“If relatives inherited the gene, they should inherit the data.” Bringing the family into the room where bioethics happens

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Biological kin share up to half of their genetic material, including predisposition to disease. Thus, variants of clinical significance identified in each individual’s genome can implicate an exponential number of relatives at potential risk. This has renewed the dilemma over family access to research participant’s genetic results, since prevailing US practices treat these as private, controlled by the individual. These individual-based ethics contrast with the family-based ethics – in which genetic information, privacy, and autonomy are considered to be familial – endorsed in UK genomic medicine and by participants in a multi-method study of US research participants presented here. The dilemma reflects a conflict between US legal and ethical frameworks that privilege “the individual” and exclude “the family” versus actual human genetics that are simultaneously individual and familial. Can human genetics succeed in challenging bioethics’ hegemonic individualism to recognize and place the family at the center of the room where bioethics happens?

**Keywords:** ethics of disclosure; genetic information; confidentiality and consent; family genetics; relational autonomy

The genetic self is the connected self. (Philosopher Heather Widdows 2013, 1)

Many of the ethical dilemmas opened up by the applications of biotechnology offer ironically the opportunity to re-think and re-theorise human genetics as a social critique of Western individualism. (Anthropologist Monica Konrad 2005, 152, emphasis added)
Ethical dilemmas over family disclosure in the genomics era

Pay attention to dilemmas and controversies, Foucault instructed; they can expose taken-for-granted truths that are being problematized, disassembled, disarticulated (Foucault 1997). The journey traversed by the human genome project, particularly the deployment of new genomic technologies, has surely left a trail of such “problematizations.” One of the most persistent and ethically troublesome dilemmas has been over disclosure of an individual’s genetic results to family members (Wertz 2001). Given that first-degree biological relatives share 50% of their genetic material, individual research participants’ genomic results may also have significant relevance to the health and lives of family members. Like genetics itself, genetic information is both individual and shared.

Yet prevailing United States (US) laws and ethics policies1 regard genetic research information like any other protected health data – personal information belonging to the individual who gave the blood sample (“the proband”) and supported by the two core ethical commitments and practices of human subjects research: protection of individual privacy through confidentiality and respect for individual autonomy and control through informed consent (Wolf et al. 2015, 445). In short, individuals’ genetic information is private and, barring exceptional circumstances, may be shared with others only with the subject’s consent. This is but one example of a prevailing legal and ethical framework explicitly built around and for “the individual” and the protection of his or her “rights” and interests.

For a field so inherently and materially based on the family, the dominance of an individualist ethical framework for genetics is indeed curious and problematic (Fox, Spencer, and Torkamani 2018). From the science and technology of human genetics – the new genetics were developed from family linkage studies – to the diagnosis of an individual’s genetic risk, clinical and social studies have demonstrated how human genetics are an inherently family affair (Wertz 2001; Doukas and Berg 2001; Konrad 2005; Finkler 2000; Ballard et al. 2020; Knoppers 2002).

Next-generation sequencing technologies are generating so much genomic information, it is nearly inevitable to find variants in the DNA of every research participant (Chan et al. 2012; Mackley et al. 2017). Many of these are incidental findings (IFs) – research findings that were not the initial target of testing but were found “incidentally” – that are increasingly clinically actionable and judged worthy enough to return to the participants (Chan et al. 2012; Bombard, Offit, and Robson 2012).

This return of IFs to probands has reactivated a persistent and often neglected question: “what about the family?” (Wolf 2015, 437). Should these results also be offered to probands’ relatives? And what if a participant’s clinically significant IFs outlive the participant, due to death or storage in biobanks, yet consent is lacking or uncertain (Amendola et al. 2015; Tassé 2011; Ursin and Stuijfbergen 2018)? And if a proband has not communicated or has prohibited communication
of clinically significant results to relatives, do researchers have any “duty to inform” them (Rothstein 2018; Chan et al. 2012; Bombard, Offit, and Robson 2012; Knoppers 2002; Weaver 2016)? These were the novel questions raised by the unexpected discovery of clinically significant IFs among biobank samples of deceased pancreatic cancer research participants at Mayo Clinic in Rochester, Minn. These questions concerning family benefit inspired the project we present in this article.

Considering that for every individual newly diagnosed at risk, the number of family members at potential risk could increase exponentially, genome sequencing can produce populations of relatives “at risk,” as they are called (Tabor et al. 2011). What’s more, as anthropologists Kaja Finkler and Marilyn Strathern point out, using the frames “heredity” and “genetics” themselves socially produces relatives: “Genes do not just provide an individual with identity, they also … relate persons to one another and give them an identity as relatives” (Strathern 1995, 104; Konrad 2005, 13), relatives, we should note, based on biogenetic inheritance (Finkler 2000).

And here the dilemma returns: how to allow families the access necessary to benefit from individual participants’ genetic results within a legal and ethical framework that only recognizes and protects individual’s rights to privacy and autonomy but not family rights or autonomy? What to do with the ever-growing number of kin implicated, willingly or not, in probands’ genetic results but who lack any direct and legitimate access to them, including posthumously, beyond probands’ consent or exceptional circumstances?

Many social, cultural, economic, and political processes are entangled in the dominance of particular disclosure practices in particular times and places (Gordon and Paci 1997). The legal and ethical mechanisms of governance around disclosure do more than regulate and prescribe behavior. They embody, produce and reproduce particular understandings and ideals of the self and personhood, of individuals, families, societies and the relationships among them. Even more basically, they embody particular understandings of what it means to be human, and how (Widdows 2013, 2). These foundational assumptions show up in how genetic information and its communication within the family are conceived, governed and practiced.

