What does 'respect for persons' require? Attitudes and reported practices of genetics researchers in informing research participants about research
Author(s): Fiona Alice Miller, Robin Zoe Hayeems, Li Li and Jessica Peace Bytautas
Source: *Journal of Medical Ethics*, Vol. 38, No. 1 (January 2012), pp. 48-52
Published by: BMJ
Stable URL: https://www.jstor.org/stable/23072130
Accessed: 10-08-2020 15:04 UTC

PAPER

What does 'respect for persons' require? Attitudes and reported practices of genetics researchers in informing research participants about research

Fiona Alice Miller,[1] Robin Zoe Hayeems,[1] Li Li,[2] Jessica Peace Bytautas[1]

[1]Department of Health Policy, Management and Evaluation, University of Toronto, Toronto, Ontario, Canada
[2]Department of Statistics, University of Toronto, Toronto, Ontario, Canada

Correspondence to
Dr Fiona Alice Miller, Department of Health Policy, Management and Evaluation, University of Toronto, 155 College St, 4th Floor, Toronto, ON M5T 3M6, Canada; fiona.miller@utoronto.ca

Received 15 November 2010
Revised 5 May 2011
Accepted 24 May 2011
Published Online First
16 June 2011

ABSTRACT

Background It has been suggested that researchers are obliged to offer summary findings to research participants to demonstrate respect for persons, and that this may increase public trust in, and awareness of, the research enterprise. Yet little research explores researchers' attitudes and practices regarding the range of initiatives that might serve these ends.

Methods Results of an international survey of 785 eligible authors of genetics research studies in autism or cystic fibrosis are reported.

Results Of 343 researchers who completed the survey (44% response rate), the majority agreed that their team should (i) inform participants of summary findings (90.7%) and (ii) ensure they gain an awareness of developments in the field (86.9%). Additionally, the majority reported that in practice, their team (i) informs participants of summary findings (69.4%) and (ii) provides other types of relevant non-results information (eg, state of science in the field, opportunities for research participation) (67.9%).

Conclusion Researchers endorsed the obligation of communicating with research participants by providing summary findings and other research-related information in equal measure. In light of these findings, it is suggested that while the provision of summary results may contribute to efforts to discharge the obligation of respect for persons, it may be neither a necessary nor a sufficient means to this end.

> In general, the aim of clinical research should be to treat each individual participant with the utmost respect, as an end in and of himself or herself, and as a partner in research. In support of this approach, results should be shared ...[1]
>
> Respect for persons, including respect for participants' self-determination and a recognition of their integral role in research, underlies investigators' responsibilities to make aggregate research results available to participants.[2]
>
> ...fulfilling respect for participants obligates the researcher to offer to provide a summary of research results on completion of the study...[3]

INTRODUCTION

While debate continues about researchers' obligations in reporting results about individual findings to research participants, there is a seeming consensus that researchers should offer to provide a summary or aggregate set of results to participants upon study completion.[2 4–8] As the quotes that open this article suggest, this obligation is motivated by a concern for 'respect for persons' as well as supported by guidance at the national and international levels (eg, Declaration of Helsinki's Paragraph 33).[6 9] As Fernandez *et al* (2003) argue, the principle of respect for persons 'requires that individuals who are capable of making decisions for themselves be accorded high regard, and this is embodied in the concept and practice of free and informed consent.' Yet, as they point out, 'the principle of respect has been interpreted in a limited way' and there is 'little guidance for operationalizing the principle of respect over the course of research or after its completion.' In addition to redressing this apparent omission, proponents of the obligation to report summary results to research participants argue further that the offer of research results might produce both direct and indirect benefits for the individuals involved and the research enterprise as a whole, increasing public trust in, and awareness of, the research enterprise, and providing medically relevant information that might be valued by some participants.[1–3 10]

Criticism of an obligation to report the summary results of research has been muted (though see Moutel *et al*[11] cited in the article by Puri *et al*[12]). Most criticism has been directed at the potential obligation to report individualised results, because of the moral and procedural burden this places on the research enterprise and the potential harm to research participants where personal or medical decisions are influenced by preliminary and potentially erroneous information.[13–19] Yet some critics also suggest that the generally laudable practice of informing participants about the summary findings of a study is not necessarily benign, as summary results may in fact have individual implications and thus as much potential harm as deliberately individualised results.[20]

In this paper, we seek to advance the debate by considering the communication of summary research results in relation to the broader goal of demonstrating respect for persons, and the associated interest in informing research participants about the state of science in the field and the research enterprise more broadly.

