

Attitudes of Deaf Adults toward Genetic Testing for Hereditary Deafness

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Summary

Recent advances within molecular genetics to identify the genes for deafness mean that it is now possible for genetic-counseling services to offer genetic testing for deafness to certain families. The purpose of this study is to document the attitudes of deaf adults toward genetic testing for deafness. A structured, self-completion questionnaire was given to delegates at an international conference on the "Deaf Nation," held at the University of Central Lancashire in 1997. The conference was aimed at well-educated people, with an emphasis on Deaf culture issues. Eighty-seven deaf delegates from the United Kingdom returned completed questionnaires. The questionnaire had been designed to quantitatively assess attitudes toward genetics, interest in prenatal diagnosis (PND) for deafness, and preference for having deaf or hearing children. The results from this study provide evidence of a predominantly negative attitude toward genetics and its impact on deaf people, in a population for whom genetic-counseling services are relevant. Fifty-five percent of the sample thought that genetic testing would do more harm than good, 46% thought that its potential use devalued deaf people, and 49% were concerned about new discoveries in genetics. When asked about testing in pregnancy, 16% of participants said that they would consider having PND, and, of these, 29% said that they would prefer to have deaf children. Geneticists need to appreciate that some deaf persons may prefer to have deaf children and may consider the use of genetic technology to achieve this. Any genetic-counseling service set up for families with deafness can only be effective and appropriate if clinicians and counselors take into consideration the beliefs and values of the deaf community at large.

Introduction

There have been rapid advances in the molecular genetics of deafness in recent years; >60 different genes causing nonsyndromal and syndromal deafness have been discovered (Hereditary Hearing Loss 1998). It is therefore likely that diagnostic, carrier, and, possibly, prenatal genetic testing for deafness-causing genes will become part of routine clinical practice. Uptake of such testing will depend on the understanding and opinions of the populations for whom the tests are relevant (Hietala et al. 1995).

Deaf and hearing people often have different views and beliefs about genetics, primarily because deafness can be viewed from different perspectives. People who refer to themselves as culturally Deaf (written with an uppercase "D") view deafness from the cultural or sociological perspective—that deafness is a condition to be understood and preserved—as opposed to the medical perspective—that deafness is a pathology to be treated or cured (Arnos et al. 1991). The deaf (lowercase "d") community is a global category for all people with any level of hearing loss, including people who are hard of hearing or deafened and deaf people who identify primarily with the hearing world as well as with the Deaf culture. Many culturally Deaf people are positive and proud to be Deaf; they have their own language (British sign language [BSL] in the UK and American sign language [ASL] in the United States) and share a common history, social customs, and identity (Arnos et al. 1991). It is thought that, of the 70,000 prelingually deaf people in the United Kingdom, 50,000 use BSL (statistics of Royal National Institute for Deaf People [1996]). For many of these people, BSL will be their first language; this is a major indicator of Deaf cultural identity.

Culturally Deaf people are often sensitive to threats to their community; this reaction has been demonstrated clearly in the resistance to cochlear implants (Gibson 1991). Genetics is seen as a similar threat. There is anecdotal evidence to show that many Deaf people are suspicious of genetics (Holmes 1997) and fear that the use of genetic technology will reduce the numbers of deaf children born, thereby having a direct effect on the viability of the Deaf community (Grundfast and Rosen 1992). Some deaf parents have said that they will not seek genetic counseling, because they worry that they

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will be told not to have children (Israel 1995). The fear of genetic research is deeply rooted in Deaf culture, primarily because of the appalling way in which deaf people have been treated throughout history, often in the name of eugenics (Bahan 1989).

Common methodological themes running throughout the social-sciences literature as well as work by Singer (1993) and Michie et al. (1995) have been adopted in the present study. Participants for the present study were a self-selected group of deaf delegates attending a conference for deaf people. It is possible that such participants may hold views stronger than those of people from the deaf community at large.

Given the anecdotal literature, it was anticipated that many culturally Deaf adults would have a predominantly negative attitude toward genetics. The aims of the present study therefore were to describe the attitudes that a group of culturally and nonculturally deaf people have toward genetics and to look at the effect that this has on interest in prenatal diagnosis (PND) for deafness.

