

LETTERS TO THE EDITOR

Parents' responses to predictive genetic testing in their children

I am grateful for the opportunity to comment on the recent case report by Michie *et al*¹ on predictive genetic testing of children. In that paper and elsewhere^{2,3} the authors join with others⁴ in calling for systematic studies that will provide evidence to help resolve the various practical and ethical problems that arise in the context of predictive genetic testing of children for adult onset disorders. Unfortunately, this paper sets out the issues in a misleading fashion, and hence fails to contribute to the discussion of the real issues.

Bowel screening for tumours is generally offered to children at risk of familial adenomatous polyposis (FAP) from around the age of 10 years, and so there is a clear medical indication to carry out molecular genetic testing of children within known FAP families by that age. The principal ethical concerns about predictive genetic testing of children, the loss of the child's future autonomy as an adult to make his/her own decisions about testing, and the loss of the confidentiality to which an adult would be entitled, are therefore irrelevant in this context. This paper is written as if it sets out to contribute to the debate on these issues, but it cannot do so because these issues simply do not arise in this context. Of course the observations made in this report may be interesting in their own right, such as the parents' decision after the genetic testing not to adhere to their previously agreed policy of absolute confidentiality, but they are simply irrelevant to the questions that the paper claims to be tackling.

There may be good reasons often to defer the genetic testing of children at risk of FAP until near the age at which bowel screening would be recommended, but unfortunately these were not presented in the report and we do not know whether they had been discussed with the parents. Such reasons would include the possibility of more precise genetic testing in some families in the future, and the ability to involve a child of 8-10 years in discussions about the testing, when that is clearly impossible for most children of 2-4 years.

Instead, the portrayal of the issues by the parents, as reported in the paper, suggests that they had an inappropriately polarised view of the whole childhood testing issue. This is unfortunate, because the lack of a balanced view of the issues in this family makes it most unlikely that an anecdotal report of their experience will be helpful to other families or to professionals. Because the ethical issues of autonomy and confidentiality do not arise in this context, the question of when the children should be tested has to be one to be settled by an open discussion between parents and professionals; what will be most helpful, given that no-one has advance knowledge of the test results? Discussion of the various possible outcomes of testing should lead to a consensus that would be appropriate for each family. While every fam-

ily would like to know at once that their children are unaffected, some families would prefer their child to have been involved in the decision to be tested if (s)he were then going to be shown to be affected, and this desire to involve the child in decision making may be strong enough to lead a family to defer testing. I do not expect that any simple formula will be appropriate for every family in this predicament, and there will never be a substitute for the family discussing their detailed circumstances with a trusted clinician or counsellor.

The authors call for more "evidence" to resolve the difficult issues that arise in the predictive testing of children; in effect, they call for more of the same type of evidence, recognising that one report of a single family followed for 15 months is not enough to formulate and justify a policy. While the systematic description of such family experiences is crucial, and will provide a lot of helpful insights for professionals if the studies have been structured with care, it is far from clear that it will resolve the ethical issues that were not even present in this example. The addition of a few psychometric measures before and after testing will also not cause the ethical issues to disappear.

A focus on evidence in this area, to the exclusion of ethical reflection, may be understandable when it comes from those who are professionally engaged in gathering evidence, but it should not blind physicians and other health professionals to their obligations towards their patients and clients. When Michie and Marteau² call for "controlled studies of the short- and long-term effects of testing..." in the context of predictive testing in children, are they suggesting that predictive testing of young children at risk of Huntington's disease should be carried out so as to enable long term studies to document the resulting harm? It is necessary for professionals to decide now on a policy in relation at least to the extreme cases, such as the predictive testing of young children at risk of Huntington's disease, without evidence of the long term outcome for groups of at risk children who have been tested or denied such testing. We cannot wait 20 or 30 years to gather evidence of harm before deciding that such testing is inappropriate, particularly with the active marketing of commercial genetic tests looming close. Furthermore, I find it hard to imagine that any ethics review panel would approve such a controlled study of this type. And who knows what will count as evidence of harm in 30 years' time?

To take this debate further, we need a vigorous discussion about precisely what evidence will be helpful in guiding policy and practice; surely not just opinion polls among a selected subset of the interested parties? It will be essential that the evangelists of evidence remember that researchers have a duty of care towards their subjects, just as clinicians do towards their patients. Simply dismissing the relevance of ethical considerations is unhelpful,⁵ and the argument that some legal minors are able to contribute to discussions and decisions about difficult health matters is not a valid argument against the need for guidelines to protect those minors who are unable to do so.

To close, I would like to push further the analogy with adoption that was drawn by the mother in the case report. It is generally advised that adopted children should be brought up knowing that they are adopted, that there is an issue concerning their parents

that needs to be considered. This knowledge can be imparted gradually, as the adoptive parents consider appropriate. When older, the child is then free to decide whether or when to find out more about their biological parents, and perhaps even to make contact with them. In the same way, a child can be brought up knowing that there is a genetic condition in the family that may, in some way, be relevant to them when they are older. And when the child is older, he or she will be free to find out more about their genetic status, and may choose to have genetic testing. This is the way in which I understand the real parallels between information about one's parents (adoption) and information about one's genetic constitution.

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- 1 Michie S, McDonald V, Bobrow M, McKeown C, Marteau T. Parents' responses to predictive genetic testing in their children: report of a single case study. *J Med Genet* 1996;33:313-18.
- 2 Michie S, Marteau TM. Predictive genetic testing in children: the need for psychological research. *Br J Health Psychol* 1996;1:3-14.
- 3 Marteau TM. The genetic testing of children. *J Med Genet* 1994;31:743.
- 4 Clinical Genetics Society. The genetic testing of children: report of a Working Party of the Clinical Genetics Society (A Clarke, chairman). *J Med Genet* 1994;31:785-97.
- 5 Michie S. Predictive genetic testing in children: paternalism or empiricism? In: Marteau T, Richards M, eds. *The troubled helix - social and psychological implications of the new human genetics*. Cambridge: Cambridge University Press, 1996:177-83.

This letter was shown to Dr Michie *et al* who reply as follows.

We would like to respond to three issues that we consider key to the stance taken by Angus Clarke in his reply to our report of a single study.

(1) Clarke questions the usefulness of data from situations that have some aspects in common with, but are different from, the situation of interest. In this case, the situation of interest is whether parents should be allowed to have their children tested for genes for late onset disorders for which there are no medical interventions. Because of the difficulties of gathering data directly to inform this question, we studied the responses of parents to having their young children tested for a condition for which there is a medical intervention. Some of their responses related to the practical implications. The majority, however, related to the psychological implications of having knowledge for its own sake, with relief brought about by greater certainty. We consider further larger scale and controlled studies of this, and other conditions, will help inform the more ethically problematical situations of genetic testing for conditions for which there are no medical interventions.

(2) Clarke states: "A focus on evidence in this area, to the exclusion of ethical reflection, may be understandable when it comes from those who are professionally engaged in gathering evidence, but it should not blind physicians and other health professionals to their obligations towards their patients and clients." Evidence and ethics should not be counterposed, but used to inform and enrich each other. Similarly, those who gather evidence should not be counterposed against those who have clinical obligations to patients. Without evidence, no amount of obligation will guarantee high quality care. And those engaged in health services research