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Duty to disclose what? Querying the putative obligation to return research results to participants

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ABSTRACT

Many research ethics guidelines now oblige researchers to offer research participants the results of research in which they participated. This practice is intended to uphold respect for persons and ensure that participants are not treated as mere means to an end. Yet some scholars have begun to question a generalised duty to disclose research results, highlighting the potential harms arising from disclosure and questioning the ethical justification for a duty to disclose, especially with respect to individual results. In support of this view, we argue that current rationales for a duty of disclosure do not form an adequate basis for an ethical imperative. We review policy guidance and scholarly commentary regarding the duty to communicate the results of biomedical, epidemiological and genetic research to research participants and show that there is wide variation in opinion regarding what should be disclosed and under what circumstance. Moreover, we argue that there is fundamental confusion about the notion of “research results,” specifically regarding three core concepts: the distinction between aggregate and individual results, amongst different types of research, and across different degrees of result veracity. Even where policy guidance and scholarly commentary have been most forceful in support of an ethical imperative to disclose research results, ambiguity regarding *what* is to be disclosed confounds ethical action.

In recent years a growing chorus of enthusiasts have declared the emergence of an ethical imperative to return research results to research participants. Decrying as outmoded an earlier standard of routine non-disclosure, these commentators have called for the opposite norm: obliging researchers to routinely offer results to research participants.¹ In their seminal paper on the topic, Fernandez *et al* suggest that the disclosure of research results upholds the principle of respect for persons and the requirement that participants be treated as more than a means to an end.² Further, because awareness of research results potentially conveys risks as well as benefits, they argue that results should be disclosed only with fully informed consent.² Other scholars offer similar principled and consequentialist imperatives in arguing for an ethical duty of disclosure.^{1 3–5}

Yet some scholars have begun to question a generalised duty to disclose research results, highlighting the potential harms arising from disclosure.^{6 7} These authors challenge the extent to which the principles of respect for persons, reciprocity, beneficence, and justice are served by disclosure or violated by non-disclosure.^{7–11} They dispute the claim that these principles generate an

obligation, rather than a consideration, to disclose.⁹ Further, they note that commentators positing a duty of disclosure blur the ethically relevant distinctions between research practices and clinical care.^{7–11}

In support of this dissenting view we argue that current rationales for a duty of disclosure are neither clear nor consistent enough to form the basis of an ethical imperative. We review policy guidance and scholarly commentary regarding the duty to communicate the results of biomedical, epidemiological and genetic research to research participants and highlight wide variation in opinion regarding what should be disclosed and under what circumstances, as well as fundamental confusion about the concept of “research results.” To have policy traction, arguments about the ethics of disclosure require clearer conceptual distinctions between aggregate and individual results, amongst different types of research, and across different degrees of result veracity.

THE DUTY TO DISCLOSE: WHAT DO WE KNOW?

We gathered policy guidance and scholarly commentary regarding the communication of results to research participants from databases and the authors' files. We identified relevant international (eg, WHO, UNESCO), supranational (eg, Council of Europe) and selected Anglo-American national (US, UK, Canada, Australia) policies and legal instruments (1999–2006) using the HumGen International Database of Laws and Policies (GenBiblio; www.humgen.umontreal.ca). We also searched PubMed for literature published on the issue (1999–2006), focusing on scholarly commentary or reports of consensus development processes that describe ethical norms or imperatives (ie, what people *should* do). An increasing amount of empirical data that describes practices and attitudes (ie, what people think or do about disclosure) is available providing support for,^{12 13} and offering opposition to,^{14 15} result disclosure, but we exclude this descriptive literature from our review. (All published guidance is included in the bibliography; a list of unpublished policies and legal instruments is published online only, with guidance identified here by acronym and year.)

Some policy guidance holds that researchers have a duty to make clear to research participants what, if any, results will be disclosed to them, without implying a general duty to disclose (MRC2001; NCI2000). Most, however, suggest some duty of disclosure. This apparent consensus has led scholars to argue that an ethical duty has now emerged.^{3 16} Upon closer examination, however, this seeming consensus glosses over considerable disagreement and uncertainty regarding the

exact nature and extent of the obligation, and its putative justification.

First, there is some confusion about the basic duty at issue, in particular, between a *proactive* ethical duty to offer results to all participants and a *passive* duty to respond positively to requests for results from self-selected individual participants (CIOMS2002; NCI2000).^{4 16 17} These obligations are legally and ethically distinct, with privacy laws in many jurisdictions requiring that individuals be granted access to data about themselves when they make such a request (COE2005). By contrast there is no similar legal obligation to *offer* results; though, as Parker has noted, the offer of disclosure carries with it the “suggestion that one would be imprudent to refuse”.¹⁰ This confusion between a right of access and a duty to disclose is exacerbated by guidance that extends duty of disclosure discussions to the rights of third parties to gain access to the results of biologically related research participants.^{3 17}

Further, even where the focus is clearly on an active duty to disclose, fundamental differences remain regarding which types of results should be returned, and how. The first issue is whether the researcher must disclose aggregate (or global) study results (findings representing the sample of participants) or individual results (findings regarding the person herself). Most disclosure policies apply to aggregate results. In many, the return of aggregate results is an extension of a broader commitment to the community involved in the study (ACUNS2003; NIGMS2000),¹⁸ or to disseminate study findings widely (IEA2004; ACE2000). While some guidance specifies an ethical obligation to *offer* aggregate results which may be voluntarily declined by participants (UNESCO2003; MRC2001),² other guidance is disturbingly quiet on the question of voluntarism (NUFFIELD2003; CIOMS2002; NHMRC1999).¹⁹ Neglecting this issue may transform a duty of researchers to disclose results into a duty of participants to receive them (eg,^{1 20})—a controversial move, indeed.

