Health Care Anal (2010) 18:103–112
DOI 10.1007/s10728-009-0113-4

ORIGINAL ARTICLE

Public Perceptions of Ethical Issues Regarding Adult Predictive Genetic Testing

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Published online: 14 March 2009

Abstract The purpose of this study was to explore the views of members of the general public regarding ethical issues in adult predictive genetic testing. The literature pertaining to ethical issues regarding to adult predictive genetic testing is largely restricted to the views of 'experts' who have emphasized informed consent, patent issues, and insurance discrimination. Occasionally the views of patients who have undergone genetic counselling and testing have been elicited, adding psychosocial and family issues. However, the general public has not had the opportunity to contribute. In order to explore theatre as a health policy research tool, 1,200 audience members attended the play 'Sarah's Daughters' in seven Canadian cities, following which audience discussions were audiotaped. This study performed a secondary qualitative analysis of the data to identify the ethical issues of adult predictive genetic testing important to members of the general public. The identified issues were: (1) need for public education; (2) choice to undergo genetic counselling and testing; (3) access to genetic counselling and testing; and (4) obligations regarding the handling of genetic information. Audience members emphasized public education and access to information regarding potential choices, which was different from the emphasis on informed consent and other ethical issues prominent

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in the literature. Members of the general public emphasized ethical issues that were different than those identified by experts and patients. It is essential that members of the public be included in complex and controversial public policy decisions.

Keywords Health policy · Genetic testing · Ethics · Theatre · Public engagement

Introduction

The ethical implications of adult predictive genetic testing have been commented upon by ethicists, social scientists, legal scholars, policy makers and clinicians [4, 5, 12, 22, 27, 33], as well as patients undergoing genetic counselling and testing [10, 14, 17]. The ethical issues identified by these groups included: patient autonomy and informed consent; genetic discrimination [2, 7, 28]; the appropriateness of commercial genetic testing [5, 7, 13, 23, 26]; whether to allocate of resources to genetic testing services [5]; patient's rights, responsibilities and concerns with respect to their close relatives [12, 14, 16, 17, 19], and the psychosocial risks of offering genetic tests and disclosing genetic information to children [19]. However, this literature does not include the views of the general public regarding adult predictive genetic testing in general, and particularly regarding BRCA-gene-related breast and ovarian cancer.

Public engagement is a prerequisite for effective and morally legitimate health policy development, particularly in relation to complex and controversial practice [3, 6, 8, 15, 21, 25, 30]. A recent study examining the ethics of theatre as an innovative tool for public engagement in health policy development reported that audience members were engaged intellectually through a multi-directional exchange of information, and emotionally through personal identification with characters on stage [27]. It also showed that audience members were able to provide relevant opinions important to health policy development.

The purpose of the current study was to explore the ethical issues in adult predictive genetic testing important to the general public through an independent, secondary analysis of this database.

Methods

Design. This study involved an independent, secondary analysis of a previously analyzed data set exploring the ethics of using theatre as a health policy research tool [27].

Setting and Sampling. Twelve performances of the play *Sarah's Daughters* (by JN) in seven Canadian cities (Winnipeg, London, Halifax, Ottawa, Toronto, Montreal, Hamilton, and Banff) engaged approximately 1,200 audience members. The performances were advertised among the general public and the demographic make-up of the audience was not filtered. The 70 min play, *Sarah's Daughters*, is the story of a young woman living in fear of hereditary breast cancer. Script research included key informant interviews, and a search of scientific and scholarly

writings. The play was designed to incorporate scientific information about BRCA gene mutations, the prevention, diagnosis and treatment of BRCA gene-related breast and ovarian cancer, and access to genetic counselling and testing in this regard. The plot line focuses on the story of one woman, her family and a close friend, and provides the necessary scientific information in a personal context. The play in telling this story stimulates audience members to reflect in potential ethical issues, and comment if they are moved to. The play describes a women's tacit knowledge that she is at risk for hereditary breast cancer, as both women in her family, her grandmother and mother, who survives the Holocaust died of breast cancer at a young age, but that she did not qualify for BRCA gene counselling and testing in Canada as she lacked five family members with this diagnoses. A more detailed description of the play and its development was presented in a publication describes the use of theatre in health policy development [27].

