A duty to recontact in genetics: context matters

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Rapid advances in genomic technologies and their increasing clinical application are driving the need for a policy on whether to recontact patients to inform them of new genetic discoveries that may have relevance to their health. Importantly, the duty to recontact is context-specific, and professionals should be offered guidance accordingly.

Particularly in genetics, old results can take on new meaning in the course of time. Moreover, newly developed genetic technologies can yield results that contradict or clarify previous findings. In oncology, for instance, finding new targets for treatment can be a long-term objective of high-throughput sequencing studies. This brings new weight to the question of whether patients or their relatives should be recontacted about new genetic information or developments that are relevant to their health or reproduction[1–4]. In the literature, there is support that recontact is to some extent ethically desirable[2]; however, uncertainty prevails about the extent of professional responsibilities[2]. Here, we provide an outline of the arguments in favour of and against recontacting and discuss factors that influence a duty to recontact. In other words, how does one decide whether to recontact or not?

Arguments in favour of and against recontact

There is a widely supported view that people have an interest in genetic information relevant to their health[2]. Therefore, respect for autonomy and beneficence are important arguments in favour of recontact. Also, empirical studies show that both professionals and patients acknowledge the health benefits of recontact[2], and returning new information can be considered part of genetic health care. However, not all patients need or want to be (re)contacted, and there are concerns regarding potential harmful effects. The principle of non-maleficence and a person's right not to know are strong arguments against recontact. Instead, patients or participants could approach genetic professionals themselves. But it is questionable whether it is appropriate to assign this responsibility to people completely. A lay person will not be aware when new information is available and, hence, when it is the best time to recontact. In addition, professionals have specific knowledge and skills regarding genetic information that contribute to role-related responsibilities[5]. Other arguments against recontact are concerns about the efforts, the feasibility and the burden it poses on professionals, although IT developments and standard policies can contribute to some extent in overcoming these practical difficulties.

Reviewing the arguments in favour of and against recontact (TABLE 1) shows a great resemblance to the arguments presented in the debate on the return of unsolicited findings[4,6]. This observation is not surprising, as people can have an interest in receiving certain unsolicited findings just as they can have an interest in new genetic results or advice.

Contextual factors

Considering the wide variety of recontact situations, the force of the arguments differs accordingly. For example, the burden for professionals is much lower when it concerns approaching one person rather than hundreds of persons. This means that the strength of duty to recontact is context-specific, and professionals should be offered guidance on how to decide if they should recontact. We identify six contextual factors that should at least be considered to make a moral judgement on whether to recontact or not. These factors include information features, costs and efforts, personal preferences, who is contacted, clinic or research setting, and time.

Several informational aspects are important. Foremost, only information that is valid should be fed back. In addition, the level of (un)certainty, the severity and probability of the condition and the possibility to act on the new information should be considered. Note that one genetic result may have different meanings for different people. Reproductive information, for example, may be of more use to a young couple than to someone whose family is complete. Or, new information on therapeutic targets may be of particular importance for a patient who is about to start treatment. Also, the nature of the newly acquired information must be compared with the previously provided information; rectifying a former result, for instance, may be a strong reason to recontact.

Recontacting people and offering them suitable medical care and genetic counselling takes time and requires certain costs and efforts, which should be proportionate to the expected benefits[4,7]. That is, before renewing contact

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Table 1 | **Arguments in favour of and against a duty to recontact**

Arguments in favour	Arguments against
Respect for autonomy requires recontact	Respect for autonomy does not imply recontact (right not to know)
Beneficence or a duty to warn requires recontact	Recontact can have harmful consequences both on an individual and a societal level (principle of non-maleficence)
Technological developments can simplify and facilitate recontact	Recontact is not feasible
Empirical studies support a desire for recontact	Recontact poses an untenable burden on professionals
Recontact can provide protection against legal claims	Health professionals become vulnerable for legal claims
Recontact is part of (genetic) health care[a]	Recontact is the patient's responsibility[a]
Recontact engages participants (scientific citizenship)[b]	Therapeutic misconception[b]

[a]Specifically for a clinical context. [b]Specifically for a research context. Table adapted with permission from REF.[6], Cell Press.

and in view of established effectiveness, the expected efforts, costs and feasibility require evaluation. This evaluation includes an assessment of the workload for recontacting but also the long-term effects, such as benefits from preventive measures that become available when a person can act on genetic risks. Hence, empirical and health technology assessment studies, and possibly even insurance and health-care coverage decisions, are necessary.

