

CEJA Report 4 – A-95 Testing Children for Genetic Status

INTRODUCTION

As the Human Genome Project unfolds, the number of genetic conditions that medical professionals can test for is increasing rapidly. DNA-based tests are being developed to diagnose genetic diseases, to determine predisposition to genetically-linked disorders, and to identify carrier status. Genetic testing offers considerable benefits and some peril. However, for many genetic factors, such as carrier status or predisposition to an untreatable genetic disease, individuals may not want their genetic condition determined. Some people may simply prefer not to know that they are likely to develop a devastating disease, such as Huntington disease, for which no preventive or ameliorative therapies exist. In addition, because the disclosure of genetic status, including carrier status, can often lead to discrimination by insurers and others, individuals may decide that it is preferable not to undergo testing.

Genetic testing in childhood is an area of particular sensitivity, given its great implications for adult life. If parents have complete freedom to consent to genetic testing for their children, the testing may disclose information that precipitates discrimination against the children. Even if no discrimination results, the parents have preempted their children's right to decide, upon maturity, that they would prefer not to know their genetic status. Accordingly, unless there are important benefits for a child from diagnostic testing, the risks of testing suggest that parents should not be able to require genetic testing of their child. This position has essentially been taken by several professional groups, including the Institute of Medicine, the International Huntington Disease Working Group, the National Kidney Foundation and the United Kingdom's Working Party of Genetic Testing.^{1,2,3,4} In this report, the Council discusses the relevant issues and proposes guidelines for the genetic testing of children.

INFORMED CONSENT AND GENETIC TESTING CONCERNS

Like other diagnostic testing, genetic testing raises concerns about individual autonomy and the best interests of the patient. Ordinarily, physicians may not perform diagnostic tests on a patient without the patient's informed consent. The principle of informed consent recognizes that patients are in the best position to balance the advantages and disadvantages of diagnostic testing and judge whether testing would be beneficial. In addition, by resting decisionmaking authority with the patient, the principle of informed consent permits each person to exercise control over his or her life on matters of great personal importance.

POTENTIAL HARM OF KNOWLEDGE

Informed consent is particularly important when genetic testing is considered. Because the ability to diagnose a genetic disease or to identify the predisposition for a genetic disease may precede the development of any therapeutic interventions for the disease, a positive test result may provide information about an illness for which nothing can be done medically. Healthy persons who have a positive test for Huntington disease know that they will ultimately develop a devastating, degenerative disease and that there is nothing they can do to prevent or ameliorate it. This kind of information can be psychologically harmful and diminish the person's enjoyment of even the remaining years of health. Premature diagnosis might also unreasonably discourage patients from pursuing a particular life plan—they may fail to account for the chance that effective treatments or cures would emerge prior to onset of symptoms. The information can also have a positive effect, since it relieves the uncertainty about the risk and ensures that the person has sufficient notice of the future disease onset to take that into account when considering education, employment and other pursuits. Accordingly, some people at risk for Huntington disease undergo testing to resolve the uncertainty about their future. In short, people can differ greatly on the advantages and disadvantages of genetic testing. It is therefore essential to give each person responsibility for deciding whether to undergo testing.

DISCRIMINATION

Individual control over genetic testing decisions is important, given the historically discriminatory use of test results. Carriers of the gene for sickle-cell anemia have been denied employment as if they suffered from the disease. At least one health insurer unsuccessfully tried to deny coverage for a newborn with cystic fibrosis, even though its parents carried family-health care coverage, since the parents opted to continue the pregnancy following prenatal diagnosis.⁵ Genetic testing results may also lead to stigmatization of the individual by family members, friends or acquaintances. People may decide to forego genetic testing because of the risk of discrimination or stigmatization from positive test results. Given the potential disadvantages of genetic testing, it is clear that individuals should have control over the decision whether to undergo genetic testing, and that informed consent must be required.

DECIDING ABOUT GENETIC TESTING FOR CHILDREN

For children, the issue is more complicated since they lack decisionmaking capacity and can neither give informed consent to testing nor make an informed refusal of testing. In addition, testing would be performed before the child would be mature enough to understand the implications of test results. Pre- and post-test counseling would not be sufficiently effective. Accordingly, to preserve the child's right to decide about genetic knowledge, testing often must be deferred until the child attains a level of maturity sufficient to make medical decisions. Waiting may additionally benefit the child, as current tests sometimes are not refined enough to detect all mutations of a genetic condition. In cases where testing is not medically indicated, genetic tests should not be offered nor should they be performed at the request of the parents.