Individual, rather than aggregate, research results are another matter. The obligation to disclose these is advanced with respect to genetic, pharmacogenetic, biological materials and proteomic research (EFPIA2006; TRI2005; NUFFIELD2003; UNESCO2003),^{1 3 16 21 22} but remains contentious. Some guidance frames the duty to disclose individual results so narrowly that it approximates instead a duty to warn, or a duty of care.¹ For example, such guidance suggests that individual-level research results should be disclosed only when data are valid and confirmed, have significant health implications, and a course of action to ameliorate or treat the problem is available (NBAC1999).^{3 21} Other guidance suggests that a duty to disclose applies where research results are merely clinically relevant or useful (EFPIA2006; NUFFIELD2003),²³ or have relevance for the health of the individual (EC2004; CIOMS2002; UKP2001; NHMRC1999). Others reject narrow clinical rationales for disclosure, and instead suggest that results warrant disclosure whenever they may affect an individual's interests (MRC2001), and that participants' preferences should guide decisions about what types of results should trigger the offer to disclose.^{1 24–27} Indeed, some guidance derives the duty entirely from these

preferences, without regard for the validity or clinical utility of the information.^{4 28–30}

However, many guidelines include the qualification of data veracity before disclosure is imperative, that is, individual information should be disclosed only when valid or confirmed (NUFFIELD2003; NBAC1999),^{1 21 23 31} or when its reliability (CP2002), or integrity² is established. Some guidance suggests that the duty to disclose depends on the stage of the research program, but in these cases, stage serves as a proxy for the veracity of research results (ie, the more mature the research program, the more certain the findings and the greater the obligation to disclose). For example, several guidelines hold that pharmacogenetic, biobank or genetic population research currently involves a limited duty to disclose because of its exploratory nature (EFPIA2006; IAGEG2003).^{16 21 22 31 32} Similarly, Knoppers and colleagues suggest that the duty to disclose applies only to translational or clinical trials research and not to fundamental research.³

UNDERSTANDING “RESEARCH RESULTS”

The ethical duty to disclose research results is inconsistently specified by available policies, legal instruments and scholarly commentary. It is further undermined by confused use of three core concepts. The first is the distinction between aggregate and individual results. Aggregate results may have highly individual implications. For example, Fernandez *et al*² illustrate their call for the disclosure of aggregate results with the imagined story of a childhood cancer survivor who was not informed that the research in which she had participated as a child had revealed an elevated lifetime risk of cancer for survivors such as herself, and who had therefore failed to manage her risk. Aggregate results may, on the other hand, merely indicate the incidence or prevalence of certain health conditions in a population as a whole. It is therefore not clear why the ethically-salient distinction should be drawn between type of result (aggregate versus individual) rather than type of information (population-salient versus individual-salient). In genetic research, for which a distinctive duty to disclose individual results is argued most forcefully, aggregate results may also have individual implications. For example, research that links visible phenotypic characteristics (eg, sex, specific dysmorphology, etc.) to aggregate genetic results (eg, the prevalence of specific mutations) can be interpreted as individually relevant by participants. In such cases, an ethical duty to disclose aggregate results may well conflict with an ethical duty not to disclose individually-relevant results where they have limited veracity, and restricted or uncertain clinical benefit (NCI2000; NBAC1999).^{3 21 32}

A second questionable distinction is between definitive (validated) and preliminary (unvalidated) results. This distinction is almost impossible to define and apply;³³ indeed, available guidance implies two different standards. Some argue that results should be peer reviewed before being disclosed,^{2 23 24} suggesting a standard concerning scientific rigour. More commonly, guidelines call for results to be clinically validated or confirmed (NUFFIELD2003; CP2002; MRC2001; NBAC1999), suggesting a standard concerning clinical validity. In evidence-based clinical practice these two standards converge: clinically valid results are also scientifically robust. But in the research context they may diverge, relying on different appraisal processes or achieving resolution at different stages of a research project or program. Scientific peer review² or empirical replication²³ may be required to establish scientific rigour, with study (or research program) completion a condition of disclosure (CIOMS2002).² By contrast, a standard of clinical

¹Parker¹¹ suggests that the duty to warn serves as the one clear and exceptional instance in which research results should be returned. The commentators discussed here also interpret disclosure as exceptional, but justify the return of results as a duty to disclose rather than a duty to warn, and use broader criteria than are typically relied upon for a duty to warn (ie, averting immediate and life- or severe-health-threatening harm).

validity may be met simply when researchers use technologies that are in regular clinical use (eg, routinely available, nonexperimental diagnostic tests) to generate the research data and findings. This standard is more difficult to address when the research technologies are not yet in clinical use (see²¹), or where disclosure in the research context is to be triggered by lower standards of clinical validity than are typical in the clinical context.¹ In short, the veracity of information generated in the course of research is a function not only of the progress of the research project, but also the background state of knowledge, technology, and the broader scientific agenda. For this reason, the idea that research at different stages involves inherently different duties³ provides an unsound basis for ethical policy. Further, meaningful lines cannot be drawn between fundamental or applied research, or between exploratory, transitional or clinical research for the purpose of defining a duty to disclose. Ethical obligations that rest on such fluid distinctions are sure to be frequently upset.