In this article we articulate two paradigmatic approaches to ethics governing the sharing of genetic information within the family – one dominant and more individual-based, the other marginal and more family-based (Parker and Lucassen 2004). Central to their difference is the degree to which the ethics ignore (individual-based) or recognize (family-based) the special, shared nature of human genetics. These approaches embody different answers to questions about the nature of genetic information, privacy, autonomy, and individual/family relations, and different priorities when addressing the basic question, “what about the family?”

Part 1 highlights ways in which prevailing US discourse, laws, ethics, and policies favor and produce an individual approach to familial communication of
genetic research information and how, despite recent commitment to increase family access to individual’s genetic results, many of these ways continue implicitly and explicitly to reproduce individuals’ limits to family access.

Part 2 moves beyond the individualist paradigm and describes a more family-based ethics of communication that we found articulated in two different populations, one lay, the other professional. The lay population consists of participants in pancreatic cancer genetic research in the US who, in our interviews and survey, contested the accepted view of genetic information belonging only to the individual. Instead, and explicitly based on the unique, shared nature of human genetics, they took a distinctly familial approach to privacy and autonomy around communication of genetic information within the family.

The second population is an interdisciplinary collection of leaders in clinical genetics, clinical ethics and genetic law involved in the development of genomic medicine in the United Kingdom (UK). Arguing that the genomics era requires changes in foundational assumptions and practices around confidentiality and consent, they propose a reconceptualization of genomic information, ethics and law as familial (Parker and Lucassen 2004, 2018; Gilbar 2007; Dheensa, Fenwick, and Lucassen 2016, 2017; Lucassen and Hall 2012; Samuel et al. 2017; Widdows 2013; Royal College of Physicians, Royal College of Pathologists and British Society for Genetic Medicine 2011, 2019).

We conclude by asking, whose ethics do official US bioethics actually represent? We call for the right ethics for the right job that adequately reflect and build on the shared nature of human genetics and of human being, which necessarily require bringing the family, and in turn the group, into a bioethical framework.

The genesis of the project

It was a first-hand experience of a moral dilemma around disclosure to relatives that sparked this project on communication of genomic IFs to relatives. While directing the bioethics program at Mayo Clinic in Rochester, Minn., co-author anthropologist/bioethicist Dr Barbara Koenig was approached by a Mayo Clinic genetic epidemiologist, Dr Gloria Petersen, with a dilemma: Petersen ran two biobanks that collected health records and biological samples (including DNA) from patients diagnosed with pancreatic cancer and their biological relatives: one from a biobank of cases, another from patients with strong family history of the disease.

The purpose of the biobanks was to contribute to pancreatic cancer gene discovery. However, in conducting this genomic research, in addition to discovering the sought-after novel pancreatic gene variants in the samples of the diseased probands, Dr Petersen’s team unexpectedly revealed strong associations with three other known disease genes, variants in genes conferring risk of hereditary breast and ovarian cancer (BRCA2), malignant melanoma (CDKN2A/p16), and carrier status for cystic fibrosis (CFTR) (Petersen, de Andrade, and Goggins 2006;
Zhen et al. 2015). Given their potential treatability, preventability, and impact on health and reproductive choices, results of these three gene variants were traditionally offered to patients in clinical care but not in research studies.

The Mayo team was sitting on the results of about 75 deceased pancreatic cancer patients that could potentially benefit their living family members. Consent to share these results with relatives, however, had neither been requested nor granted from the deceased probands. This raised the question to Dr Petersen’s team: should these research IFs be shared with surviving relatives, even without the consent of the proband? Did the standard policy that prohibited sharing without consent still make sense? This constituted a “moral dilemma” for the researchers (Couzin-Frankel 2014).

To address this “bioethics crisis,” Dr Petersen of the Mayo Clinic engaged Dr Barbara Koenig research at Mayo, to formulate a plan. They soon enlisted the assistance of University of Minnesota law professor, Susan Wolf. Joining forces, the team applied for and received a grant from the National Institutes of Health. The project explored questions of law and policy about the return of genetic IFs to family members, with or without consent of the deceased proband, among two main stakeholder groups, one professional, one lay.

First the team convened a multi-disciplinary “Working Group” of experts from the US and Canada. Over a three year period they produced guidelines for policy makers, Institutional Review Boards (IRBs), funders, and researchers for addressing questions around familial disclosure of genomic information. The Working Group’s analysis and recommendations on “Returning a Research Participant’s Genomic Results to Relatives,” together with a large set of background papers, were published in a special issue of The Journal of Law, Medicine & Ethics (Wolf et al. 2015). Given the family focus and the breath and expertise of this Working Group, we refer to some of their discourse and guidelines in our synthesis of prevailing and evolving US approaches to familial communication of genetic research information.

The second stakeholder group was patients and families recruited through two pancreatic cancer research projects affiliated with the Mayo Clinic: the patients from the pancreatic cancer biobank; the family members, including unrelated blood relatives and spouse/partners, from the pancreatic cancer family registry (Radecki Breitkopf et al. 2015). With the aim of providing empirical data to inform the Working Group’s developing policy, two studies of patient and family stakeholder “preferences” and reasonings about sharing genetic information, particularly the 3 IFs, within the family were conducted, first through semi-structured interviews (see Gordon et al. 2019 for full description of the study), then by survey (see Radecki Breitkopf et al. 2015, 2018 for full description of the survey).2

In her dual role of anthropologist/bioethicist and Co-Principal Investigator of this National Cancer Institute-funded project, Koenig served as a bridge by participating in all phases of the normative and empirical components of the project.
Working both inside and alongside teams of researchers, bioethicists, clinicians, policy makers, and research participants, she developed an “embedded ethics” model.

