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The Concept of "Genetic Responsibility" and Its Meanings: A Systematic Review of Qualitative Medical Sociology Literature

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The acquisition of genetic information (GI) confronts both the affected individuals and healthcare providers with difficult, ambivalent decisions. Genetic responsibility (GR) has become a key concept in both ethical and socioempirical literature addressing how and by whom decision-making with respect to the morality of GI is approached. However, despite its prominence, the precise meaning of the concept of GR remains vague. Therefore, we conducted a systematic literature review on the usage of the concept of GR in qualitative, socioempirical studies, to identify the main interpretations and to provide conceptual clarification. The review identified 75 studies with primarily an Anglo-American setting. The studies focused on several agents: the individual, the family, the parent, the healthcare professional, and the institution and refer to the concept of GR on the basis of either a rational/principle-oriented approach or an affective/relational approach. A subtype of the rational/principle-oriented approach is the reactive approach. The review shows how the concept of GR is useful for analyzing and theorizing about socioempirical findings within qualitative socioempirical studies and also reveals conceptual deficits in terms of insufficient theoretical accuracy and heterogeneity, and in the rarity of reflection on cultural variance. The vagueness and multiplicity of meanings for GR in socioempirical studies can be avoided by more normative-theoretical explication of the underlying premises. This would provide a higher degree of differentiation of empirical findings. Thereby, the complex findings associated with the individual and social implications of genetic testing in empirical studies can be better addressed from a theoretical point of view and can subsequently have a stronger impact on normative and policy debates.

Keywords: genetic responsibility, systematic review, qualitative studies, genetic counseling, decision making, lay persons, professionals, bioethics

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INTRODUCTION

In the past 30 years, the technical possibilities for producing risk information in genetics have increased substantially and quantitatively. From visual chromosome analysis of aberrations (e.g., for Down syndrome) to more advanced testing for rare genetic disorders based on polymerase chain reaction, next-generation sequencing, genome-wide association studies, and "personalized" medical

treatments, the field of predictive medical genetics has grown dramatically. However, skepticism about the consequences of using genetic information (GI)[1] and the validity of most of the current tests remains. This has led to a series of unsolved questions: What does this information mean? Do we want to have this type of information? If so, how can we manage and act upon the genetic risk? If not, is it responsible to continue testing? Does this information impose personal and familial responsibilities to minimize risk for genetic disease development?

These questions counter the optimistic opinions of some moral philosophers (Harris, 1998; Buchanan et al., 2007) who have suggested that genetic risk information allows us to move away from our dependency on nature and take control of our fates. They argue, from the perspective of the promotion of individual autonomy and choice, that neglecting genetic risk information and not using it for one's own and the common good is morally dubious.

Apart from the rhetoric of choice, another moral concept has shaped the debate on genetic testing (GT) since the 1970s: that of "responsibility" or, more concretely, "genetic responsibility" (GR). The latter term was coined by Lipkin and Rowley (1974) to argue in favor of reproductive and positive eugenic considerations from a collective responsibility point of view. They stated that mankind should use the opportunities of emerging genetic science to act "responsibly" toward the next generations by avoiding the inheritance of diseases (Fletcher, 1974). This revival of eugenic virtues was embedded in the emerging awareness of limited natural resources, a growing world population and an ecological crisis, as prominently discussed in the "Club of Rome" report (Meadows, 1972). Another approach to the concept was initiated by the sociologist Nikolas Rose and followers, who theorized that GR reflects the biopolitical impact of a more and more genetic "denkstil" (thought style) on individual life styles and thus on conceptualizations of the self and its relation toward the sociopolitical realm (Lemke, 2006; Rose, 2007). Summing these notions up, the term appears to be associated with ambivalence, individual and social conflict, and a critical attempt at improving genetic practice.