METHODOLOGY

Sample, recruitment and questionnaire

With approval from the University of Toronto Health Sciences Research Ethics Board and the

Hospital for Sick Children Research Ethics Board, we conducted an international survey of autism spectrum disorder (ASD) and cystic fibrosis (CF) genetics researchers in 2009. Our methodology has been described in detail elsewhere.[15] Here, we provide a brief overview. We generated a list of eligible researchers from a review of published scientific literature (published 2005–8 on ASD or CF genetics); all authors of eligible papers were included in our sampling frame, but the final sample of 877 potential participants (418 ASD, 459 CF) was limited to those authors with publicly available email and postal addresses. Following the Dillman-tailored method of mixed-mode survey design, we contacted potential participants up to five times over an 8-week period. All contacts were made by email, with a link and password to the survey online, except for the fourth contact by post, which enclosed a paper copy of the questionnaire. The survey instrument included non-experimental and quasi-experimental components (the latter, a factorial survey[21]). It was developed and pretested by the study team drawing on a review of the literature,[7 19 22] and previous research that pointed to the many different obligations toward participants that researchers perceive and seek to act upon.[14 20]

The data reported here derive from the non-experimental component of the survey, which assessed (i) respondent characteristics (three items); (ii) research and related practices, including efforts to provide participants with information (eg, summary results from the study in which they had participated, information about scientific findings of interest, information about relevant research studies in which they might participate) (seven items); and (iii) attitudes, including attitudes about providing research participants with summary results from the study in which they were involved, and ensuring that participants gain an awareness of developments in the scientific field of interest (12 items). The attitude questions were measured using five-point Likert scales from strongly agree to strongly disagree.

Analysis

We computed descriptive statistics for all independent variables, including frequency counts and percentages. We used cross tabulations to explore relationships between practices. Because we compared proportions from non-independent samples, we used the z-test to assess differences in the beliefs or reported practices of researchers. A probability level of <0.05 was used to determine statistical significance. Finally, we generated an index to assess the degree to which each potential respondent was involved in relevant ASD or CF genetics research (based on number and relevance of publications in which they were listed as an author), and to assess whether respondents and non-respondents differed in this regard. All statistical analyses were completed using the R (version 2.10.1, 2009) software package.

RESULTS

Response rates and characteristics of respondents

For 92 potential participants we could not locate a valid email and/or postal address after receiving returned mail and they were deemed ineligible, leaving a total of 785 eligible respondents. Of these, 343 researchers completed the survey for a final response rate of 44%; 175 (51%) were CF researchers and 168 (49%) were ASD researchers. Respondents were more likely to be authors of more relevant publications (ie, discovery research rather than incidence/prevalence studies, human subjects research rather than Biobank research, research with populations rather than case studies) than non-respondents (OR=1.8, 95% CI 1.25 to 2.69). One hundred and ninety (55%) respondents were from North America and 126 (37%) were from Europe; a further 27 (8%) were from other countries (ie, Australia, Brazil, Chile, Colombia, Hong Kong, India, Israel, Japan, Malaysia, and Taiwan). One hundred and eighty (52%) of respondents reported holding a medical degree (MD or equivalent) and 191 (56%) held a research-based doctoral degree. One hundred and sixty-four (48%) reported a primary research role in molecular genetic analysis, 149 (43%) in clinical interpretation of results, and 114 (33%) in statistical interpretation of research results.

Attitudes of researchers toward informing research participants

The majority of ASD and CF researchers (86.9%) agreed (agreed or strongly agreed) that their research team should ensure that research participants gain an awareness of developments in their respective fields. The proportions of CF and ASD researchers agreeing to this statement were essentially identical (86.8% and 86.9%, respectively) (table 1). In addition, the majority of researchers (90.7%) agreed that their research team should ensure that study participants, as a group, are informed of the summary findings from the study in which they were involved. Once again, the proportions of CF and ASD researchers agreeing to this statement were nearly identical (90.9% and 90.5%, respectively). There was no statistically significant difference between attitudes towards the obligation to ensure a general awareness of research developments and the obligation to report summary results (p=0.07).