Part 1. US bioethics’ individual-based ethics of disclosure of genetic information within the family

A typical scenario captures the individual focus of this prevailing ethics of disclosure of genetic information: Two members of the same family arrive together to participate in a genetic research project, and to protect each person’s confidentiality, are quickly led into two separate rooms with two different genetic counselors. Because an individual’s genetic status is only understandable in the context of a family history, consultations usually begin by constructing a family tree based on family information of living and deceased relatives collected without explicit consent (Parker and Lucassen 2004). Even within a family context, each person individually consents to participate in a study and gives a blood sample for analysis, a sample, we should remember, that includes genetic material shared with relatives. The research analysis of each sample, however, is returned as the exclusive private property – the “individual’s genetic information” – of each study participant.

While guidelines vary nationally and locally (Branum and Wolf 2015; Dheensa et al. 2016), the default model of sharing genetic information currently dominant in the US – what Parker and Lucassen call “the personal model” – holds that individuals’ genetic information, like all health information, must be kept confidential, and barring exceptional conditions, should not be disclosed to relatives without the individual’s consent (2004, 166).

Protection of individual privacy remains a relatively unquestioned, almost sacred, absolute good, and it is generally assumed that people are concerned about “their privacy” and that it is professionals’ and institutions’ responsibility to protect it. And protect it they do, even up to 50 years post-death as required by institutions governed by Health Insurance Portability and Accountability Act (HIPAA) rules.

The “no disclosure without consent” policy enacts the second unquestioned pillar of the ethical framework, autonomy, i.e. individuals’ right to control their genetic information, including withholding it from relatives pre-and post-mortem (Wolf et al. 2015; Parker and Lucassen 2018; Dheensa, Fenwick, and Lucassen 2016).

As effective interventions for genetic risks are increasingly available and the number of relatives potentially affected rapidly increases as genetic testing expands, much is at stake in sharing results with relatives. Professionals reveal strong commitment and a sense of moral obligation and responsibility to make this sharing happen. The problem, however, is that it is difficult to offer these potentially valuable results when they are safely guarded inside these deeply
rooted protective laws and ethics practices. The available avenues for disclosure are mostly indirect through the probands — offering education, encouragement and possibly some moral pressure —, sometimes through health providers, and occasionally through the few legal pathways for exceptional or specific circumstances (ASHG 1998; Wolf et al. 2015).

In the US and elsewhere, this challenge of expanding family access to individual participants’ results is most often framed as a potential conflict between two distinct entities — “the individual,” who is clearly favored, and “the family” — or more specifically, between “protecting individual privacy” vs. the “health benefit to relatives” (Lucassen and Gilbar 2018; Dheensa et al. 2016; Gilbar 2007). Branum and Wolf framed it in terms of “balancing:”

Genomic researchers and professionals across the world face the challenge of balancing the privacy and wishes of individuals against the potential utility of genetic information for relatives. We need ways that American policy can better reconcile the priorities of privacy protection, respect for autonomy and consideration of the interests of research participants and relatives. (Emphases added [2015, 577])

Guidelines emphasized that “supporting the rights of individuals is compatible with supporting families” (Wolf et al. 2015, 445). Despite the intent, however, statements such as these inadvertently reiterate the status quo by reasserting that while individuals have “rights” to the information, families have “interests” that “should be supported.” Families do not have rights to others’ information except in specifically indicated conditions; they are neither legally nor ethically recognized, essentially making sharing optional and access conditional.

Despite clinicians’ and researchers’ commitment to expand sharing with family members, and despite the potential benefit of knowing results that so many professionals endorsed, support for the default of “no sharing without consent” continued to receive strong support. For example, while studies repeatedly document that probands rarely, explicitly refuse to share results with relatives (Mackley et al. 2017), professionals are advised not to disclose information to relatives if probands’ post-mortem wishes are unknown, which is often the case (Branum and Wolf 2015, 589).

Similarly, most guidelines continue to support participants’ freedom to prohibit sharing posthumous research results with relatives “out of respect for their autonomy and privacy” (Branum and Wolf 2015; Wolf et al. 2015, 453). In fact, in a survey, 88% of US IRB chairs and vice-chairs strongly opposed overriding the deceased preferences which they considered to be a breach of confidentiality (Beskow and O’Rourke 2015, 504–505). Of note here is that the abstract principles of individual privacy and autonomy are honored over the potential practical real-life benefit for relatives.

One familiar and widely-supported solution to respecting probands’ wishes is to solicit their post-mortem preferences for communication of their results when they enter a research project (Goodman et al. 2017; Radecki Breitkopf et al. 2018). But
not everyone agrees with this proposal to solicit participants’ preferences. Presenting a minority voice, one prominent research team spoke against the idea, believing that “…a research participant does not have the ethical authority to preemptively prevent relatives from obtaining information that could be of clinical benefit to them after his or her death…” (Chan et al. 2012, 10). Uniquely, this latter argument does challenge the status quo by proposing new limits to individuals’ autonomy to withhold posthumous results from relatives.

Finally, a widely supported pathway for individual professionals to circumvent institutional legal and ethical restrictions of communication is to establish and communicate their own policies about sharing results with relatives to prospective participants as their condition for entry into their research program (Gert, Berger, and Cahill 1996; Branum and Wolf 2015).