The widespread usage of the term GR and the related notion of "responsibilization" in health care is often related to the sociopolitical paradigm of invoking individual responsibility and the citizen's need to take actions for health risks instead of promoting collective or state solutions to handle those risks. From a theoretical point of view, this is a very intriguing phenomenon because, in bioethics, "responsibility" has so many conceptual and historical meanings [for an overview, cf. Schicktanz and Schweda (2012)]. Furthermore, philosophers, bioethicists, social scientists, and medical professionals often use it in different ways without providing sufficiently clear definitions. Starting from these observations, in this review, we have examined the following research questions: which concepts of GR are applied in qualitative, socioempirical studies and how are these concepts used to explain empirical findings on moral attitudes toward GT and GI?[2]

The objectives of this review were threefold. First, we aimed to summarize and analyze how existing qualitative studies have applied, described, and theorized about the notion of GR; for this, we used a general philosophical notion of responsibility as a theoretical premise to detect references to GR in the material. Second, we aimed to provide a classification of approaches to the concept of GR and to analyze similarities and variances within this classification. We based our classification on a philosophically informed structure that addressed the main agents and contexts of GT and GI. Third, we aimed to discuss important inconsistencies in the literature on GR and, to promote a closer interaction between empirical medical sociology and bioethics, to make suggestions for overcoming this inconsistent terminology.

MATERIALS AND METHODS

Theoretical Background

For the purposes of this systematic review, we have used a general, meta-ethical framework of responsibility that captures the various local and contextual conceptual meanings of "responsibility" (Schicktanz and Schweda, 2012). This offers the advantage of integrating various meanings of "responsibility" into one consistent general concept. Based on the idea that responsibility could be seen as a formal expression of any moral statement by which we account other people as moral beings (Strawson, 2008), this general concept not only accounts for the plurality of approaches to GR in bioethics and medical sociology literature but also avoids a one-sided, reductionist interpretation, where "being responsible" is equivalent to "being blameworthy" or "being guilty" (Schicktanz and Schweda, 2012; Schicktanz, 2016).

Overall, the concept of responsibility can be seen as involving seven *relata* (Schicktanz and Schweda, 2012): someone (*the moral agent*) is, in a particular time frame (*time*), retrospectively or prospectively *(temporal direction)* responsible for something or someone (*the moral object*) with respect to someone (*the norm proofing authority/enforcement*) on the basis of certain standards (*norms*) with certain consequences (*sanctions or rewards*).

We expected that the current literature on GT would show aspects of all seven dimensions of responsibility. At the same time, we did not assume that the colloquial concept of GR in empirical studies would encompass all of these dimensions at once.[3] Thus, this general formula of responsibility was used to

[1] We refer to GT as a medical test that identifies changes in chromosomes, genes, or proteins. GI in a narrow sense refers to information acquired *via* GT, yet the term may also include other information about an individual acquired in the context of GT and genetic counselling that is not a direct result of the technological application (e.g., family trees) (Lister Hill National Center for Biomedical Communications, 2016).

[2] This review is part of a larger project in which yet unclarified conceptual uses of GR in the wider bioethical and biopolitical discourse about GT are being analyzed. A second analysis, in which we aim to analyze the concept of GR in the ethical literature, is currently in preparation.

[3] Although the temporal dimension of responsibility is well acknowledged in the theoretical discussion, the spatial dimension is yet insufficiently addressed. However, an implicit way of addressing the spatial dimension can be seen in the way how the relationship between the moral agent and moral object is construed and addressed. We can think of social closeness (e.g., such as in family) as a

allow us to distinguish a wide variety of concepts of GR, while analyzing the specific concepts present in the literature from one, unified point of view.

This formula enabled us to structure the variety of concepts associated with GR found in the material and to distinguish the usages of GR along such dimensions as "agent," "object," "norm," "consequences," etc., if applicable.

Search Strategies

The literature search was performed in January 2016 in the PubMed and Web of Science databases to include all articles published in English and German[4] from 1945 to the present (for search strings, cf. **Figure 1**). These searches were complemented by a search for gray literature from monographs and anthologies, drawing on resources at the University of Göttingen.

Selection Criteria

We compiled a comprehensive corpus of literature in which both "genetic risk" *and* "responsibility" were broadly discussed with regard to practical possibilities offered by the new genetics. To achieve this, we included all publications in English or German that examined GT or genetic screening and used the term "responsibility" in the title, abstract, or keywords (cf. **Figure 2**). A search limited to the narrow term "genetic responsibility" would have led to a biased picture, because in a pilot search, we identified many relevant studies closely connecting responsibility and genetic risk but not using "genetic responsibility" as a key term.