As we have reported elsewhere, a smaller but still large majority of researchers (80.8%) agreed that their research team should inform individuals of a genetic variation that has been identified in their specific sample when the variation is judged to be clinically significant.[15] The difference between attitudes towards the obligation to report summary findings and the obligation to report individualised results is statistically significant (p<0.001).

Practices of researchers in informing research participants

With respect to the practices of research teams, the majority of researchers (69.4%) reported that their team did indeed inform participants about the summary findings from the study in which they were involved, using written information and/or workshops. Smaller proportions but still majorities reported the involvement of their research team in a number of other activities to inform participants about relevant non-results issues, using similar media: specifically, 55.1% reported that their research team informed participants about scientific knowledge

Table 1 Respondent beliefs: informing participants

Respondent beliefs	ASD (n=168)	CF (n=175)	All (n=343)
In general I believe that my research team should ensure that participants gain an awareness of developments in the field of (CF/ASD) genetics research			
Strongly agree	71 (42.3)	62 (35.4)	133 (38.8)
Agree	75 (44.6)	90 (51.4)	165 (48.1)
Neutral	15 (8.9)	18 (10.3)	33 (9.6)
Disagree	7 (4.2)	5 (2.9)	12 (3.5)
Strongly disagree	0 (0.0)	0 (0.0)	0 (0.0)
I believe my research team should ensure that study participants, as a group, are informed of the aggregate findings from studies in which they were involved*			
Strongly agree	72 (42.9)	74 (42.3)	146 (42.6)
Agree	80 (47.6)	85 (48.6)	165 (48.1)
Neutral	14 (8.3)	11 (6.3)	25 (7.3)
Disagree	2 (1.2)	4 (2.3)	6 (1.7)
Strongly disagree	0 (0.0)	0 (0.0)	0 (0.0)

Results are shown as number (%).
*1 "Null" value in CF group.
ASD, autism spectrum disorder; CF, cystic fibrosis.

in the field (ie, the results of other relevant studies); 55.4% reported that they provided participants with other research information (eg, funding news, opportunities to participate in relevant studies); and, 56.3% reported providing research participants with information about the availability of relevant health services or social supports (table 2).

When compared, the proportion of respondents who reported providing summary results (69.4%) was not significantly different (p=0.45) from the proportion of respondents who reported providing *any* of the three types of non-results information (67.9%) (ie, information about scientific knowledge in the field, other research information, or the availability of relevant health services or supports). Further, while most respondents (74.9%) provided all types of information if they provided any (ie, summary results *as well as* non-results information), some respondents provided non-results information without providing summary result information (5.5%), and vice versa (7%) (table 3).

DISCUSSION

Our study confirms that researchers from different research communities (in this case, CF and ASD genetics) believe in the goal of reporting the summary results of their research to the involved participants.[23–25] Moreover, a sizeable though smaller proportion of researchers stated that their team actually reports summary results.[24–26] Further, support for the communication of summary results was higher than support for the communication of individual results where the latter are deemed clinically significant. This fact gives weight to the suggestion that the communication of summary findings to enrolled research participants is a widely endorsed goal.

Yet these findings might also imply a challenge to conventional wisdom about researchers' responsibilities in communicating with research participants, and the ways in which an obligation to pay them due respect may be met. Researchers do more to meet their responsibilities to research participants than reporting summary results. Researchers also endorsed the goal of ensuring research participants learn about developments in the research domain of interest (CF or autism genetics), which is a partial proxy for a broader interest in informing participants about various aspects of the research enterprise. Moreover, the proportion endorsing this latter communication role was not significantly different than the proportion endorsing an obligation to report summary results, suggesting that these commitments are equally valued.