What is noteworthy in many of these and other examples is that even when family needs are seriously considered, the primacy of the individual, and all that goes into making it, remains essentially unchallenged (Koenig 2001). The shared nature of genetic information is recognized but only after the information is privatized for the proband. Despite stating that individual and family interests are compatible, which they often are, in discourse they are regularly framed as opposing disparate interests that must be “reconciled.” Such a dichotomous opposition of individuals to families precludes and obscures understanding them as one mutually, constituting, sustaining and defining entity. Designating and protecting information as “confidential” within the family projects and enacts a social and conceptual buffer between each person, contributing to the social construction of distinct individual entities with distinct boundaries, territories and self-serving interests. Widdows captures these divisions in genetics with her notion of a “disconnected self” (2013, 1).

Opposite this distinct, abstract and sovereign individual that still appears in many current bioethics guidelines, policies, and laws is an abstract, undifferentiated secondary entity – the family – a family that usually refers to living relatives only. Kin exist primarily in reference to the individual, with little recognized subjectivity, rights of their own, much less recognition of their own local “informal ethics” for handling communication within a family (Konrad 2005; Gordon et al. 2019).

Part 2. Moving beyond an individualist bioethics paradigm

For many decades a minority policy view has disputed the primacy of individual privacy and control. For example, challenging the individualist approach to genetic information and based on international studies of ideas and attitudes around genetics, Wertz, Fletcher, and Berg argued that,

DNA should not be considered the “private property” of one individual. It should be possible to inform others who share part of an individual’s DNA, namely biological
relatives, about their own health risks and also to allow them access to the DNA which is shared property. (2003, 85)

More broadly, that western individualism has been granted a disproportionate amount of space in bioethics, especially in the US, has been critiqued for decades (Fox and Swazey 1984, 2010; Kleinman 1997; Wertz 2001; Wertz, Fletcher, and Berg 2003; Koenig 2001, 2014; Gilbar 2007; Corrigan 2003; Dove et al. 2017; Widdows 2009, 2013). In fact, this individualist view of bioethics has been accompanied by persistent theoretical and practical challenges from multiple sources: phenomenology, anthropology, sociology, feminism, virtue ethics, communitarianism, and public health ethics. And albeit with various emphases, most critiques challenge the unquestioned primacy of the atomistic individual and the persistent eclipse of the inherent and fundamental social and contextual nature of being human.

In contrast, many proposed alternatives to the individualist paradigm share an understanding of being human as being in relation, in which the self is inherently relational and interconnected rather than separate from others. A relational approach to autonomy, interdependence, and moral responsibility has a relatively long history in genetics (Hallowell 1999; Samuel et al. 2017; Dove et al. 2017). But while it has gained some traction in bioethics (Burgess and d’Agincourt-Canning 2001; Donchin 2001; Jennings 2016; Dove et al. 2019; Hoeyer 2005), it remains relatively marginal to the hegemonic individualism that dominates practice and policy (Dove et al. 2017).

Similarly, many critiques shine light on an ethics of care and moral obligation (Wertz and Fletcher 1991; Wertz 2001; Corrigan 2003; Weaver 2016), an approach often eclipsed by the dominant ethics of autonomy and human rights.

Empirical research findings have long challenged the assumptions of individualism, documenting, for example, how many peoples’ decisions to enroll in genetic testing studies are motivated out of social responsibility and the wish to generate information for the family, not just the individual (Hallowell 1999; Facio et al. 2011; Hallowell et al. 2003). Studies reveal that many feel relatives do have a right to genetic information (Ballard et al. 2020; Dheensa et al. 2016) and want to share that information with family members (Graves et al. 2014; Allen et al. 2014; Amendola et al. 2015; Radecki Breitkopf et al. 2015; Daly 2015).

This persistant and enduring dominance of individualism in bioethics makes the appearance of a more family approach especially significant, not only for its practical implications but also for the changes in foundational assumptions it presents. This is well illustrated in each of the two examples that follow: accounts of a sample of US research participants (2a) and emerging clinical and legal guidelines for genomic medicine in the UK (2b).
2a. US research participants’ family-based ethics of disclosure of genetic information within the family

Let’s begin with a little information about the project interview and survey studies. All study participants (hereafter referred to as “participants”) were recruited from two pancreatic cancer resources based at the Mayo Clinic. Fifty-one people – 17 probands and 34 relatives of either living or deceased pancreatic cancer patients – participated in 50–120 minute semi-structured telephone interviews (Gordon et al. 2019). One thousand, nine hundred and three people responded to the survey – 464 cancer patients, 1040 blood relatives, and 399 spouses/partners (Radecki Breitkopf et al. 2015, 468) – based on Likert-type rating scales and forced-choice questions. Both studies explored preferences for family communication about genetic research information, including mutations related to breast and ovarian cancer, cystic fibrosis, and pancreatic cancer, and some of the reasonings and assumptions behind them. Interviewees were evenly divided between males and females, survey respondents were predominantly female (62.8% vs. 38.2%); the medium ages were 66 and 64, respectively. The vast majorities were highly educated, of Euro-American heritage, economically comfortable, insured for healthcare and, among the interviewees, self-identified as Christian (Gordon et al. 2019) All were English speaking and all but one were “white.”

All participants were directly or indirectly involved in genetic research on pancreatic cancer. All had had a close experience with someone with pancreatic cancer, be it their own or that of a family member; with few exceptions, these were painful and difficult experiences that often engaged the whole family. Pancreatic cancer is difficult to detect early, difficult to treat, and rapidly fatal, with a low six-month survival rate. In fact, several participants recounted their own experiences of having had symptoms that lasted months, and sometimes a year, before a correct diagnosis was made.