Subjects, Material, and Methods

Subjects

Subjects were 87 delegates attending an international conference on the “Deaf Nation,” at University of Central Lancashire, during 1997. The delegates were deaf and hearing individuals and professionals with an interest in issues relevant to the worldwide Deaf community. Consent was received, from one of the conference organizers, to put a questionnaire on every seat in a large auditorium for one of the main presentations at the conference. A presentation prior to the questionnaire distribution looked at ways in which deaf people could educate the hearing world to improve services for the deaf. A Deaf chairwoman who introduced the questionnaire tried to capture this message by informing delegates that they could make a difference to genetics services for deaf people, if they completed the questionnaire, or could exercise their right to refuse, by ignoring it. Completed questionnaires were collected as people left the auditorium.

There were 140 delegates in the auditorium; completed questionnaires were collected from 124 British individuals (response rate 89%), 83 of whom considered themselves “deaf” and 4 of whom considered themselves “hearing impaired.” Responses from hearing-impaired subjects fitted the pattern of responses from deaf subjects, so these two groups were pooled ($n = 87$).

Of the 87 individuals in the sample, 46 (53%) considered themselves culturally Deaf, and 37 (43%) identified equally with the Deaf and hearing communities; the remaining 4 participants either identified with the hearing community or gave no indication of community involvement (i.e., their questionnaires did not provide

complete data). Therefore, approximately half of the study sample could be considered culturally Deaf and the other half nonculturally Deaf. The cause of deafness in each of the participants was not assessed. The majority of participants (71/87 [82%]) were within their reproductive years (i.e., 20–49 years old). Of the 87, 37 (43%) were male, 50 (57%) were female, 51 (59%) were either married or living with a partner, and 36 (41%) were either single, divorced, or separated. Ethical approval for the project as a whole was received from a hospital ethics committee.

Questionnaire

A structured, self-completion questionnaire was designed that used original questions as well as themes from established-attitude literature. The questionnaire consisted of 11 closed questions on the following topics: opinion on whether genetic testing will do more harm than good or more good than harm (based on a question by Singer [1993]); intention to have PND for deafness; preference for having deaf or hearing children; feelings about new discoveries in genetics (based on a question by Michie et al. [1995]); thoughts on whether genetic testing for deafness devalues deaf people, and sociodemographic data. The written introduction to the questionnaire informed participants that the research would be used to educate medical professionals as to the opinions of Deaf/deaf people. No written information about genetics was given, and participants were not assessed to see whether they already had participated in genetic counseling. The questions had been developed and modified as a result of three different pilot studies. Data was analyzed by means of the statistical package SPSS (Statistical Package for the Social Sciences). Statistical analyses were performed in accordance with prior hypotheses (which looked to see whether having a particular attitude toward genetics had an effect on interest in PND for deafness). Since this was an exploratory study, specific calculations were done on specific data sets; outcomes of all calculations are documented in the Results section.

Results

Attitudes toward Genetics and Genetic Testing (Figs. 1–3)

The sample group as a whole had a negative attitude toward genetics and genetic testing for deafness. Some participants had extreme views; for example, 21% were horrified by new discoveries in genetics. Participants were five times more likely to tick negative adjectives than to tick positive adjectives, to describe how they felt about new discoveries in genetics. Of the 87 individuals, 71 (82%) ticked no positive words at all, to describe how they felt about new discoveries in genetics.

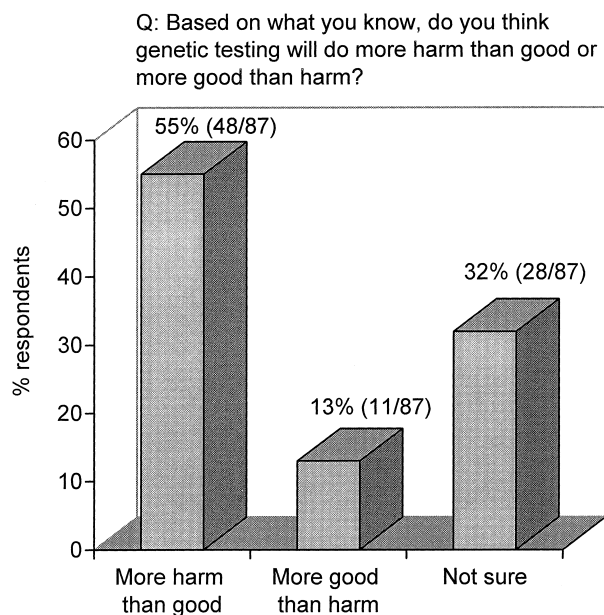


Figure 1 Attitudes toward genetic testing (1)

PND for Deafness, and Preference for Having Deaf or Hearing Children (Figs. 4 and 5)

There were trends showing that participants who thought genetic testing did more harm than good were more likely not to be interested in PND for deafness (χ^2 [with Yates’s correction] 3.5, 1 df, $P = .06$); and those who thought that the potential use of genetic testing for deafness devalued deaf people were more likely not to be interested in PND for deafness (χ^2 [with Yates’s correction] 3.4, 1 df, $P = .07$), although with Yates’s correction these were not significant. There was no association between already having children, gender, and community involvement and interest in PND.