We also included related articles from the gray literature where "responsibility" was only indirectly addressed *via* reference to duties and obligations to conform to social or moral norms in the context of decision making about GT. Articles referring to "responsibility" or "being responsible" exclusively in the sense of "being the cause for" were excluded, as such expressions did not convey the moral meaning of GR we wanted. As we aimed to track GR in the scientific discourse, we only included original scientific work, scientific review papers, peer comments to these works, and reports of ethics committees. Editorials, conference abstracts, letters to the editor, method reports, and reader surveys were thus excluded.

Thematically, we excluded papers that focused on (a) genetic ancestry testing; (b) GT of non-human animals; (c) the efficient administration of genetic screening programs; (d) storage of genetic material in the laboratory; (e) the biotechnical function of genetic tests; (f) insurance procedures to assess genetic risk; (g) the construction of scientific entities such as the concept of the "gene"; (h) genetic discrimination, in cases where there was no obvious relation with GR; (i) national and international legal regulations on the management of GI; (j) responsibility of healthcare professionals, if there was no explicit reference to the management of GI; (k) responsibility of researchers working on the Human Genome Project and on whole-genome sequencing; and (l) prenatal or pre-implantation testing if GT or GI was not mentioned.

Search Strategies in Web of Science and PubMed Database

Web of Science

Search String: [Topic] genetic testing AND [Topic] genetic responsibility

Refinement:

RESEARCH AREAS: (GENETICS HEREDITY OR SOCIOLOGY OR MEDICAL ETHICS) AND **RESEARCH AREAS:** (GENETICS HEREDITY OR FAMILY STUDIES OR SOCIOLOGY OR MEDICAL ETHICS OR SOCIAL ISSUES OR LEGAL MEDICINE OR PHILOSOPHY) AND **RESEARCH AREAS:** (GENETICS HEREDITY OR FAMILY STUDIES OR SOCIOLOGY OR MEDICAL ETHICS OR SOCIAL ISSUES OR LEGAL MEDICINE OR SOCIAL SCIENCES OTHER TOPICS OR PHILOSOPHY) AND **Databases:** (MEDLINE OR WOS OR SCIELO) AND **DOCUMENT TYPES:** (ARTICLE OR REVIEW OR ABSTRACT OR CASE REPORT) AND **LANGUAGES:** (ENGLISH OR GERMAN)

Timespan: 1945-2015.*

Search language=Auto

Results retrieved: 423

PubMed

Search String: **((((genetic testing[Title/Abstract]) AND genetic responsibility[Title/Abstract])) OR ((genetic screening[Title/Abstract]) AND genetic responsibility [Title/Abstract])) OR ((genetic testing[Title/Abstract]) AND responsibility[Title/Abstract])) OR ((genetic screening[Title/Abstract]) AND responsibility[Title/Abstract])**

Results retrieved: 115

FIGURE 1 | Research strategy for socioempirical studies referring to 'responsibility' in genetic testing. *The default time restriction in Web of Science is 1945.

A total of 228 duplicates were removed from the corpus. The selection criteria were applied by assessing each remaining paper after reading the title, abstract, and keywords. At least two researchers (Jon Leefmann and Manuel Schaper/Silke Schicktanz) assessed the papers independently. If a decision could not be made on this basis, the whole paper was read and a consensus was sought. With regard to our aim of reviewing the conceptual uses of GR in qualitative studies, our assessment of the robustness of the evidence drawn from the studies did not

constructive manner of space as well as of narrow distances that matter (e.g., exchanging GI in a same room of a hospital). The latter is then a geographical manner of space (Simmel, 1971).

[4]The search was performed with no time limitations. However, it was limited by the maximal timeframe offered by the databases (i.e., 1945–2016) and the authors' language skills.