However, it would not always be in the child's interests to have genetic testing withheld until the child gains decision making capacity. Some children may be at risk for a genetic disease for which there is preventive therapy.⁷ For example, if there is a family history of retinoblastoma, a malignancy that almost always occurs in the first few years of life, genetic testing can eliminate the need for risky and costly screening, including ophthalmologic examination that might need to be performed under general anesthesia. In other cases in which a child is suffering from an unexplained illness or developmental delay, genetic testing may confirm or rule out a diagnosis, thus indicating the appropriate course of therapy or the need for further diagnostic tests. Accordingly, in some circumstances, parents must be given authority to consent to genetic testing for their children. When genetic testing during childhood may benefit the child, it is appropriate for the parents to consent to testing.

Because parents should have some, but not full, discretion to consent to genetic testing for their children, it is important to establish the extent of their decisionmaking authority. As with other medical decisions for children, parents should have discretion to accept or reject genetic testing as long as they are acting reasonably in the child's interests. Parents have a fundamental "duty to provide their child with adequate medical care."⁹ Accordingly, parents must be able to make testing decisions that will enhance their children's well-being. The parents' decision about testing should be respected as long as it reflects a reasonable balance of the advantages and disadvantages of testing.¹⁰

In sum, because children upon reaching adulthood may not want to know their genetic status, genetic testing often will not be appropriate for children. Before testing can be performed, there must be some potential benefit from the testing that can reasonably be viewed as outweighing the disadvantages. When there is such a potential benefit, parents should decide whether their children will undergo testing. In some cases, testing might be unreasonable either because it does not afford sufficient benefit generally or because it would not appreciably benefit a specific child. If parents unreasonably request or refuse testing of their child, physicians should take steps to change or, if necessary, override the parents' choices by legal means if those choices are inappropriate for adequate medical care of a child. In the next several sections of this report, the Council will apply these principles to different kinds of genetic testing.

TESTING FOR CONDITIONS FOR WHICH PREVENTIVE OR OTHER THERAPEUTIC MEASURES ARE AVAILABLE

Routine neonatal and childhood testing for certain medical conditions, including phenylketonuria and hyperthyroidism, has been a successful public health initiative for decades. Early detection of many conditions may afford an opportunity to limit the development or progression of illness; for many conditions prognosis can be improved by careful monitoring, dietary modifications or the early administration of medication. Deferring testing until the child reaches maturity may result in irreversible harm. Childhood testing can also help narrow the group of individuals at risk for a particular disease, thereby allowing physicians to target their interventions where they will do the most good and sparing many children from receiving costly, painful and anxiety-producing medical procedures.⁵ In short, for many genetic diseases, childhood testing may offer important benefits to the child. Given these benefits, genetic testing should be offered or, in some cases, required.

TESTING FOR CONDITIONS WITH PEDIATRIC ONSET FOR WHICH PREVENTIVE OR OTHER THERAPEUTIC MEASURES ARE NOT AVAILABLE

Testing for untreatable childhood diseases raises somewhat different issues from other genetic testing during childhood. Because the disease develops during childhood, it is not possible to preserve the child's ability to decide about testing upon reaching adulthood. Concerns about the child's individual autonomy are not relevant. Rather, the issue is whether the testing would be in the child's interests.

In general, reasonable people can differ on the question whether the testing would be in the child's interests. There are clear potential benefits from testing, but, because the disease is unpreventable and untreatable, the benefits may not be substantial. The primary benefit is to relieve uncertainty for both the parents and the child about the child's likelihood of developing the disease. While not perfectly analogous, studies of pediatric cancer patients indicate that the earlier children are informed of their status, the better they cope with the consequences of the condition.¹¹ Early ascertainment of risk also can give parents of an affected child time to plan for the onset of disease. Finally, if a child's genetic status would influence the parents' future reproductive decisions, and if there were no other ways to ascertain the parents' risk, this might also be grounds for testing. While these are all potential benefits, they may often have an insubstantial impact on overall physical or psychological health of the child and other members of the family. Moreover, there are many disadvantages to testing. The child's self-esteem may suffer from the knowledge of the test results, the tendency to feel responsible for the genetic risk, and the reactions of family and friends to the results.³ Insurance companies may not only deny coverage for the child's medical expenses, but also might decline to renew coverage for the rest of the family. Parents may place unnecessary limitations on the child's activities or divert their limited resources to or from the upbringing of their other children.