Of the 14 participants who said that they would be interested in PND for deafness, 8 (57%) were culturally Deaf, and 6 (43%) were nonculturally deaf. Of the 4 participants who were interested in PND and preferred to have deaf children, 3 were culturally Deaf, and 1 was nonculturally Deaf.

Differences between Culturally and Nonculturally Deaf Participants

Culturally Deaf participants were more likely than nonculturally deaf participants to think that genetic testing for deafness devalued deaf people (χ^2 [with Yates’s correction] 4.1, 1 df, $P = .04$). Culturally Deaf participants were more likely than nonculturally deaf participants to think that genetic testing will do more harm than good (χ^2 [with Yates’s correction] 12.7, 2 df, $P = .002$).

Culturally Deaf participants were seven times more likely to tick negative words rather than positive words, to describe how they felt about new discoveries in genetics. There was a significant association between ticking negative words and being culturally Deaf (Mann-Whitney test $P = .0005$). There was an association between being culturally Deaf and preferring to have deaf children, but this was not significant (χ^2 [with Yates’s correction] 1.9, 1 df, $P = .2$).

Although nonculturally deaf participants demonstrated a more negative than positive attitude toward genetics and genetic testing, the majority of responses from this group were neutral. Therefore the majority were “not sure” how they felt about genetic testing; that is, 20 (50%) of 40 said that they were not sure whether either genetic testing did more harm than good or did more good than harm. Of these 40, 17 (43%) said that they were not sure whether the potential use of genetic testing for deafness devalued deaf people. Overall, nonculturally deaf participants ticked more neutral words than positive or negative ones, to describe how they felt about new discoveries in genetics.

Discussion

This study shows that some culturally and nonculturally deaf people have a negative attitude toward genetics and are concerned about the implications of genetic testing for deafness. Most participants in this study were not interested in PND for deafness, and there was a trend that showed that the participants who thought

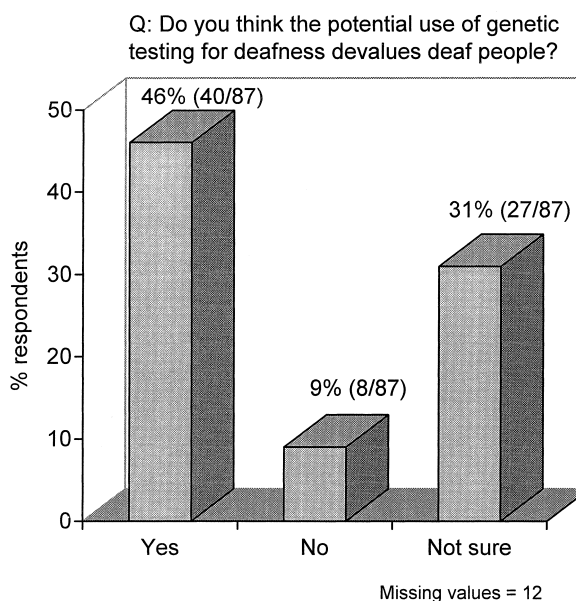


Figure 2 Attitudes toward genetic testing (2)