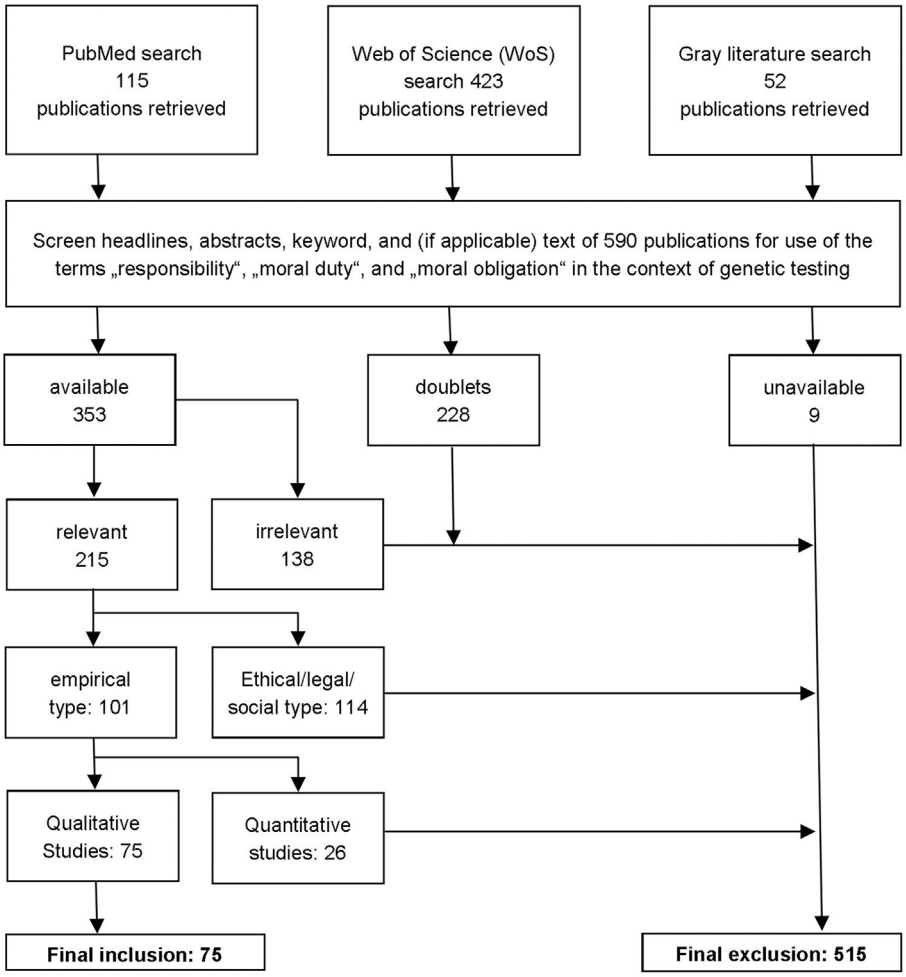

FIGURE 2 | **Final inclusion and exclusion of studies to the corpus**.

consider criteria such as representativeness of the included studies as central. Instead, we excluded qualitative studies only if their methodology was missing or if the paper used qualitative data only anecdotally. Application of these selection criteria yielded a total of 353 publications.

Data Extraction

We then divided the publications into empirical studies and non-empirical papers reflecting on ethical, legal, and social aspects of the new genetics for separate analysis and later comparison. Non-empirical papers were excluded. Every paper reporting data acquisition and/or presenting methodologically sound data analysis (quantitative and qualitative) was considered as an empirical study. This criterion also applied to studies that drew on empirical data for ethical analysis or to theorize about social processes related to genetics ($n = 101$). Of these we excluded review papers, quantitative studies, and case studies ($n = 26$), limiting the reviewed literature to qualitative, socioempirical studies. This restriction is justified because our research questions aimed at the various interpretations of GR generated from empirical material. As quantitative studies tend to prompt the respondent's attitudes using predefined concepts, the conceptual uses are limited, and the process of conceptual construction is usually not documented in the studies. Hence, qualitative studies provided much better background for our interests (Flick, 2009). Thus, we obtained a comparatively homogeneous corpus in terms of study design (cf. Table 1 in Supplementary Material). Finally, 75 qualitative, socioempirical studies that referred to GR in the context of GT were identified (cf. **Figure 2**).