Generally, parents should have discretion to decide about genetic testing for childhood diseases that are unpreventable and untreatable. As previously discussed, parents should have discretion to decide about testing when a balancing of the advantages and disadvantages suggests that either testing or not testing would be a reasonable decision. Since, with unpreventable and untreatable genetic diseases, there are both benefits and risks to genetic testing, and neither the benefits or risks clearly outweigh the other, parents generally should be allowed to decide about testing for their children.

Nevertheless, physicians need to be sensitive to the possibility that the parents might not be acting in the child's interests. With surrogate decisionmaking in general, there is a chance that the surrogate will be unduly influenced by conflicts of interest and make decisions that serve the interests of the surrogate or others rather than the interests of the patient. Accordingly, if a physician suspects that parents are seeking genetic testing inappropriately, the physician should not undertake testing until there are assurances that testing will not unduly compromise the child's interests.

TESTING FOR CONDITIONS WITH ADULT ONSET FOR WHICH PREVENTIVE OR OTHER THERAPEUTIC MEASURES ARE NOT AVAILABLE

In the case of adult-onset disorders which cannot be prevented and which cannot be ameliorated, genetic testing of children generally should not be undertaken. The disadvantages of testing substantially outweigh the benefits. In such cases, as with Huntington disease, the child may decide at maturity that testing is not desired, and parental consent to testing during childhood would preclude that choice. Indeed, studies have found that adults at risk for genetic disease, including perhaps 85-90% of those at risk for Huntington disease, often decline genetic testing.^{3,13,14}

Besides the harm from a compromise of autonomy, there are other potential harms to the child from testing. The psychosocial impact of such early risk assessment has yet to be fully ascertained, but harmful effects could include diminished self-esteem, lowering of expectations by family members, and inhibitions in relationship development.¹⁵ Parents may be less willing to devote financial resources for education or other life activities on the grounds that the child will not live long enough, or healthily enough, to justify expensive investments in the child's future. Parents' attitudes towards the child may turn towards disappointment or rejections and sibling tension could arise. And, despite state government efforts to curb genetic discriminations and federal and state attempts at universal health coverage, the child's future opportunity for employment, education and insurance still may be jeopardized.⁴

The arguments for offering testing are not trivial. Resolution of parental uncertainty, chance for gradual child adjustment, acceptance within the family and financial planning are all potentially beneficial results of testing. In addition, it is possible that harm to family members resulting from not testing could be greater than testing in some instances. For some parents, the provision of professional counseling would not resolve the anxiety of constantly anticipating indications for a degenerative disease their child might not even have.

Nevertheless, parental curiosity and stress can be managed without testing and should not supersede the interests of the child in not being tested. Unless there is an unusual benefit for the child from testing, testing should be postponed until children can make the decision for themselves, as mature adults. The real need to make important life plans, particularly involving marriage, reproduction and higher education, does not come until the child has reached maturity and is capable of making medical decisions.

These conclusions apply not only to uncommon inherited diseases like Huntington disease. Genetic testing for risk of developing more common disorders such as breast cancer and Alzheimer's disease would also fall into this category. The benefits from childhood predispositional testing generally would be small. For breast cancer, careful self-examination, mammograms or possible prophylactic mastectomy can be postponed until adolescence when a person with the BRCA1 gene can decide their own course of treatment. Alternatively, if the child tests negative for a gene the child may be led to believe he or she has no chance of developing that condition, when in fact the chance of developing only the heritable version has been assessed.

While testing may not be undertaken, families should still be informed of the existence of tests and given the opportunity to discuss reasons why it is not available for children. Without such disclosure, parents will feel deceived if they later discover the existence of the tests. In addition, if they discuss the child's risk with the child (e.g., in response to questions about risk from a child who has older affected family members), they will be able to explain to the child that testing will be available at the discretion of the child when the child reaches maturity.

TESTING FOR CARRIER STATUS

In general, the justifications for carrier status testing suggest that testing should be deferred until either children reach maturity and can make their own reproductive decisions or a child becomes pregnant and a reproductive decision needs to be made on the child's behalf. Being a carrier for a recessive gene does not affect the individual's health; accordingly, there is no need to determine a

child's carrier status to ensure that the child receives appropriate medical care. The primary justification for determining carrier status is to ensure that the individual has information that can be very important to making reproductive decisions. A person's carrier status can significantly affect decisions regarding marriage as well as the use of reproductive technologies, like prenatal genetic testing, in vitro fertilization, Reimplantation genetic diagnosis, artificial insemination by donor, and surrogacy. Testing for carrier status therefore should be deferred until children have decisionmaking capacity, either because they reach the age of adulthood or because they are emancipated or mature minors. In addition, when minors are partners in a pregnancy, carrier testing should be offered to the minors or their surrogate decisionmakers to inform decisions about the pregnancy.