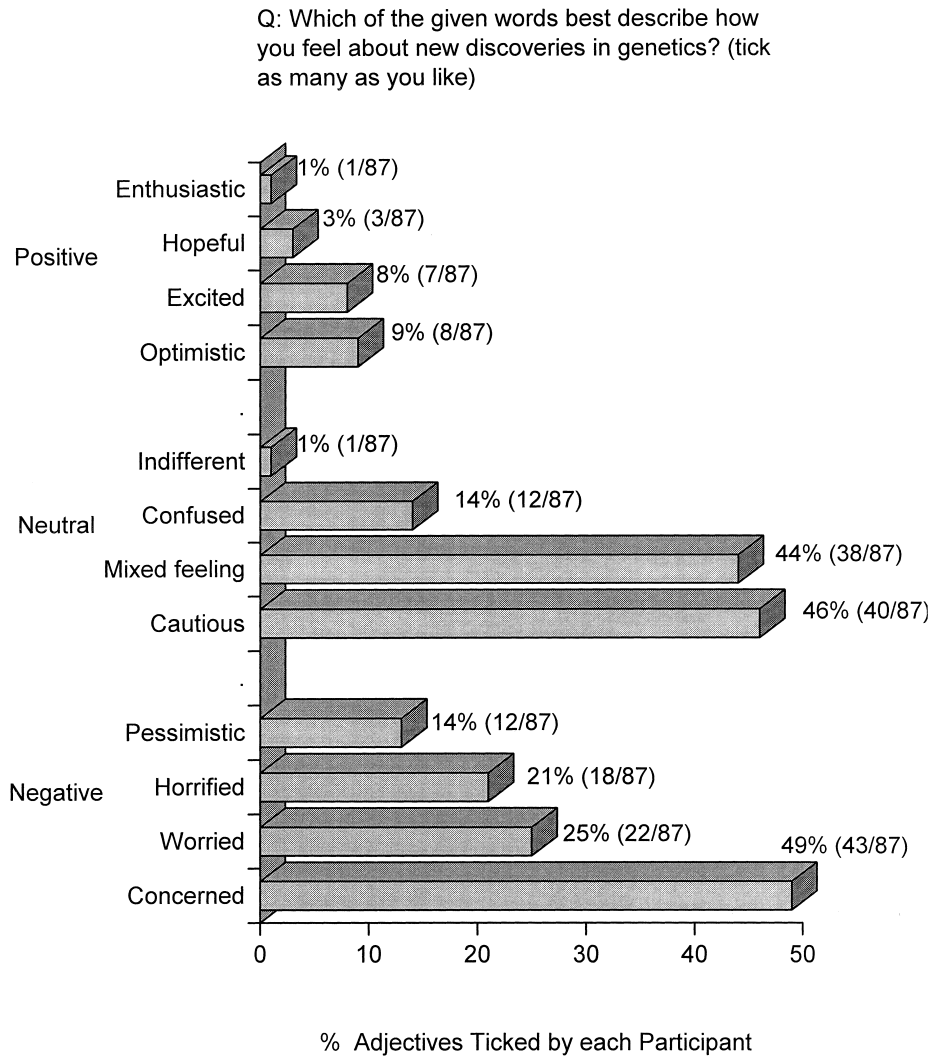


Figure 3 Words used to describe feelings about new discoveries in genetics

that genetic testing for deafness either did more harm than good or devalued deaf people were least likely to want PND for deafness. However, 13 (15%) of the 87 individuals in the entire sample, and 4 (29%) of the 14 who were interested in PND for deafness, said that they would prefer to have deaf children. These results are supported by the work of Kalla et al. (1996), who showed that 14 (19%) of a sample of 74 deaf and hard-of-hearing college students also had a preference for having deaf children. It is understandable that culturally Deaf persons may want to have deaf children, since this would allow them to pass on their language, identity, and history to the next generation, thereby keeping the Deaf culture alive.

Negative feelings about new discoveries in genetics were strong, particularly on the part of the culturally

Deaf participants. More nonculturally deaf participants ticked neutral words to describe their feelings; this fits in with results reported by Michie et al. (1995), who showed that the U.K. public also was more likely to choose neutral words to describe their feelings. Therefore, in this instance, it is clear that participants with a *defined* culture are more likely to have a *defined* view about genetics, whereas those with only partial or no cultural involvement are more likely to be less definitive about their views on genetics.

The majority (55%) of subjects thought that genetic testing would do more harm than good. This compares to a report that only 21% ($n = 1,000$) of the public from the United States think that genetic screening would do more harm than good (Singer 1993), thus indicating that deaf and hearing people may have different views about