Importantly, researchers' practices reflected their commitment to broad communication. In addition to asking about practices in offering summary results, we offered researchers three examples of the kinds of information research teams sometimes provide—information about scientific knowledge in the field (findings from other studies), other research information (funding news, opportunities for participation in research) and information about available health and social supports. Small majorities of researchers reported that their teams do indeed provide this kind of non-results information to participants. Moreover, respondents reported that, in total, their teams provided this kind of non-results information just as much as summary result information. Finally and suggestively, a small proportion of respondents indicated that some teams that *do* provide information nonetheless avoided providing summary results.

These descriptive data provide some basis for normative argument. Because researchers' attitudes and practices appear to conform closely with ethics recommendations to report summary results, we draw the reader's attention to researchers' attitudes and practices related to *other* types of scientific information—which have not attracted ethical attention. Specifically, we would suggest that two normative inferences arise. The first is that the provision of summary results alone may not be *sufficient* to discharge the obligation of informing research participants. Researchers endorsed both the specific goal of offering summary results and a broader goal of informing research participants about scientific developments in the field. Thus, if the goal is to respect the personhood of research participants by providing information, summary results do not have a unique status. Summary results may indeed provide valuable or desired information, but they may not be sufficient to address the larger and, arguably more meaningful, goal of respecting the personhood of research participants by informing them about the research enterprise as a whole.

A second inference from these findings is that the provision of summary results may not be *necessary* to discharge the obligation of informing research participants. This inference is limited as our data are only suggestive on this point. However, the fact that a few research teams endeavoured to provide information

Table 2 Respondent practices: information known to be communicated to participants

Respondents practices	ASD (n=168)	CF (n=175)	All (n=343)
The research team with which I am affiliated informs participants about…			
Summary results: "…aggregate (ie, global)findings from studies in which they were involved."			
Written material and/or workshops	113 (67.3)	125 (71.4)	238 (69.4)
Nothing/don't know	55 (32.7)	50 (28.6)	105 (30.6)
Non-results information: (any of the three types of information listed below)			
Written material and/or workshops	107 (63.7)	126 (72.0)	233 (67.9)
Nothing/don't know	61 (36.3)	49 (28)	110 (32.1)
Scientific knowledge in the field: "…the findings from other relevant studies."			
Written material and/or workshops	83 (49.4)	106 (60.6)	189 (55.1)
Nothing/don't know	85 (50.6)	69 (39.4)	154 (44.9)
Other research information: "…other research information (eg, funding news, opportunities to participate in relevant studies, etc)"			
Written material and/or workshops	80 (47.6)	110 (62.9)	190 (55.4)
Nothing/don't know	88 (52.4)	65 (37.1)	153 (44.6)
Other support information: "…the availability of relevant health services or social supports."			
Written material and/or workshops	80 (47.6)	113 (64.6)	193 (56.3)
Nothing/don't know	88 (52.4)	62 (35.4)	150 (43.7)

Results are shown as number (%).
ASD, autism spectrum disorder; CF, cystic fibrosis.

Table 3 Reported practices: relationships between communication practices

Reported practices	ASD (n=168)	CF (n=175)	All (n=343)
The research team with which I am affiliated …			
Provides any type of information (summary results or non-results information)	121 (72.0)	136 (77.7)	257 (74.9)
Provides no information at all (or none that is known by the respondent)	47 (28.0)	39 (22.3)	86 (25.1)
Provides only summary results	14 (8.3)	10 (5.7)	24 (7.0)
Provides only non-results information	8 (4.8)	11 (6.3)	19 (5.5)

Results are shown as number (%).
ASD, autism spectrum disorder; CF, cystic fibrosis.

to research participants but did not provide summary results may point to judgements about the readiness of summary results for release. Summary result information may not be ready for circulation for several years after a research study begins, or may never be available for circulation (eg, where studies are terminated or results are never peer reviewed and published). Moreover, in some circumstances, summary result information may be both too provisional and too potentially harmful to *ever* communicate to research participants. This is especially likely where summary results have significant implications for individuals—as when population characteristics that research participants can readily identify in themselves or family members (eg, sex, geographic location, age at onset) are associated with a susceptibility, for example, to specific clinical outcomes.[20] In these cases, the apparent distinction between aggregate and individual results falls away,[20] and concerns related to reporting individual results come to the fore.[14 15]

CONCLUSION

Drawing on an international survey of researchers involved in either CF or autism genetics research, we demonstrate that researchers endorse equally a commitment to communicating summary results and a commitment to ensuring that participants gain an awareness of developments in the scientific field of interest. Further, researchers do not give privilege to summary results from the specific study in which the participant has been involved over and above non-results information in their communications with research participants.