Participants’ belief in the determinative and predictive power of genetic information was consistently stronger and less discriminating than among the US professionals (Gordon et al. 2019); some even equated information with medicine: “Information is to your disease what insulin is for diabetes;” a few called information “life-saving!” Knowing one’s risk could allow for early diagnosis of a disease, but even if knowing could not prevent death, the information would be useful for planning the future: “How are you going to plan your life if you don’t know [your genetic information]?” (Gordon et al. 2019, 9). Most wanted minimal limits to participant and family options to know all types of genetic information (Radecki Breitkopf et al. 2015; Gordon et al. 2019).

However, whereas bioethics procedures and policies generally treat genetic information the same as other personal health information, our participants repeatedly emphasized that their opinions about communication were specific to communication of genetic information:
This is not legal information this is genetic information… Genetic information needs to [go] to next of blood kin… Genetic information is not just one person’s information. (Gordon et al. 2019, 11)

In fact, nearly two-thirds (62.6%) of the survey sample agreed that “genetic information belongs to all blood relatives, not just the person who gave the blood sample” (Radecki Breitkopf et al. 2015, 472). Genetic information belongs to the family members who share the genes, who are or could be affected by them. Information should follow the genes, as if part of a family inheritance, not be controlled only by the person who gave the sample: “After death, a family, … if they inherited the gene, they ought to inherit the data” or, “the next-of-kin genetic tree … should be notified. I think it is their genetic right to know.” Those who share the genes, “immediate family members … have a right to know … The genetic code isn’t just hers [the proband], it is also the daughters.” When interviewees were asked, “Does anybody own genetic findings?,” a common answer was: “To me, it [the genetic information] should be available to whoever it affects” (Gordon et al. 2019, 12).

Nearly all agree that given that genetic information is shared and that knowledge is so valuable, relatives, particularly parents, are morally obligated to learn the information; it is part of taking care of yourself and your family, of being a good parent – “I would certainly want to be able to share that information [of a BRCA2 mutation] with both of my kids …. I think as a parent I owe that to them. …” In this way, many described an ethics of sharing and familial interdependence: “Because if I just say, ‘No, don’t tell me,’ then I’m also saying, ‘No, my kids don’t get to know.'” (Gordon et al. 2019, 12)

In this framework, individual privacy takes on a different look and different value. In contrast to US traditional bioethics described earlier, concern for privacy within the family was essentially a non-issue among respondents of both studies, and familial communication of genetic information did not pose an ethical dilemma (Gordon et al. 2019). Only 7.6% indicated they would be “quite” or “extremely concerned” if “your biological family members learned your [medically useful] genetic research results?”; only 7.5% agreed that “I would not want my blood relatives to know about my genetic research results” and only 4.4% agreed that “I would want my genetic results to be kept PRIVATE, even after my death” (Radecki Breitkopf et al. 2015, 473, emphasis in the survey instrument).

Respondents even questioned the rarely disputed right to keep information private: only a third (32.7%) agreed with a survey scenario that a hypothetical Pat “should be able to keep the information [about the BRCA2 mutation] private from others in the family” (Radecki Breitkopf et al. 2015, 470). Only 8.9% thought their parents’ results should be kept private, including after death (Radecki Breitkopf et al. 2015, 470). For many, keeping one’s own genetic
results private was valued as wrong – even deemed selfish – because it meant withholding information from others.

The potential benefit of knowing genetic information was not only for living relatives but for “generations to come” and justified a refusal to honor an individual’s pre-death wishes to withhold results from relatives.

If it is a disease that does not have a genetic influence, then that is different. But if it is something that other generations are going to have to be concerned about …, then I think that it’s not so important to keep it so private. (Gordon et al. 2019, 12)

Survey respondents echoed these interviewees’ views. Where Pat was deceased and Pat’s post-mortem wishes about sharing were unknown, greater than 80% of respondents agreed that the information that Pat carried the BRCA2 mutation should be offered to Pat’s blood relatives, while if Pat had requested that they not be shared, only one third agreed those wishes should be honored (Radecki Breitkopf et al. 2015, 470).

Respondents prioritized collective family health over individual autonomy and privacy. When asked to choose “the most important factor to consider in returning genetic research results,” nearly two-thirds (65.8%) of survey respondents indicated it was “whether blood relatives will benefit,” while only a third (34.6%) chose “the wishes of the person who provided the sample” (Radecki Breitkopf et al. 2015, 472).

Finally, rather than describe the individual and the family as two separate and potentially opposing genetic entities, many participants implicitly depicted the family as one genetic unit, often extending into the past and the future, albeit composed of individuals with shared genetic inheritance and diverse but co-existing desires to know genetic information. Specific families have their own pre-existing or emerging “family ethics of disclosure” (Konrad 2005; Gordon et al. 2019), such as knowing which family members would or would not want to know the information or could be trusted to learn and protect it. The families they described were heterogeneous but not atomistic; while each person is different and unique, they accepted and accommodated those differences.

Interviewer: How important is it to honor the wishes of people who do not want to hear any news about their genes?
Participant: Well, I don’t know. I’m thinking I have five kids. So two don’t want to know and three do. It would be different for each person. (Gordon et al. 2019, 15)

These differences in family members’ interests in knowing genetic information or not were described as known facts of family life. To our interviewees, families are made up of specific people living specific circumstances, well captured by their frequent answers to questions, “it depends” and “everyone is different” (Gordon et al. 2019).
2b. **UK genomic medicine’s proposed family-based ethics of disclosure of genetic information within the family**

In a chorus that echoed our US research participants’ assertion that “genetic information belongs to all blood relatives, not just the person who gave the sample,” many patients in a UK study asked about their own clinical genetic results: “Is this knowledge mine and nobody else’s? I don’t feel that.” They too perceived genetic information, like a mutation, as familial (Dheensa, Fenwick, and Lucassen 2016).