From these 75 studies, all 3 authors independently collected data on the size of the data sample, country of origin, study design (type of data acquisition and method of analysis), research topic, and the following outcome measures: medical genetic context, agents of responsible behavior, objects of responsible behavior, norms of responsibility, potential conflicts of responsibility norms, definition of responsibility, and purpose of conceptual use (cf. Table 1 in Supplementary Material).

Analysis

We have used a descriptive approach to the literature corpus (part 1) and a narrative synthesis approach (part 2). In part 1, we used the criteria commonly used in the PRISMA protocol for systematic reviews, as applicable to the qualitative data (Liberati et al., 2009), providing a detailed protocol for inclusion and exclusion criteria (**Figure 1**), the sizes of each of the thematic categories or subcategories, and a systematic characterization of the literature corpus in terms of sample size, date of publication, national context, and disease model for each of the studies. In part 2, we based our analysis on the narrative synthesis approach developed by Popay et al. (2006) and also described by Ryan (2013). This approach is suitable for reviewing large sets of studies, which vary greatly in methodology and research focus and which cannot be summarized using statistical tools. Our narrative synthesis in principle follows the recommended steps of (i) theorizing about the phenomenon under investigation, (ii) developing a preliminary synthesis of the findings from the literature by thematically categorizing the studies and by summarizing key findings, (iii) exploring relationships within and between the different studies, and (iv) assessing the robustness of the synthesis of the findings [see also (Strech and Sofaer, 2012)]. As this methodological approach is normally used to review the effects of clinical interventions, however, we modified our narrative synthesis according to our purpose: instead of a theory of the changes produced by the clinical intervention (Weiss, 1998; Popay et al., 2006), which is suitable for investigating cause–effect mechanisms, we have referred to the theoretical analysis of the concept of responsibility as provided by the general formula outlined in Section "Theoretical background." Like the theory of intervention leading to changes, as in the original approach, this formula provided the framework for searching for factors in the studies, which could explain similarities and differences in the conceptual use of the term GR. For instance, the most central relata (e.g., agent, object, norm) often function as indicators for the thematic contextualization of a study (e.g., family communication, professional duties, agency, etc.). These similarities were used to develop a preliminary synthesis of all our findings and to thematically categorize the studies accordingly. The results of this preliminary synthesis of the findings are plotted in Table 1 in Supplementary Material. We further analyzed these findings to identify overall patterns concerning references to or conceptual uses of GR. Close reading of the material allowed us to generate hypotheses on conceptual uses, which we repeatedly and iteratively revised in the light of further gathered data from the material. Using this inductive process resulted in two stable categories of conceptual uses of GR: one use referring to a rational/principle-oriented meaning and another use relating to an affective/relational meaning. A particular subtype that occurred within the first category is the "reactive" conception, as we will explain later on. Interpretations based on these categories form the main part of part 2 of the Section "Results." We have also provided an overall, critical assessment of our synthesis of the findings in the section "Discussion."

RESULTS

Part 1: General Description of the Literature Corpus

Quality of Included Studies

The sample sizes and methodology of most of the studies (cf. Section "Methods of Data Acquisition and Analysis" and Table 1 in Supplementary Material) indicated that most were of sufficient quality in terms of basic methodology. For an assessment in terms of the conceptual use of GR, we looked for conclusive definitions of GR in the studies and tracked quotations and references that might expose a theoretical discussion of GR. While we only found 21 studies offering a conclusive definition of GR (Burgess and d'Agincourt-Canning, 2001; d'Agincourt-Canning, 2001, 2006; Downing, 2005; Hallowell et al., 2006; Arribas-Ayllon et al., 2008a,b, 2011; Bickerstaff et al., 2008; Etchegary and Fowler, 2008; Etchegary et al., 2009, 2015; Raz and Schicktanz, 2009; Dancyger et al., 2010; Hines et al., 2010; Clarke et al., 2011; Weiner, 2011; Donnelly et al., 2013; Boardman, 2014b; D'Audiffret Van Haecke and de Montgolfier, 2015; Ross, 2015), all but 17 of the included studies made reference to non-empirical literature, such as philosophical or bioethical papers referring to or using GR (cf. Data Sheet 1). We took both these measures as indications of the authors' awareness of the conceptual discourses on GR in philosophy and bioethics.