Not only do the benefits of carrier testing generally not justify testing before maturity, there are also important risks to testing for carrier status before maturity. Discovery of carrier status can have a variety of social, psychological and economic effects. Stigmatization of and discrimination against heterozygote individuals has been documented.¹⁹ Carrying a gene for a recessive disorder has also been shown to have a detrimental effect on insurability, employment and marriageability.²⁰ Although carriers of a recessive gene suffer no physical manifestations of the disease and may even retain a selective advantage, knowledge of carrier status nevertheless may have a harmful effect on an individual's perception of health,^{17 22} reproductive anxiety²³ or general stress.²⁴ Among children self stigmatization could result in alienation from peer groups.²⁵ Ability to cope with carrier status is also complicated when the individual has a sibling who has the disease itself. Carrier testing could raise family tension over the healthy sibling's future reproductive risks and exacerbate existing "survivor guilt."²⁶ Additionally, a child might not comprehend carrier information even with counseling, to the point that they might perceive dire physical ramifications for themselves. As with other reproductive issues, parents may improperly or insufficiently inform children about the implications of genetic information. Testing during childhood may be more detrimental than testing in adulthood because testing for some conditions elicits a high rate of false negative tests; by the time the child reaches adulthood, the testing may be much more accurate.²⁷

Finally, as with other genetic testing, carrier testing calls into question the individual's right to autonomy in medical decisionmaking.²⁸ This principle should particularly be adhered to in cases affecting reproductive decisions, about which people have very deeply and strongly felt views that often reflect important religious or other personal beliefs. Parents cannot predict what their child's reproductive choices will be at maturity.

The potential for incidental discovery of carrier status, particularly for autosomal-recessive disorders such as sickle-cell anemia or cystic fibrosis, during related genetic testing presents an additional problem for obstetricians, clinical geneticists, family physicians and pediatricians. Prenatal testing may also elicit a positive carrier status for a fetus. And in research, minors might be used as part of familial studies to establish genetic linking and their status might be uncovered in the process.²⁹ In all instances, the information should be retained by the physician and entered into the patient record. Discussion of the existence of this finding should then be taken up at the appropriate time, when reproductive issues emerge in adolescence, so that the mature individual can make a decision regarding disclosure of the information. It is important that physicians treat positive and negative results in the same way. Otherwise, if physicians only raise the existence of the testing results when the results are positive, individuals will know what the results must be. Generally, this information should not be disclosed to parents or other third parties, even if they request it. Genetic information should be maintained in a separate portion of the medical record to prevent mistaken disclosure.³⁰

TESTING FOR BENEFIT TO FAMILY MEMBERS

Situations may arise where the performance of linkage analysis to diagnose a relative, genetically counsel parents, or prenatally screen a fetus could be aided by testing a child.³¹ Or, testing and disclosure might take place for an untreatable childhood disease to help the parents make an informed decision about their own future reproduction. For example, knowing whether a child will be afflicted by a devastating genetic condition could affect parental choices about additional children. However, in the vast majority of cases, the necessary information can be obtained

without testing the child. Sufficient, accurate information about the parents' reproductive risks generally can be obtained through carrier testing of the parents. Indeed, in some cases more accurate information can be obtained through parental testing since a child might have a genetic test result not attributable to either parent, which may indicate the problematic discovery of nonpaternity. Similarly, a child need never be tested for the purpose of informing the parents about their own predispositions for a particular disease. The parents can make an informed decision to be tested directly. Ultimately, minors should not be subjected to genetic testing that is not in their interest. Genetic testing for the benefit of a family member should not be performed unless two conditions are met. First, the test results must be able to prevent substantial harm to a family member. Second, there must not be any reasonable alternate ways to avoid the harm.

GENETIC TESTING AND ADOPTION

The same respect for autonomy regarding genetic information and the child's interests should be applied to children who are candidates for adoption. Accordingly, the guidelines for genetic testing of children who are candidates for adoption are the same as for other children.