More significantly, however, this US lay familial approach to genetic research information closely resembles the “joint account” model of genetic confidentiality proposed in 2004 in the UK by medical ethicist Michael Parker and Anneke Lucassen (2004), professor of clinical genetics, clinical ethics and law. In contrast to the prevailing individual “personal” model described earlier, this model considers genetic information to be familial, to “belong” to family members and thus be potentially available to other at-risk relatives. It protects confidentiality at the familial rather than the individual level. The default in this model is that information “should be available to all ‘account holders’ (family members), unless there are good reasons to do otherwise,” when exceptions should be made (Parker and Lucassen 2004, 166). It should be emphasized that the main difference between the professional US and UK positions is in their defaults – both recognize the need for exceptions.

This familial approach reflected a deliberate rethinking, informed by parallel qualitative and quantitative research, of how the traditional practices of consent and confidentiality should operate in a genomics era and in the development of clinical genomic medicine (Gilbar 2007; Lucassen and Hall 2012; Dheensa, Fenwick, and Lucassen 2016, 2017; Samuel et al. 2017; Lucassen and Gilbar 2018; Parker and Lucassen 2018; Horton and Lucassen 2019; Dove et al. 2019; Ballard et al. 2020).

This joint account or “familial” model was endorsed and included in 2011 and again in 2019 in the official, non-binding clinical guidelines of the joint committees of the Royal College of Physicians, Royal College of Pathologists, British Society for Human Genetics (2011, 2019), as well as in the 2017 Annual report of the Chief Medical Officer in the UK (Lucassen, Montgomery, and Parker 2017). Yet despite these new guidelines, as of 2017, few clinicians had actually put them into practice; sharing an individual’s information with relatives felt like a breach of the deeply-ingrained tradition of doctor/patient confidentiality (Dheensa et al. 2016; Montgomery 2019; Lucassen, Montgomery, and Parker 2017).

In order to weaken this resistance to putting these new ideas into practice, Parker, Lucassen and others proposed a solution in the 2017 UK’s General Medical Council’s guidelines: rather than approach all genetic information as personal and confidential, thus preserving the status quo, an effort could be made to distinguish between an individual’s personal and confidential information, such as
a clinical diagnosis of breast cancer, and “familial information,” such as familial genetic factors explaining the breast cancer, that should not be confidential at the individual level (Montgomery 2019). They explained:

Close relatives have a greater degree of genetic code in common than unrelated people, and a genetic predisposition to disease may be common to several members of a family. Can a familial predisposition therefore be considered “personal and sensitive” when it is not identifying of any one person? (Parker and Lucassen 2018, 955)

In other words, they recommended changing the boundaries of confidentiality in genomics to be seen, at least in some situations, at a familial rather than an individual level. The authors acknowledge that this may not always be possible, but venture that “in many cases relatives could be alerted via communication between respective health care providers without a breach of confidence of a patient’s clinical information” (Parker and Lucassen 2018). As of 2019, this separation of approaches to confidentiality of individual clinical information from familial genetic information had not yet gained widespread traction in practice (Montgomery 2019).

Like the family-oriented approach endorsed by our US research participants, and in contrast to the US professional research approach, this UK clinical approach maintains that relatives also have rights to genetic information that is relevant to them. This in fact was supported in a ground-breaking ruling the British Appeal Court held in the “ABC v St George Healthcare NHS Trust” 2017 case (hereafter referred to as “ABC”). In this ABC case, a daughter claimed that her father’s clinicians were negligent for not telling her that her father had Huntington’s disease (HD) despite her repeated requests, choosing instead to respect the father’s insistence that they not respond to his pregnant daughter’s requests. After the birth of her baby, the daughter inadvertently learned of her father’s HD diagnosis based on a genetic test. New genetic tests revealed that both she (the mother) and her infant tested positive for HD (“ABC v St George’s Healthcare NHS Trust” 2017).

In a reversal of a previous ruling, the British Appeal Court ruled that the principle of personal autonomy can justify a claim brought not only by the patient, but also by the relatives, emphasizing “that relatives’ interests in autonomy and disclosure are no less important than a patient’s rights to autonomy and confidentiality” (“ABC v St George Healthcare NHS Trust” 2017). They called for limits to the absolute rule of individual confidentiality and privacy – “The assumption that confidentiality towards individuals is always paramount is as inappropriate as the assumption that disclosure is always permissible” (Montgomery 2019) – and took a strong stand against what they called, “the unbridled rule of autonomy;…” they regarded the withholding of important health interventions that could benefit others without any reason as wrong (Parker and Lucassen 2018, 957). They affirmed that “respect for privacy should not prevent advising family members” (“ABC v St George Healthcare NHS Trust” 2017).
Taking as its starting point the dual nature of genetic relations implicates a different understanding of the self and personhood: “Genetics is profoundly relational since one’s genetic inheritance may bind the self to others” (Konrad 2005, 13). In this way, genetics supports a relational understanding of the person, well captured in Widdows’ notion that “the genetic self is a connected self” (2013, 1).