Publication Types and Disciplinary Context of the Reviewed Studies

Among the reviewed studies, only three appeared in edited volumes. The remaining 72 studies appeared in 31 distinct journals from different subfields of the social and the medical sciences (all journals are listed in the Data Sheet 1). The most frequent journals in our corpus were *Journal of Genetic Counseling* ($n = 13$), *Social Science and Medicine* ($n = 10$), *Sociology of Health and Illness* ($n = 5$), and *Clinical Biochemistry* ($n = 5$). We used the Scientific Journal Ranking Subject Area Classification (cf. www.scimagojr.com) to assess, which academic audience was mainly addressed by the journals. Among the 31 journals, only 9 can be unambiguously identified as medical journals. The remaining 22 journals were classified as social sciences or the humanities journals with an interdisciplinary audience ranging from social scientists to medical practitioners to applied ethicists. However, it is remarkable that the 9 journals clearly identified as medical journals contributed to 27 studies, which amounts to more than a third of the whole literature corpus. This comparatively large number suggests the existence of a significant readership interested in GR beyond medical sociology and medical ethics.

Frequency of Studies over Time

Even though the use of GR as a theoretical concept dates back at least to the 1974 anthology by Lipkin and Rowley (1974), we found that the term did not play a significant role in qualitative socioempirical studies before the early 1990s. From then on, we found a steady increase of publications referring to GR between 1993 and 2015 in our material (cf. **Figure 3A**). We assume that this increase is likely due to an augmentation of the number of studies addressing

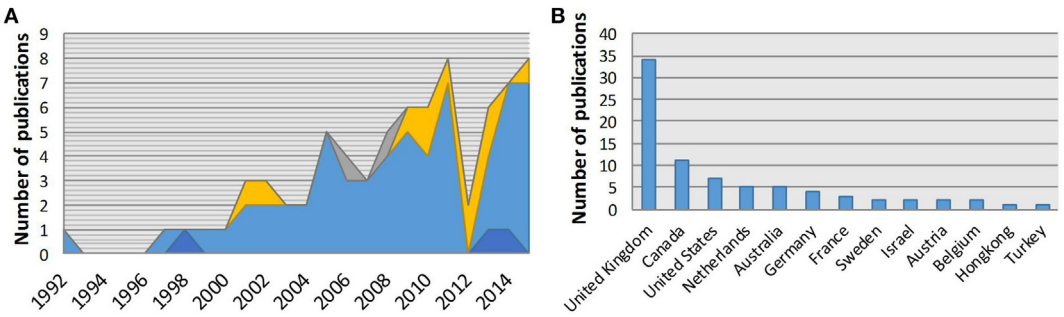

FIGURE 3 | Frequency of studies over time (A) and distribution of the retrieved qualitative socioempirical studies over different countries of origin (B). (A) The different colored planes indicate different thematic contexts of responsibility: GR for agency (dark blue), GR of at-risk people in the context of the family (light blue), professional GR (yellow), and GR of institutions (gray). The size of a plane represents the frequency of studies in a context over time. Planes add up to the total frequency per year. **(B)** Studies, which emerged from collaborations of researchers from different national backgrounds, were counted once for each national background involved.

GT as such. However, we found also that GR was mainly addressed as "responsibility to manage information about genetic risk in the family context" (cf. below). The number of studies focusing on other thematic contexts did not increase over the years.

Methods of Data Acquisition and Analysis

Of the 75 qualitative studies included, 59 used semistructured interviews for data acquisition (cf. Table 1 in Supplementary Material). In five studies, semistructured interviews were combined with focus groups or questionnaires, and eight studies used focus groups for data collection. In one case, (video) recorded counseling sessions were analyzed. Other methods included ethnographic field work ($n = 1$) and narrative interviews ($n = 1$). The number of participants in the studies varied between 8 and 66. Various approaches were chosen for data analysis (cf. Table 1 in Supplementary Material). The most frequently used analytical tools were grounded theory ($n = 11$), thematic analysis ($n = 8$), inductive thematic analysis ($n = 8$), qualitative content analysis ($n = 8$), constant comparative analysis ($n = 7$), rhetoric discourse analysis ($n = 5$), and interpretative phenomenological analysis ($n = 4$). Other methods were applied as well, and 16 studies did not explicitly mention a method used for data analysis.