We have argued that these descriptive findings support the normative inference that the focus on research results as a primary vehicle through which to honour the principle of respect for persons may be overly and inappropriately specific. Yet our intent is not to identify an additional set of universal obligations incumbent upon researchers—for example, an imperative to inform research participants about the state of science in the field of interest. Rather, our goal is to question the status of one specific practice—that of reporting summary research results—as itself a principled obligation. The obligation that researchers have and that is unquestioned is to demonstrate respect for the personhood of research participants. This includes a recognition of the independent needs and interests of research participants, and thus the importance of their own reasons for seeking to or agreeing to participate in research, rather than only the ways in which their participation may serve the ends of the research and the research team. There are surely many different ways to discharge this important obligation. Of particular relevance to the debate about the communication of research results are efforts to discharge this larger obligation by informing research participants about the state of scientific knowledge in the field of interest and the research enterprise as a whole. The provision of summary results information can contribute to efforts to meet this obligation, but it is only *one way* to do so. Ethicists and research ethics committees err if they confuse the means (ie, provision of summary findings) with the end to be served (ie, principle of respect for persons), thereby establishing an obligation to the proxy measure rather than the thing itself.

Limitations

The study reports findings from the authors of ASD or CF genetics research publications, and therefore necessarily represents this selected population. Further, the representativeness of the data is limited by the 44% response rate. This limitation is balanced by the international nature of the survey, and by the fact that, according to the index of involvement that we generated, respondents were more involved than non-respondents in the type of research where issues related to reporting results to research participants are most likely to arise.

The survey instrument we developed was designed to explore several constructs rather than attending exclusively to the topics explored in this paper. Thus, we were necessarily constrained in the detail we could capture. We were, for example, able to measure only some of the attitudes and practices that might serve to inform and engage research participants in the research enterprise. The practice data are further limited by the fact that some respondents, though they are authors, may not be sufficiently involved in the central coordination of the study to know the communication strategies used by their research team; in addition, because we did not differentiate between 'do not' and 'do not know' in the questionnaire, we cannot estimate the proportion for whom this was the case. Further, we identified only select communication media (newsletters and workshops/conferences), and do not know the quality of the communication involved. Now that this exploratory study has suggested the value of research on these topics, we hope that future studies will correct these limitations and explore these important issues in greater depth.

Acknowledgements We thank our study participants for taking the time to complete our survey as well as Genome Canada for funding this project. We also thank our clinical collaborators in the context of cystic fibrosis genetic research (Drs Peter Durie, Julian Zielenski and Mary Corey) and autism spectrum disorders genetic research (Drs Stephen Scherer, Peter Szatmari, Wendy Roberts and Lonnie Zwaigenbaum). FAM is supported by a new investigator award from the Canadian Institutes of Health Research (80495). RZH has been supported by a CADRE postdoctoral fellowship from the Canadian Institutes of Health Research and the Canadian Health Services Research Foundation, and a CF Canada Research Fellowship. Sponsors' support for this work should not imply endorsement of the conclusions, for which the authors retain sole responsibility.

Funding Genome Canada, 150 Metcalfe Street, Suite 2100, Ottawa, ON K2P 1P1, Canada.

Competing interests None.

Ethics approval This study was conducted with the approval of the University of Toronto Health Sciences Research Ethics Board and the Hospital for Sick Children Research Ethics Board.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

1. **Partridge A,** Winer E. Informing clinical trial participants about study results. *JAMA* 2002;**288**:363–5.
2. **Shalowitz D,** Miller F. Disclosing individual results of clinical research: implications of respect for participants. *JAMA* 2005;**294**:737–40.
3. **Fernandez C,** Kodish E, Weijer C. Informing study participants of research results: an ethical imperative. *IRB* 2003;**25**:12–19.