Data Collection

The audience was told before the performance of the play that their participation in a research study was being requested and that their reactions/views would be sought after the performance. Following each performance of the play, a trained facilitator moderated audience discussions in the theatre audition. The facilitator asked for audience comments, answered scientific and clinical questions, clarified audience member comments if necessary, but did not steer the discussion's direction. The comments of audience were taped and transcribed for qualitative analysis. The data were collected between October 2001 and October 2003.

Data analysis. A modified thematic analysis of the database to identify and describe other ethical issues raised by audience members was conducted. Four steps were involved in the analysis: (1) the transcripts were read several times in order to become familiar with their content; (2) segments of data that related to a specific concept were identified, the relevant text was underlined, labelled (i.e. coded), and notes were written in the margins; (3) codes were compared within and between transcripts to ensure comprehensiveness and consistency; and (4) concepts that were related were grouped into themes based on these relationships.

Research ethics. Prior to and immediately following each performance, an information letter was read that emphasized that if audience members chose to remain in the theatre, their choice to participate in the research was implied, but did not require their offering of comments. Research Ethics Board approval for Non-Medical Research Involving Human Subjects was obtained from the University of Western Ontario. No participant has been identified.

Results

Four overlapping themes emerged from the audience members: (1) need for public education; (2) informed choice to undergo genetic counselling and testing; (3) access to genetic counselling and testing; and (4) obligations regarding the handling

of genetic information. In this section we describe the audience members' views about each and provide illustrative verbatim quotes.

Need for Public Education

Audience members frequently commented on the need for public education. There were concerns regarding the compromise of health resulting from lack of public education regarding BRCA gene-related breast cancer in general, as well as particular education campaigns for women at high risk so that they would have the ability to choose whether or not to pursue genetic counselling and prevention strategies.

> … it's an important issue that people need to know about, especially because it affects people of my heritage but beyond that. It's information that women can get to prevent them from passing away from disease.

Audience members felt it important for the public to have access to unbiased sources of information regarding genetic testing, such as through government-sponsored programs rather than direct-to-consumer advertising in the US. Other audience members felt it important that public education not overemphasize the genetic component of disease to the detriment of the investigation into other contributing factors to illness, such as "socio-economic or environmental conditions", as well as causing the public to neglect the possibility of non-genetic preventative measures, or to be overly worried about a disease for which they are not at high risk.

Audience members were also concerned that physician lack of information regarding BRCA-gene-related breast cancer was a contributor to the lack of public education and particularly for women at high risk and wondered, "How come doctors don't make their patients aware of this". Some audience members worried that incomplete or insufficient information among physicians could lead to a fear of broaching the subject with their patients, let alone attempting to educate them in this regard.

Informed Choice to Undergo Genetic Counselling and Testing

Participants felt that public education would promote informed choice to undergo genetic counselling, undergo genetic testing, and allow strategies of prevention of BRCA-gene-related breast cancer.

> … absolutely essential that they [women at high risk] know so that they can make sorts of choices in their lives that will permit them to live their lives in a more, I guess, more knowledgeable, and certainly a more planned fashion.

Some audience members felt that information regarding all three areas might be required for a woman to choose to talk to her physician about BRCA-gene-related breast cancer or access another entry point for genetic counselling, such as a cancer-prevention unit.

Some audience members drew attention to the possible tension between individuals desiring to be provided with choices, and that providing individuals with choices is not always in the best interests of the health care system when considered against initiatives to improve health in the general population. Although the vast majority of audience members who commented supported the imperative that women at high risk of BRCA-gene-related breast cancer be made aware through public education that they are at high risk so that they at least have the choice to access genetic counselling and preventative strategies, others offered contrary comments.

> I'm a little reluctant to make our health priorities in terms of what individual choices are when there is so much we ought to be doing about systemic changes to make people less likely to get sick.