Agencies or prospective parents may request genetic testing that is normally not permitted to assess the child's risks of disease more fully; indeed, some adoptive parents have turned to adoption to have a child because of a familial risk for an inheritable disorders. However, this kind of "quality check" inappropriately treats the child as a commodity

It could be argued that, since natural parents have the option of prenatal testing and can make reproductive choices accordingly, potential adoptive parents should also have access to genetic information about a child they are considering for adoption. However, testing of a child would compromise entitlements that the fetus does not have. The potential for adopting a child with a genetic disorder should be an accepted risk of the adoption process.

NEED FOR FURTHER EDUCATION AMONG HEALTH PROFESSIONALS AND THE PUBLIC

Recent research assessing attitudes toward genetic testing has found some important differences in attitude among various types of medical practitioners.^{3,31} Compared to pediatricians and other primary care practitioners, medical geneticists on the whole exhibited far greater caution in performing genetic tests on children. These differences underscore the need for continued dialogue and consensus on these issues as genetic technology emerges.

As tests for genetic conditions proliferate, it will become increasingly difficult to resist their use, especially as industry vigorously markets tests regardless of ethical implications.^{32,33,34} Continuing medical education should incorporate current information on clinical genetics and its practical applications. Technologies such as multiplex testing (i.e., screening for several conditions at once) will further complicate issues of application and counseling. Expertise is needed in a broadening array of specialties, from obstetrics to psychiatry. There will never be enough specialists in genetic counseling to meet the need for pre- and posttest counseling. Accordingly, more physicians will need to incorporate genetic counseling skills into their practices. Given the litigious potential of clinical genetics,³⁵ coordinating a professional standard of care for application of emerging genetic technologies is vital.

The general population should also be more comprehensively educated. Genetics curriculum within school systems should be strengthened. And the lay press should be particularly careful about how it represents genetic conditions and prospects for predictive testing. All these entities must participate in ensuring equitable access to beneficial genetic innovations.

NEED FOR FURTHER RESEARCH

Research should be undertaken to better assess the advantages and disadvantages of genetic testing in children. Consideration should be given not only to physical medical benefits but also to the psychosocial impact of genetic testing. Where testing for pediatric-onset conditions, later-life

diseases, or carrier screening has already been undertaken, it would be appropriate to perform prospective and retrospective analysis of the consequences. Information regarding the transmission, retention and application of genetic information would be particularly valuable. And where requests for testing are denied, assessment of how the ongoing uncertainty impacts the family is warranted. Finally, there should be ongoing assessment of social attitudes towards genetically-affected, at-risk and carrier individuals.

RECOMMENDATIONS

For the foregoing reasons, the Council on Ethical and Judicial Affairs issues the following guidelines:

1. Genetic testing of children implicates important concerns about individual autonomy and the interests of the patients. Before testing of children can be performed, there must be some potential benefit from the testing that can reasonably be viewed as outweighing the disadvantages of testing, particularly the harm from abrogating the children's future choice in knowing their genetic status. When there is such a potential benefit, parents should decide whether their children will undergo testing. If parents unreasonably request or refuse testing of their child, physicians should take steps to change or, if necessary, use legal means to override the parents' choice. Applying these principles to specific circumstances yields the following conclusions:
 - a. When a child is at risk for a genetic condition for which preventive or other therapeutic measures are available, genetic testing should be offered or, in some cases, required.
 - b. When a child is at risk for a genetic condition with pediatric onset for which preventive or other therapeutic measures are not available, parents generally should have discretion to decide about genetic testing.
 - c. When a child is at risk for a genetic condition with adult onset for which preventive or other therapeutic measures are not available, genetic testing of children generally should not be undertaken. Families should still be informed of the existence of tests and given the opportunity to discuss the reasons why the tests are generally not offered for children.
 - d. Genetic testing for carrier status should be deferred until either the child reaches maturity, the child needs to make reproductive decisions or, in the case of children too immature to make their own reproductive decisions, reproductive decisions need to be made for the child.
 - e. Genetic testing of children for the benefit of a family member should not be performed unless the testing is necessary to prevent substantial harm to the family member.
2. When a child's genetic status is determined incidentally, the information should be retained by the physician and entered into the patient record. Discussion of the existence of this finding should then be taken up when the child reaches maturity or needs to make reproductive decisions, so that the individual can decide whether to request disclosure of the information. It is important that physicians be consistent in disclosing both positive and negative results in the same way since if physicians raise the existence of the testing results only when the results are positive, individuals will know what the results must be. This information should not be disclosed to third parties. Genetic information should be maintained in a separate portion of the medical record to prevent mistaken disclosure.
3. When a child is being considered for adoption, the guidelines for genetic testing should be the same as for other children.

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