4. **Ravitsky V,** Wilfond B. Disclosing individual genetic results to research participants. *Am J Bioeth* 2006;**6**:8–17.
5. **Bredenoord A,** Kroes H, Cuppen E, *et al*. Disclosure of individual genetic data to research participants: the debate reconsidered. *Trends Genet* 2011;**27**:41–7.
6. **Knoppers B,** Joly Y, Simard J, *et al*. The emergence of an ethical duty to disclose genetic research results. *Eur J Hum Genet* 2006;**14**:1170–8.
7. **Beskow L,** Burke W, Merz J. Informed consent for population-based research involving genetics. *JAMA* 2001;**286**:2315–21.
8. **Kollek R,** Petersen I. Disclosure of individual research results in clinico-genomic trials: challenges, classification and criteria for decision-making. *J Med Ethics* 2011;**37**:271–5.
9. **World Medical Association.** *World Medical Association Declaration of Helsinki, Ethical Principles for Medical Research Involving Human Subjects*. Seoul, Korea: Ferney-Voltaire, 2008.
10. **Fernandez C.** *The Return of Research Results to Participants: An Ongoing Debate Modeled in Cancer Research*. *Harvard Health Policy Rev*, 2007;**8**:16–28.
11. **Moutel G,** Duchange N, Raffi F, *et al*. Study group communication of pharmacogenetic research results to HIV-infected treated patients: Standpoints of professionals and patients. *Eur J Hum Genet* 2005;**13**:1055–62.
12. **Puri K,** Suresh K, Gogtay N, *et al*. Declaration of Helsinki, 2008: Implications for stakeholders in research. *J Postgrad Med* 2009;**55**:131–4.
13. **Miller F,** Giacomini M, Ahern C, *et al*. When research seems like clinical care: a qualitative study of the communication of individual cancer genetic research results. *BMC Med Ethics* 2008;**9**:4.
14. **Miller F,** Hayeems R, Bytautas J. What is a meaningful result? Disclosing the results of genomic research in autism to research participants. *Eur J Hum Genet* 2010;**18**:867–71.
15. **Hayeems R,** Miller F, Li L, *et al*. Not so simple: A quasi-experimental study of how researchers adjudicate genetic research results. *Eur J Hum Genet* 2011;**19**:740–7.
16. **Meltzer L.** Undesirable implications of disclosing individual genetic results to research participants. *Am J Bioeth* 2006;**6**:28–30.
17. **Parker L.** Best laid plans for offering results go awry. *Am J Bioeth* 2006;**6**:22–3.
18. **Ossorio P.** Letting the gene out of the bottle: a comment on returning individual research results to participants. *Am J Bioeth* 2006;**6**:24–5.
19. **Klitzman R.** Questions, complexities, and limitations in disclosing individual genetic results. *Am J Bioeth* 2006;**6**:34–6.
20. **Miller F,** Christensen R, Giacomini M, *et al*. Duty to disclose what? Querying the putative obligation to return research results to participants. *J Med Ethics* 2008;**34**:210–13.
21. **Wallander L.** 25 years of factorial surveys in sociology: A review. *Soc Sci Res* 2009;**38**:505–20.
22. **Murphy J,** Scott J, Kaufman D, *et al*. Public expectations for the return of results from large-cohort genetic research. *Am J Bioeth* 2008;**8**:36–43.
23. **Partridge A,** Wong J, Knudsen K, *et al*. Offering participants results of a clinical trial: sharing results of a negative study. *Lancet* 2005;**365**:963–4.
24. **Rigby H,** Fernandez C. Providing research results to study participants: support versus practice of researchers presenting at the American Society of Hematology annual meeting. *Blood* 2005;**106**:1199–202.
25. **Di Blasi Z,** Kaptchuk T, Weinman J, *et al*. Informing participants of allocation to placebo at trial closure: postal survey. *BMJ* 2002;**325**:1329.
26. **Partridge A,** Hackett N, Blood E, *et al*. Oncology physicians and nurse practices and attitudes regarding offering clinical trial results to study participants. *J Natl Cancer Inst* 2004;**96**:629–32.