Audience members were concerned that external pressures may affect the capacity for free choice, such as pressure from family members, either to be tested or not to be tested, because of the impact of the resulting knowledge. Audience members were also concerned regarding the impact of the choice whether or not to pursue testing could have on future choices, such as to choose not to have children.

> … there may be an ethical obligation on the part of women primarily, men too I guess, who test positive, not to have children.

and

> … for most people their life's work is to have children. And to deny them that when you have knowledge of a genetic disease … I don't think that this should even be considered.

In addition regarding women's choices, audience members were concerned that the range of women's choices could be narrowed by insurers and employers who could discriminate against women based on genetic test results, or even on having undergone genetic testing, such as in the comment.

> In the States there are several lawsuits underway because there are women who have been denied access to insurance because they've had the testing.

Access to Genetic Counselling and Testing

Audience members were concerned about ensuring equal and timely access to genetic counselling and testing for all women at high risk. The subthemes that emerged were: (1) equal access for all women at high risk; (2) inappropriate wait times; (3) concerns regarding appropriate cost of testing and the effect of patenting genetic tests; (4) appropriate criteria for genetic counselling and testing; (5) and resource allocation to genetic counselling and testing in terms of competing demands for health-care funding.

Audience members voiced concern that access to BRCA-gene-related genetic counselling was not equally accessible to all Canadians. They linked inequality in access to the ability of individuals to pay privately for testing, and to whether their

province of residence funded BRCA-gene testing and funding province's tolerance of waiting lists that can exceed 18 months.

> I am living daily with the consequences of knowing women who cannot get the BRCA testing in British Columbia … I think we have to find some answers to getting genetic testing available to women who are [at] high risk for the BRCA1 and BRCA2 mutations.

The ability of private companies to hold a patent on a genetic test was seen as inflating the cost of the test, raising questions such as "who pays for research, who owns it, and what happens to it after" when much of the research occurred in publicly-funded institutions. These questions were considered to be especially relevant to testing for the BRCA-gene which could be "so lifesaving and so necessary", rather than holding patients on discoveries that are not important to an individual's health.

Other audience members argued that improving access to adult predictive genetic testing could make economic sense as preventing breast cancer was likely less expensive than treating it. Still others were concerned that instead of accessibility to genetic counselling and testing increasing in the future, it may actually decrease due to budget cuts in the health care system.

Many of the audience members who commented were particularly concerned with "wait times" and the resulting emotional and potential physical harms to women at high risk. There were three stages of waiting mentioned: time to counselling/testing, time to receiving results, and time to strategy of prevention. Audience members described "wait times" as "sad" and linked them to funding issues. Audience members were particularly concerned that "wait times" for prevention strategies could be lethal and considered the length of waiting "tremendous", "ridiculous", and indicative of a "lack of continuum of care being provided to these women".

Audience members were also concerned regarding "limited resources" in the healthcare system and the potential "moral dilemma" in "choosing between competing alternatives". Many were concerned that resources may be spent on genetic testing that target only a small percentage of the population rather than factors affecting the broader population such as "socio-economic or environmental conditions", "smoking", and "diet". Audience members were also concerned about the discovery of "endless genes" for which genetic testing would require funding, such as the one who asked,

> … where do we go if we decide, as a society, to support and even pay for BRCA testing, what do we do with all the other genetic tests coming down the pipe …?

As a guide to funding decisions regarding particular genetic tests, participants proposed potential criteria, such as "how effective a prevention" exists, the likelihood of finding a positive result, and the age at which the disease affects individuals.

Obligations Regarding the Handling of Genetic Information

Some audience members were worried about who would have access to the results of genetic testing besides the woman and her physician, such as the woman who asked *In terms of the information, once the test is obtained, how private is the information?*

Audience members also wondered what obligation an individual has to reveal the results of genetic testing to others, such as insurance companies, or family members at risk of the genetic condition, and whether it was "ethical to suppress knowledge". Participants identified an "ethical obligation about maintaining the trust between the doctor and patient". They commented that this duty to maintain patient-physician confidentiality could lead to information being kept from family members at risk, and struggled with the issue of whether it might be justified under certain circumstances to compromise patient–physician confidentiality.