There are signs that the relational approach to autonomy and privacy put forth by theorists and embodied in the UK professional genetic guidelines noted above may even be making its way into English law. In fact for some, the significance of the ABC case went beyond recognizing a relative’s right to know. Roy Gilbar and Charles Foster – both genetic lawyers who had fought long and hard for recognition of the family in genetic law – titled their celebratory article: “It’s arrived! Relational autonomy comes to court!” They explained the significance of the ruling through contrasting paradigms of personhood:

Patients have conventionally been seen as atomistic entities, distinct from those around them. Hence, for instance, the individual patient controls the flow of her medical information to others — including her close relatives. Patient confidentiality is the rule: relatives’ interests in making informed decisions are recognized only in exceptional circumstances, when it is felt that serious harm can be avoided. (Gilbar and Foster 2018, 131)

In contrast, they argue, the judges adopted and imported a relational approach to autonomy (Mackenzie and Stoljar 2000) into law:

The supporters of this approach perceive the individual primarily as a social being, influencing, and being influenced by, one’s significant others (Donchin 2001) … . The Court of Appeal in ABC plainly endorsed this relational construction of the principle of autonomy, because it acknowledged that any decision made by one individual has implications for her significant others. The court recognized that the personal decision of the patient to undergo genetic testing had implications for the family as a unit and for the interests of individual relatives in making informed decisions. They implied that dealing with inherited diseases is not a task the patient faces alone: it is a familial challenge. They declared that if a patient refuses disclosure, clinicians as the agents of society might have a responsibility to help relatives put at risk by that refusal. (Gilbar and Foster 2018, 132)

The authors welcomed “the arrival of relational autonomy into the English courts.” And while the court suggested this widening of clinicians’ duty of care should be restricted to genetic medicine, the authors saw its greater significance:

ABC entails a redefinition of the ambit not only of the duty of care, but of the nature and ambit of humans. Humans are quintessentially relational entities. They cannot rationally be quintessentially relational entities for the purpose of genetic counselling but not for the purpose of obstetrics or orthopaedics. (Gilbar and Foster 2018, 132)
Discussion

We began this article asserting that legal and ethical mechanisms of governance around disclosure embody, produce and reproduce particular understandings and ideals of the self and personhood, of individuals, families, societies and the relationships among them, and that these foundational assumptions show up in how genetic information and its communication within the family are conceived, governed and practiced.

It is difficult to describe any better than these genetic lawyers the foundational cultural assumptions and understandings embedded in prevailing ethical and legal policies and practices, and in turn, what’s involved in changing them. Among many other entanglements preserving the status quo, such as economic, political, psychological, and historical, the difficulty keeping ethics in sync with the ever-changing realities new technology presents derives from the need to also change deeply held, embodied beliefs, conceptions, and habitus about questions as basic as what it means to be a person, an individual, a relative, a patient, and most fundamentally, what it means to be human.

Even in developing new guidelines aimed at increasing family access to individual genomic results in the US, the assumption that genetic information derived from an individual’s research sample is private and under his or her control remains unquestioned; the ethical pillars of autonomy and privacy retain their dominant power, albeit with a few minority inroads, while the priority of individual rights continues to eclipse those of kin and of sharing and caring. Finally, and perhaps most entrenched, the view of the individual as separate, self-governing and distinct from the family – Widdow’s disconnected self – remains preserved in discourse, policy and law, while the family and the familial, shared dimension of genetic information, albeit recognized, remain excluded from the official dominant ethical models (Widdows 2009).

Each of these traditional individualist foundational assumptions and values are challenged and replaced in the family-based ethics proposed by the US genetic research participants and the proponents of the new clinical genetics guidelines in the UK. The family or joint account model of genomic information puts the family squarely inside the legal and ethical framework and proposes conceptualizing and distinguishing confidentiality at the familial, not just the individual level. Family rights to know are recognized, the absolute rule of individual autonomy challenged. Taking the dual nature of genetic relations as a starting point supports a relational and inherently connected understanding of the person. Yet even here, despite these new family-based proposals for genomic medicine, actual enactment awaits accompanying changes in these deeply entrenched and entangled reaches of western individualism.

Conclusions

This essay raises several important questions and answers.
1. Whose ethics, what ethics? How much do we really know about “lay” peoples’ ethics, including their diversity, around sharing genetic information within the family, especially the family ethics of those with something at stake? Are they even recognized? How consonant are they with official legal and ethical regulations, policies and priorities? This familial approach to genetic information we encountered among a Euro-American, highly educated and economically comfortable population – how common, how rare is it? Is it new or just newly revealed?

There is good reason to expect that this familial approach, found in our own and others’ research and proposed in the UK guidelines, is not nor will be idiosyncratic. The extreme uptake of sequencing will “produce” more and more relatives, which will challenge the feasibility and the ethicality of the US framework that excludes the family, leaving them socially and culturally unrecognized and legally and ethically unprotected (Widdows 2009). And what about the many people, majorities, minorities, immigrants, whose social conditions, family structures and cultural practices fit poorly into the norms, priorities and practices governing familial sharing of genetic information (Garrison 2015)?

2. What ethics for what genetic information? We may assume that the specific nature of the three incidental findings associated with pancreatic cancer – well known, relatively preventable and treatable, and associated with a lethal disease – was reflected in the responses of the participants in the genetic pancreatic cancer research studies. Professional and lay ethics of disclosure reflect the particular meanings of the genetic information in question, and as diseases and conditions have diverse and mutating meanings for different people in different positions, we can expect that official and informal ideas and practices around privacy and consent will or will not vary accordingly and evenly. For example, will family privacy be more of an issue when dealing with genetics of stigmatized behaviors and diseases? How will genetic profiles of the increasing multi-variant risk factors be handled in the family context? The inevitable progression of technological development in genomics will continue to challenge the continued appropriateness of prevailing ethics and policy.

