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Online questionnaire development: Using film to engage participants and then gather attitudes towards the sharing of genomic data ☆

A. Middleton [a,*], E. Bragin [a], K.I. Morley [b,c], M Parker [d], on behalf of the DDD Study [a]

[a] Wellcome Trust Sanger Institute, Cambridge, UK
[b] Addictions Department, Institute of Psychiatry, King's College London, UK
[c] Centre for Molecular, Environmental, Genetic and Analytic Epidemiology, Melbourne School of Population and Global Health, The University of Melbourne, Melbourne, Australia
[d] The Ethox Centre, Department of Public Health, University of Oxford, UK

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ABSTRACT

How can a researcher engage a participant in a survey, when the subject matter may be perceived as 'challenging' or even be totally unfamiliar to the participant? The Genomethics study addressed this via the creation and delivery of a novel online questionnaire containing 10 integrated films. The films documented various ethical dilemmas raised by genomic technologies and the survey ascertained attitudes towards these. Participants were recruited into the research using social media, traditional media and email invitation. The film-survey strategy was successful: 11,336 initial hits on the survey website led to 6944 completed surveys. Participants included from those who knew nothing of the subject matter through to experts in the field of genomics (61% compliance rate), 72% of participants answered every single question. This paper summarises the survey design process and validation methods applied. The recruitment strategy and results from the survey are presented elsewhere.

1. Introduction

The aim of the Deciphering Developmental Disorders (DDD) research project is to identify new genetic causes for developmental disorders in children (Firth et al., 2011). This involves testing children and their parents, using 'exome sequencing',[1] a technique in which each of the individual's 20,000 genes are explored – in this case, to uncover an explanation for the child's disability.

The process of examining every gene offered by exome sequencing, also affords the opportunity to look at genes that are not known to be related to the child's disability. The ethical dilemma here is whether to take this opportunity or not and what to do with any findings; such a dilemma can apply both to clinical sequencing in a health service and also sequencing

* Corresponding author. Address: Ethics Researcher and Registered Genetic Counsellor, Wellcome Trust Sanger Institute, Cambridge CB10 1SA, UK.
E-mail addresses: am33@sanger.ac.uk (A. Middleton), eb4@sanger.ac.uk (E. Bragin), kimorley@unimelb.edu.au (K.I. Morley), michael.parker@ethox.ox.ac.uk (M Parker).

[1] 'Exome sequencing' is the laboratory technique where the protein-coding regions of genes are examined. 'Whole genome sequencing' is the laboratory technique where the protein coding regions of genes, as well as the DNA between genes, are examined. Irrespective of the actual method used, the ethical dilemmas are the same with both techniques.

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in a research setting. The advantage of taking the opportunity to screen the exome or genome, as a whole, is that findings unrelated to the original investigation (in the case of the DDD, this was concerned with developmental disorders) could still be very relevant to the child in later life as well as to other family members. Such, 'incidental findings' could, for example, offer information about life-threatening and serious conditions. In the DDD research project, the decision was made not to seek findings other than those relating to the developmental disorder under investigation. However, we were interested in carrying out some complementary social scientific research – the Genomethics Study (Middleton et al., 2013) - to get a better understanding of stakeholder views on this important topic and uncover attitudes towards sharing of data from research sequencing projects.

Pressure is mounting from policy makers and ethicists for those who use whole genome approaches to share clinically significant incidental findings, the thinking being that it is unethical to withhold genetic information that could enable the research participant to take preventative or therapeutic action to protect their health (Affleck, 2009; Evans and Rothschild, 2012; Wolf, 2012). However, some genomic researchers are concerned that the time spent searching for, interpreting and reporting incidental findings unrelated to the research aims might jeopardise attaining those aims. Many health professionals are concerned about how such data would be managed and used in clinical practice and the potential difficulties this might generate. Empirical data is required on the attitudes of multiple stakeholders affected by and working with incidental findings and policy is urgently needed to guide researchers on what to do in terms of incidental genomic findings (Wright et al., 2011). Our study aims to gather empirical data and inform such policy.

The objectives of this research are to ascertain the attitudes of multiple stakeholders, i.e. members of the public (who could be recipients of genomic data), genetic health professionals (who would deliver the genomic data), genomic researchers (who would create the genomic data) and other health professionals (who would also interact with members of the public who have received genomic data). We aimed to investigate the following:

(1) Attitudes towards sharing of 'pertinent findings' from whole genome studies.
(2) Attitudes towards sharing of 'incidental findings' from whole genome studies.
(3) Attitudes towards receiving genetic information in different categories.
(4) Attitudes towards risk perception.
(5) Attitudes towards the sharing of raw genomic data.
(6) Attitudes towards genomic researchers having a duty to search for incidental findings.
(7) Attitudes towards who might filter genomic data.
(8) Attitudes towards possible consenting procedures for genomic studies.
(9) Socio-demographic information and how this might affect the above variables.

Each of the above themes considered complex ideas about what could be learnt from a genome or exome sequence and required a superficial level of understanding about genetics. Thus, particularly for the lay members of the public, it was important to ensure these were carefully explained in lay language. Film was used as the medium for this.

This methods paper summarises the questionnaire design process that was undertaken to develop and implement an online survey, which aimed to introduce, in an unbiased way, the ethical dilemmas raised by genomic technologies and ascertain attitudes towards these. The challenge in this was to create a questionnaire that was suitable for completion by a wide range of people. Thus, it needed to be engaging and interesting enough for participants who knew nothing of the subject matter as well as those who were experts in the field. This paper considers the extensive design process that was completed in order to meet this remit.

1.1. Overview of the genomethics study

The study adopted a mixed methods approach, utilising both quantitative and qualitative techniques. Non-parametric data was collected through 32 closed questions and analysed using descriptive statistics.

Throughout 2011–2013 a link to the online questionnaire was made available; the Wellcome Trust Sanger Institute in Cambridge, UK hosted the survey and associated website. Numerous recruitment methods were used to invite survey completion, including national media reports (e.g. coverage on the UK Channel 4 and BBC news), direct invitation from health professionals participating in the molecular DDD study through to advertisement of the study via Facebook, Twitter, a Genomethics blog and through Google ads. As the survey was online it had the capacity to be retrieved by anyone who had access to the Internet and thus also by people who had not received a direct invitation from the researchers. The complete recruitment strategy is discussed in more detail elsewhere (Middleton et al., 2014). The aim was to specifically recruit research participants who were genomic researchers, genetic health professionals, other health professionals and members of the public. Research Ethics Committee approval was granted for the study by the National Research Ethics Service for the UK (REC reference: 11/EE/0313).

The online questionnaire can be accessed at www.genomethics.org; although active online recruitment ceased on 16th July 2013 the survey can still be viewed on the website. This paper summarises the questionnaire design process that was completed to create a bespoke questionnaire. The survey used 10 films that described some of the ethical dilemmas raised by genomic technology; film was specifically chosen as a medium that could be easily incorporated in an online