A final conceptual challenge involves identifying the “types” of research for which specific duties of disclosure are relevant. The duty to disclose individual research results, for example, has been specifically applied to genetic research, but the justification is often unclear or insufficient. Ravitsky and Wilfond¹ offer the most sustained argument, grounding the distinctive duty in the growing prevalence of genetic research as its scope broadens from the study of rare to more common disease, the probabilistic nature of genetic information and its capacity to inform about diverse clinical outcomes (ie, pleiotropy), and finally, by the potential impact of genetic information on family relationships, reproductive decision-making and personal identity. This defense relies on an argument of “genetics exceptionalism” that is now widely discounted,^{34 35} and explicitly countered by commentators concerned with pharmacogenetics research who seek to limit the duty of disclosure to extraordinary circumstances (EFPIA2006; NUFFIELD2003).^{16 36} While “exceptional” impacts may arise from the generation or disclosure of some genetic information, genetic information is too broad and leaky a conceptual category to ensure that these impacts arise with *all* such information, or that all *non-genetic* information is immune. Finally, if the exceptional status of genetic research does not justify the assertion of a distinctive duty of disclosure it is not clear what else might. Such an obligation cannot rest on the unique capacity of genetic research to generate individually relevant results: consider, for example, the results generated by behavioural or imaging research. This fact raises the uncomfortable prospect that ethical imperatives are driven by scholarly fashions—obligations arising wherever ethicists happen to shine their spotlights, not wherever they belong.

DISCUSSION AND CONCLUSION

A new convention is emerging in research ethics: a general ethical duty on the part of researchers to disclose research results to research participants. Several scholars have offered cogent critiques of the consequences of, and the ethical presumptions underlying, this purported duty. We argue that a fundamental lack of clarity about “what” to disclose undermines any generalised ethical obligation.

The voluminous policy guidance and scholarly commentary addressing the communication of research results has failed to clarify the nature or extent of a duty to disclose. Diverse claims have been made under the aegis of such a duty—concerning a duty to respond to participant’s requests for data access, regarding the data access needs of biological relatives of research

participants, or involving the obligation to offer, provide or require the receipt of results by participants themselves. These several obligations have been applied narrowly or more broadly to aggregate or individual results, and specified with respect to all or only some types of biomedical research. Further, these duties have been limited by consequentialist concerns about the veracity or utility of results, with consequences ranging from a narrowly conceived clinical utility which emphasises objective assessments of the veracity and value of research results, to an exclusively participant-based assessment of utility that rejects objective or researcher-defined measures.

Ambiguity of the guidance regarding the duty to disclose is compounded by the weak definition of three crucial concepts. First, regarding aggregate results: it is not clear why their disclosure should be so widely and uncritically endorsed given the profound range of possible impacts of these on individual research participants. Further, if the ethically-salient distinction is actually between those results that have relevance for individuals rather than those that have relevance only for populations, much of the guidance governing the disclosure of genetic research results will prove pyrrhic in practice. Attempts to restrict the disclosure of unreliable individual genetic research results may be trumped by the broader enthusiasm for the disclosure of all aggregate results. Second, the argument for a distinctive duty to disclose individual research results in genetic research lacks both clarity and ethical justification. Genetic research is not homogeneous with regards to the implications of its results, nor will all of these implications be “exceptional”; further, genetic research is not the only form of biomedical research that generates individual results, and the implications of these other types of research may also sometimes prove “exceptional”. Finally, attempts to restrict the disclosure of results to findings with some veracity are plagued by challenges. In the research context, standards of scientific validation of data do not necessarily align with standards of clinical validation of information, and neither standard will be adequately approximated by broad categories of research stage or type (eg, preliminary, translational). Further, it is not clear that rationales for a duty of disclosure that are consequentialist—that account for result veracity and clinical utility—can be reconciled with rationales that derive the duty of disclosure from the preferences of participants and disregard result veracity. Indeed, each rationale potentially establishes a fundamentally *different* duty of disclosure.

Scholars who argue for a duty to disclose concede that pragmatic questions remain concerning how to carry out this duty in practice.^{3 5 37} Based on this review of policy guidance and scholarly commentary, we suggest that the duty itself requires careful conceptual development. It is quite likely that this work will result in several qualitatively distinct responsibilities regarding the communication of research results to participants, resting on different ethical rationales and applying to different research situations. In the meanwhile, it is clearly premature to declare a general ethical duty to disclose.

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