoptive children have an interest in being 'matched' to suitable parents;
- Premise 3: The state must do its best to place children with suitable parents, creating a duty to obtain enough information to make a good decision. This information includes genetic information;

- Premise 4: The principle of consistency creates an unnecessary obstacle to this;
- Conclusion: Restrictions on some genetic tests called for by the principle of consistency prevents the state from exercising its duty to do its best to place children.

Premise 1 is uncontroversial, as if this were not respected then pre-adoptive children's interests would not be protected and they may come to harm. Premise 2 is, however, arguable, as the fate of genetics dictates who biological children are 'placed with' and this is not always ideal. Although children are not (and should not be) routinely removed at birth for reassignment to more suitable parents, natural 'matching' is far from perfect and most children being placed for adoption are in that situation because they have been removed from their biological families.

Premise 3 has prima facie merit, but we disagree that genetic information (as opposed to family history information) is always relevant. This leads to our querying of Premise 4, as the claim that predictive genetic testing will help the state to do its best to place children with adoptive families requires further deliberation. The lack of empirical evidence in adoption makes it difficult to prove that the state is not currently doing its best through providing family history information. Indeed, even a definition of 'the best' standard for matching has not been defined. Adoption agencies currently attempt to create a realistic parenting environment by providing pre-adoptive parents with risk information and allowing them to subsequently work with their local clinical genetics service to determine whether testing should go ahead. Arguably it is part of the experience of parenthood to deal with uncertainty and to make these kinds of decisions for children. We do accept, however, that this counter-claim is based on an assumption that information provision is effective in practice, which may not always be the case.

In querying Premise 4, we also undermine the conclusion. In advocating the matching argument, Jansen and Friedman Ross claimed that the consistency view "in effect directs state agencies to choose homes for these children on the incorrect assumption that they are healthy."[8, p. 217] Yet this overlooks the fact that children like George and Lisa can be described as 'at risk' based on their medical background. The matching argument also contains an assumption that genetic certainty is the only way by which a good match can be achieved, as family history won't provide all the information that full matching would (if it included genetic testing). Yet genetic certainty may in fact bring with it a whole new set of clinical uncertainties. Prospective adoptive parents need to consider what kinds of risks they can take on, regardless of outcome. They need to be able to access a clinical genetics service and relevant support groups. George and Lisa's best interests can be served not by having the state subject them to predictive genetic testing, but by finding and adequately supporting couples to adopt an at-risk child.

The interests of adoptive parents

Another argument in favour of pre-adoptive testing is that in choosing to adopt George or Lisa, adoptive parents will face a great deal of uncertainty about their future physical and psychological health, including George's risk of FAP or Lisa's risk of breast cancer. This uncertainty can be frustrating and stressful and the reality of information provision is far from perfect. Pre-adoptive parents also cannot influence the prenatal environment or social circumstances their prospective child has been exposed to. So if some elements of this uncertainty can be reduced, this should be done to support pre-adoptive parents to have some control over the process and reduce anxiety caused by uncertainty, even if this does depart from standard practice with non-adoptive families.

However, uncertainty is inherent in the adoption process and offering genetic testing may lead to unrealistic expectations for George or Lisa's future health or may, as we have mentioned above, introduce new uncertainties. Prospective adoptive parents will benefit from support to discuss their ideas about the consequences of certain genetic conditions and their ability to cope with them. Without this, pre-adoptive parents may decide not to adopt a child who otherwise would have been a perfect 'match' for them, when in fact they may have coped very well. Many parents whose child unexpectedly develops a serious medical condition find that they can cope far better than they would have predicted they would have, had they known of the illness in advance.

Some may reply that this claim is paternalistic and that it should be up to pre-adoptive couples to decide what kinds of information they should have access to. However, as we have stated above we are not claiming that pre-adoptive couples need to be kept in total ignorance about their child. Rather, they should have access to genetic risk information to weigh in the balance with other considerations. An important component of this access is that prospective parents should also have access to clinical genetics services, who can work together with adoption agencies and parents to help the minority of children like George and Lisa being placed for adoption who face these kinds of genetic risks.