3. Protecting, performing and producing individuals. The vast disparity in the importance attributed to privacy and autonomy in traditional and prevailing bioethics versus our patient/relative participants leads us to ask: whose concerns and priorities about privacy do these bioethics actually represent? Clearly the practices of consent and confidentiality provide essential pragmatic protection and control to individuals in contexts where it is needed and wanted. But the surplus significance and power attributed to them suggest that these non-negotiable practices don’t just affirm and protect a pre-existing individualism. Rather, they contribute to producing and reproducing the very thing they claim to protect and respect: individuals, specifically, individuals clearly distinct from their families. Like biomedicine in general, these bioethics practices, policies, laws and concepts around individual privacy and autonomy are important means through which individualism is enacted, performed, maintained and prioritized over the family and,
by implication, the group. The impact of this support of individualism extends well beyond the medical arena (Gordon 1988), another example of how, in the words of sociologist Renée Fox, bioethics is “more than bioethics” (1996, 5).

4. Human genetics needs the right ethics for the right job. Adjustments, modifications and additions to the prevailing individualist framework will not do. To consider alternative policy choices, we must give up an unquestioning reliance on the primacy of the individual and individual choice and consider the primacy of the family (Koenig 2001).

In this way, we regard the efforts of the UK group and others to promote new shared understandings of genetic information, autonomy, and confidentiality as necessary in the genomics era, as significant challenges to the inviolable, almost sacred principles and practices of individualist bioethics. They do so by first distinguishing between ethics appropriate for individual, personal health information vs. those appropriate for shared, genetic information. And they do so by importing into official genomic medical guidelines and law some concepts long fought for inside and outside of bioethics, such as “relational autonomy,” “relational self,” “familial genetic information,” “familial confidentiality,” “familial rights to autonomy,” and “familial and group consent.” Whether or not these new assemblages are allowed to establish deeper roots in this genomics territory is yet to be seen (Gilbar and Foster 2018, 133).

The prevailing professional bioethical architecture is a private dwelling for individuals, benevolently protected by a series of laws, policies, principles and practices. While the interests and welfare of the family are increasingly recognized as legitimate and pressing, the family remains outside the structure. A new blueprint is needed, based on a new foundation and transformation of individual private rooms – into which the family currently has conditional access – into family rooms that allow individuals space of their own. This would constitute a fundamental step towards bringing the family into the room where bioethics happens.

In this article we addressed a current dilemma in bioethics triggered by questions about family access to the abundant individual genomic data produced by genomic sequencing. Mirroring its absence in the materials we reviewed, we began in the middle of the story, considering the ethics of the diffusion of genetic data that had already been produced. This of course evades the most fundamental questions: should all this data be produced? How beneficial is knowing them and who reaps these benefits? And what are the social, cultural, psychological, political, and economic costs of the investment, production, distribution, and clinical interventions that flow from all of this data and who pays them? Identifying genetic predispositions and people at risk nurtures the dream of preemptive medicine and a cure for fear and uncertainty (Aronowitz 2015). But we know too well, and have been wickedly reminded by the COVID-19 pandemic today, that nothing predisposes people to illness and death more than social conditions, such as poverty, racism, hunger, homelessness, marginalization, and that extensive societal investments in genomic medicine can inadvertently usurp essential monies and resources needed to provide the basic needs and services for
individuals and families already at social risk. Needed are bioethics grounded in real life, not in abstract principles; needed are bioethics that actively address the fundamental social nature of human being and human suffering, that support connection and recognize and protect families, communities and individuals. Perhaps, as Monica Konrad notes in the epigraph that opens this essay, human genetics can function as “a social critique of western individualism” by insisting on the indivisible human connection at the core of it all and its centrality to human survival.

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Notes

1. As bioethics in many countries has been strongly influenced and shaped by the US (Fox and Swazey 2010), and as individualism is particularly strong in the US, we focus on it, acknowledging that many of the same values and practices are shared in other countries, particularly in western Europe and other English-speaking countries.
2. The Mayo Clinic obtained approval from the Institutional Review Board (IRB).
3. The prevailing and dominant US bioethics policies are reflected in: federal and state privacy laws, the Common Rule (“The Federal Policy for the Protection of Human Subjects”); the federal Health Insurance Portability and Accountability Act (HIPAA) Privacy Rule of 2003. They are also evident in professional society guidelines, such as the American Society of Human Genetics (ASHG 1998); and in Presidential Commissions, such as The Presidential Commission for the Study of Bioethical Issues: “Anticipate and Communicate: Ethical Management of Incidental and Secondary Findings in the Clinical, Research, and Direct-to-Consumer Contexts” (2013).
4. Because the interview and survey responses, on the one hand, and patients’ and relatives’ responses, on the other, were so concordant, we combined each of these pairs here (Radecki Breitkopf et al. 2015, 2018; Gordon et al. 2019).
5. The main actors developing and proposing this approach include:
   (a) Anneke Lucassen, MD, Dphil (Oxon) FRCP, Clinical Ethics and Law, Faculty of Medicine, Professor Clinical Genetics at University of Southampton, UK; Chair of the British Society
of Genetic Medicine; together with her clinical and research colleagues, Ballard, Crawford, Dheensa, Fenwick, Hall, Horton, Samuel, Shkedi-Rafid and others (see References); (b) Michael Parker, MA, PhD, Lecturer in medical ethics at Ethox Center, Nuffield Department of Population Health, University of Oxford, UK; (c) Roy Gilbar, School of Law, Netanya Academic College, Israel; and School of Law, University of Leicester, UK; and (d) Sir Jonathan Montgomery, Professor of healthcare law, University College London, UK.

6. They are not the only ones who promote this distinction but are unusual in having integrated it more formally and systematically into official clinical guidelines.

7. The original empirical study on which part of this essay is based had limitations, described in detail elsewhere (see Gordon et al. 2019 and Radecki Breitkopf et al. 2015, 2018). Like too many studies, our interviews and surveys tapped a homogeneous white, well educated, relatively economically and medically advantaged population and suffers from all the limitations that brings and the lacune it maintains.

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