In addition, audience members linked the potential for genetics-based discrimination to the importance of maintaining the confidentiality of genetic information on the patient record. One audience member felt that genetic testing could be requested to be anonymous, similar to that possibility existing for people seeking HIV testing, as a means of circumventing privacy concerns, and asked: *Has anyone ever considered anonymous BRCA testing à la anonymous HIV testing?*

Discussion

This study is the first to use theatre to explore the views of the public regarding ethical issues pertaining to a specific health policy issue (adult predictive genetic testing). Our analysis suggested that the public *qua* theatre-audience members focussed on different ethical issues regarding adult predictive genetic testing than experts (i.e. ethicists, social scientists, legal scholars, clinicians) and patients. While the audience members emphasized the need for public education and access to equal and timely genetic counselling, the experts emphasized concerns regarding patient autonomy and informed consent [1, 19]. While the audience members felt that lack of public education (including lack of physician education) denied women at high risk of breast cancer the opportunity to have a choice regarding genetic testing at all, let alone an autonomous one, the experts focused on the importance of *patient* education [4, 11, 20]. Indeed no research emphasizing *public* education has previously occurred, including that for members at high risk of BRCA-gene-related breast cancer and other adult-onset inherited conditions. Public education regarding genetic issues has been well established in the European Union, especially the UK's Wellcome Trust, and this public education process has also used theatre [32]. However, our study was the first to focus on research and use rigorous qualitative methods to analyze audience members' comments.

Similarly, although a large literature exists regarding autonomy of the *patient* (e.g. [1, 11, 13, 17]), no previous research has identified concern for the autonomy of women (and men) who are not yet patients to become aware of their risk of developing a genetics-based disease so they can decide whether to become patients.

Audience members also emphasized the impact that policy decisions can have on individuals, as reflected in the theme of the inappropriateness of long wait times to counselling, testing and preventative care strategies.

Audience members also identified issues that had already been noted in the academic literature, such as: genetic discrimination; the appropriateness of commercial genetic testing; whether to allocate of resources to genetic testing services; patients and their families, patient's rights, responsibilities, and concerns with respect to their close relatives, and the psychosocial risks of offering genetic tests and disclosing genetic information to children. However, because of the engagement provided by the play, audience members provided a distinctly personal and fresh discussion of these issues [27].

Members of the general public are key stakeholders that should be involved in health policy decisions making because the system is designed for, and funded by, the public [3, 24, 29]. Moreover, public engagement encourages better support for policy decisions, thus improving trust and confidence in the health system [9, 31]. In addition, the findings of this study further indicate that members of the public should be involved in policy decisions, particularly those with significant value components, because the public can provide important insights not considered by researchers and scholars, and also not necessarily evident to patients—engaging the public in complex value-based decisions is a kind of 'due diligence', which leads to higher quality decision making [18].

Limitations

This research was limited in that the composition of the audience likely over-represented certain groups, particularly those interested in the content of the play, such as women with a family history of breast cancer [27]. Additionally, the results of this study can not be assumed to be generalizable, but to represent the opinions of the audience members. However, as this study included the views of several hundred members of the general public, these results provide meaningful insight into public perceptions regarding the ethics of adult predictive genetic testing.

Conclusion

The emphasis of audience members on the importance of public education and access to information regarding potential choices regarding adult predictive genetic testing added to the emphasis on informed consent and other issues prominent in the writings of ethicists, social scientists, clinicians and other policy makers, and the psychosocial and family issues surfaced by research on patients. It is essential that members of the public be included in complex, controversial and ethically charged public policy decisions.

Acknowledgments This study was supported by Genome Canada. DKM was supported by a New Investigator award from the Canadian Institutes of Health Research. Jeff Nisker's research is supported by the Canadian Institutes of Health Research (CIHR) through the Institute of Human Development, Child

and Youth Health, and Genome Canada. Douglas Martin is supported by a Career Scientist award from the Ontario Ministry of Health and Long-Term Care.

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