One of the main concerns with recontact is that uninvited contact may harm people in several ways, such as eliciting adverse emotional responses, potential intrusion of privacy and the violation of someone's right not to know[2,8]. Ideally, people are only recontacted if they choose to be. Therefore, before making contact, a professional must at least check whether information about a person's preferences is available. However, this is a limited solution to the risk of harming people. First, to date, recontact is often not discussed during informed consent procedures for genetic testing. Second, even if recontact is discussed at the time of testing, future relevance cannot be foreseen, and it is impossible for people to predict their future preferences. These objections can, however, be taken into account when shaping future recontact policies, for instance, by making recontact a standard element of consent procedures and designing online interfaces.

New genetic information may be important for not only the person in question but also family members. Patients or study participants may have passed away when new information becomes available. Although the duty to recontact of a professional health-care provider is stronger towards the person who had a genetic test than towards a relative, it does not automatically mean that there should be no recontact when the patient or study participant is deceased. If the information is sufficiently important, (re)contacting relatives may be desirable.

Traditionally, biomedical ethics distinguishes between clinical care and research. Whereas the primary aim of clinicians is to advance the best interest of the individual patient, the primary aim of researchers is to yield scientifically accurate and generalizable knowledge[6]. Based on a duty of care, a clinician's duty to feed back important health-related information is stronger than for a researcher. However, reciprocity and scientific citizenship are specific arguments for researchers to offer individual research results (TABLE 1). Furthermore, the distinction between research and clinical care is increasingly blurred. Therefore, although there are differences between the professional duties of clinicians and researchers, researchers also need to carefully consider their recontact obligations and — if possible — align with clinicians in their research teams.

The main issue in extending the debate on unsolicited findings to the broader definition of recontact is the time factor, which poses a challenge. It has been stressed that a duty to recontact cannot last indefinitely[7]. Subsequently, it has been questioned how long professionals have a recontact duty, and fixed time frames have been proposed[7]. However, predetermined time frames are somewhat arbitrary. Also, it seems questionable that people benefit less from new information, or make less autonomous decisions, simply because of the time factor. Hence, we propose not to focus on establishing a fixed time limit and not to consider time as an overriding factor not to recontact. If confronted with the time factor, it would be better to take into account the other factors discussed, such as the expected degree of benefit and personal preferences, where known.

Concluding remarks

The duty to recontact should be considered a prima facie moral duty rather than an absolute one. Practical guidance is needed, as professionals such as clinical geneticists, laboratory specialists, oncologists and researchers continue to encounter new genetic information. The continuous, large amount of data generated by genome-wide techniques and growing genetic knowledge, which may even provide new treatment options, make the need for a clear policy even more urgent. The six factors discussed here provide a framework for the decision whether to recontact patients when confronted with new genetic information and can be used to weigh the strength of this obligation.

1. Pyeritz, R. E. The coming explosion in genetic testing — is there a duty to recontact? *N. Engl. J. Med.* **365**, 1367–1369 (2011).
2. Otten, E. et al. Is there a duty to recontact in light of new genetic technologies? A systematic review of the literature. *Genet. Med.* **17**, 668–678 (2015).
3. David, K. L. et al. Patient re-contact after revision of genomic test results: points to consider — a statement of the American College of Medical Genetics and Genomics (ACMG). *Genet. Med.* https://doi.org/10.1038/s41436-018-0391-z (2018).
4. Carrieri, D. et al. Recontacting patients in clinical genetics services: recommendations of the European Society of Human Genetics. *Eur. J. Hum. Genet.* **27**, 169–182 (2019).
5. Wouters, R. H. P. et al. Am I my family's keeper? Disclosure dilemmas in next-generation sequencing. *Hum. Mutat.* **37**, 1257–1262 (2016).
6. Bredenoord, A. L. et al. Disclosure of individual genetic data to research participants: the debate reconsidered. *Trends Genet.* **27**, 41–47 (2011).
7. Wade, C. H. & Kalfoglou, A. L. When do genetic researchers have a duty to recontact study participants? *Am. J. Bioeth.* **6**, 26–27 (2006).
8. Ploem, C. et al. A duty to recontact in the context of genetics: futuristic or realistic? *Eur. J. Health Law* **25**, 537–553 (2018).

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