The experience of predictive genetic testing for children in a biological family context also suggests that the desire for testing may well be allayed by an open exchange of information about the condition, discussion about attitudes to risk and provision of contacts for sources of further support. Pre-adoptive parents will be advised that predictive testing in childhood is not recommended, and the rationale for this will be explained to them. Once it is established that testing will still be available in future (after adoption), when George is old enough to participate (at least to some extent) in the discussion, or Lisa has reached a standard of capacity such that she can decide about BRCA1 testing for herself (supported by genetic counselling), the demand for testing may dissipate. This explanation and the offer of support is often sufficient to provide reassurance and to allow parents to come to terms with waiting for genetic information. Occasionally pre-adoptive or post-adoptive parents will not be able to come to terms with this, and may experience anxiety from the uncertainty. In such rare cases, as happens in a non-adoptive context, genetic professionals and parents may together decide to have a child like George tested; although it would be less likely that a request for testing for Lisa would be granted. There is no reason why such discussions would not be able to take place with adoptive parents, either pre or post adoption. However, if testing is merely commissioned by an adoption agency without appropriate deliberation with the prospective adoptive parents, a vital opportunity to make a decision in the best interests of the child and their family will have been lost.

Implications for practice

We have argued that the role of genetic information in the pre-adoptive process should be restricted to provision of genetic risk information rather than predictive genetic tests, unless medical need dictates that testing cannot be delayed or there is an in-depth discussion with pre-adoptive parents to establish a sound justification for testing now. To this end it is important that an accurate family history is taken as soon as possible when adoption seems likely. It is also important that adoption services liaise with clinical genetics departments to allow provision of accurate risk information, and to allow for prospective parents to discuss any risks that they are concerned about with a trained clinical genetics professional. Further it is vital that once adoption has taken place, the link between the adoptive family and the clinical genetics department is maintained to allow for ongoing provision of information and to arrange for discussions about testing at an appropriate time. If pre-adoptive testing is considered, the burden of proof of the exceptionalism of an individual case should be on those requesting testing, rather than assuming testing will be done as part of 'routine' workup to

provide information to adoptive parents. There is also a pressing need for further empirical research to explore the opinions of pre-adoptive parents, the role of predictive genetic testing in adoption breakdown and follow-up of cases where testing does take place.

Conclusion

The debate over pre-adoptive genetic testing can be summarised as a conflict between treating pre-adoptive children as we would any other child; and the need to provide full and accurate information to prospective adoptive parents. We have presented a pragmatic ethical argument to suggest that pre-adoptive predictive genetic testing for familial cancer in young children cannot be justified solely on the basis that the child in question is being placed for adoption. This argument is based on a rejection of the exceptionalism of the process of adoption and an endorsement of existing guidelines sanctioning testing only in situations of medical need. Predictive genetic testing for cancer can have serious and life-long implications and should not therefore be routinely undertaken by local authorities (either with or without a request from prospective adoptive parents) in the context of a pre-adoption work-up. We have also highlighted the importance of ensuring that clinical genetics services are able to stay in touch with adoptive families post-adoption to ensure that any necessary screening or follow-up can be discussed and that testing might be performed at the most appropriate stage. Any decision to test should then ideally be a matter of discussion and deliberation with children like George and Lisa and their adoptive parents, once they are old enough to engage with this process. In Lisa's case, this engagement may be limited to ensuring Lisa is appropriately informed of her at-risk status and how to access help once she is considering testing.

This argument does not, however, imply that pre-adoptive genetic testing can never take place. There may, on occasions, be circumstances where it is apparent that genetic testing would be in the best interests of a particular child and his or her prospective family. If testing is to proceed in these circumstances, it should only take place after all other possibilities to avoid testing have been carefully explored (including ethical aspects), in full collaboration with the clinical genetics service. This should include the opportunity for prospective parents to have genetic counselling to make an informed choice about their need for the genetic test information. Similarly, our argument does not rule out the additional exceptional circumstance of testing a younger child like George post-adoption. As with all cases of predictive genetic testing in younger children, competing considerations may, on occasion justify testing—such as issues of familial well-being or long-term, worsening parental anxiety. In this situation, George's adoptive parents would have exactly the same fiduciary and legal responsibilities for him as if they were his biological parents. In such circumstances the need for this information may outweigh the child's future autonomy. Again, this decision to test should be reached collaboratively, post-adoption, by the child's adoptive parents together with a thorough discussion with the clinical genetics department, maximising the chance that a fully informed decision can be made.

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