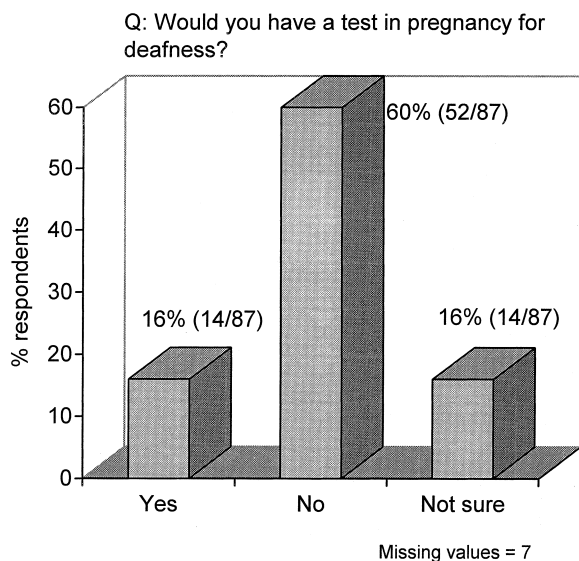


Figure 4 Interest in PND for deafness

the implications of genetic testing. The findings of the present study are consistent with the anecdotal literature, in demonstrating concern and worry about the implications of genetic testing (Grundfast and Rosen 1992; Israel 1995; Holmes 1997).

It is clear from the present study that the medical model of deafness is not a perspective that these subjects hold, irrespective of their personal cultural identity. Since the conference attended by the study participants had an emphasis on Deaf culture issues, the responses may have been influenced by the context within which the questionnaire was distributed. It could be that the deaf delegates, within the “safety” of the Deaf environment at the conference, were more likely to express openly their negativity toward genetics, compared with how they might have responded if they had been on their own in their everyday lives. Such a social desirability bias has been recognized (Bowling 1997), and all steps to limit the effect of any biases were taken into consideration, by designing the questionnaire and study in accordance with advice from the psychological and social-sciences literature (Oppenheim 1992).

To counter the effect of any biases in the present study and to document the attitudes of people with many differing perspectives on deafness, a much larger study currently is being conducted. This study involves Deaf, deaf, hard-of-hearing, deafened, and hearing individuals, from a number of different sources from all over the United Kingdom, who have given their views on genetic testing for deafness. Most previous literature has concentrated on the attitudes of the culturally Deaf and has

not considered the views of deaf persons who integrate in the hearing world; therefore, the authors’ latest research will address this issue.

Genetic counseling in general could be improved with more insight into the particular concerns and fears of clients with different disabilities. Past research has looked at lay understanding of genetics (e.g., see Chapple et al. 1995; Richards 1997) and at case-study discussion of issues relevant to clients with certain genetic conditions (Marteau and Richards 1996). However, more research is needed to fully explore particular concerns that are relevant to clients with specific genetic conditions.

One approach to improving genetic counseling for deaf people could be to use deaf genetic counselors within a clinical setting. It is unrealistic to suggest that all health professionals providing a service to people with a disability should themselves have that disability; however, it is reasonable to suggest that language and cultural barriers should be kept to a minimum, when possible. A parallel can be drawn here with genetic counseling for Asian clients, in which services could be improved by the use of Asian counselors (Darr 1997). A deaf genetic counselor would be fluent in sign language and would have a cultural awareness as well as firsthand knowledge of issues relevant to deaf people. Disability researchers repeatedly have argued for more involvement of disabled people within genetics policy making (Shakespeare, in press). Training and employment of a deaf counselor would be a positive step toward contributing a disabled voice to the genetics service.

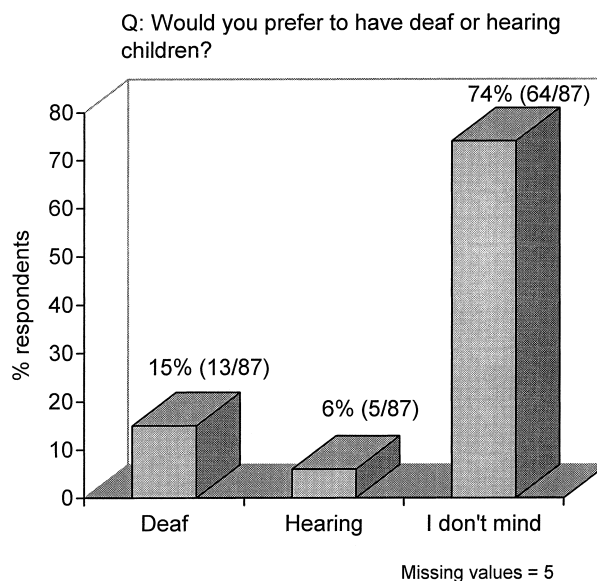


Figure 5 Preference for having deaf or hearing children

Acknowledgments

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Electronic-Database Information

URLs for data in this article are as follows:

Hereditary Hearing Loss home page (Van Camp G, Smith RJH), <http://dnalab-www.uia.ac.be/dnalab/hhh.html>

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