National Contexts

The material displayed a highly unequal distribution of studies from different countries (cf. **Figure 3B**). The majority of studies were conducted in the United Kingdom ($n = 34$). Reference to GR was also frequent in studies from Canada ($n = 11$), the United States ($n = 7$), the Netherlands ($n = 5$), and Australia ($n = 5$). The strong bias toward Anglo-American medical sociology in the sample might reflect the specificities of the selected databases used in the study but will also reflect our language restriction to English and German. However, it should not be dismissed that a particular thought school or "denkstil" is perhaps implicated.

Genetic Conditions Discussed

A total of 35 different genetic conditions were mentioned in the material, 13 of them as part of broader nosological categories such as muscular dystrophy or cardiomyopathy (for details, cf. Data Sheet 1). These 35 conditions included monogenetic diseases such as Huntington's disease (HD) or cystic fibrosis (CF), as well as multifactorial conditions and disorders such as hypercholesterolemia, obesity, or arhythmogenic right ventricular cardiomyopathy (ARVC) (cf. Table 1 in Supplementary Material). The most frequent genetic conditions discussed were hereditary breast/ovarian cancer (HBOC) ($n = 19$), HD ($n = 11$), and hereditary non-polyposis colorectal cancer (HNPPC) ($n = 6$). Twelve studies did not specify a genetic condition. As these studies focused on testing and screening for hereditary disorders, almost all of the conditions are considered rare diseases by the European Orphanet Consortium. However, in our corpus, we observed a shift over time from rather monogenic diseases toward genetic conditions or diseases with a multifactorial etiology. Hypercholesterolemia, obesity, and sickle cell anemia were not mentioned before 2011. We also found ($n = 5$) studies discussing GR with regard to various genetic mutations that increase the risk of sudden death by cardiomyopathies such as ARVC, long QT syndrome, or hypertrophic cardiac arrest. Risk for cardiomyopathy was not mentioned before 2010 in our material.

Part 2: Narrative Synthesis

In this section, we flashed out a detailed analysis of the concepts of GR occurring in the identified literature corpus and the use of these concepts for interpreting or theorizing about empirical findings. In the first step, we analyzed the moral approach to GR in our literature corpus. It is important to stress that these approaches were obtained from the examined material by an inductive process (see Analysis). In Section "Shared Assumptions on GR in the Literature Corpus," we have distilled a basic overall definition of GR that was shared by most studies, although only implicitly by some. In Section "Moral Approaches to GR: Rational/Principle-Oriented vs. Affective/Relational," we have analyzed the moral quality attached to GR, distilling two types of moral approaches from the literature corpus: *rational/principle oriented* and *affective/relational*. Furthermore, we distinguished a small subset of studies displaying a special subtype of the rational/principle-oriented approach, which we named *reactive*. Finally,

in Section "Categories of GR Studies," we have given a detailed overview of the meanings and uses of GR in the socioempirical literature.

Shared Assumptions on GR in the Literature Corpus

All 75 included studies shared the basic assumption that the knowledge derived from GT opened up a new space for action for the individuals or entities involved, including any affected person involved in the process such as partners, children, siblings, parents, and other family members, and also stakeholders such as healthcare professionals or insurance companies. Depending on the accuracy and reliability of the respective genetic test, these agents were confronted with probabilistic risk information about possible future events, over which they might or might not be able to exert control.

The examined studies assumed that managing and handling this information could be socially or morally problematic on different levels, in different respects, and for different stakeholders. In all studies, the decision to undertake GT and to act on the basis of the test result took place in a moral and social framework of norms and expectations. These norms shaped the terrain of the morally accepted or non-accepted actions. What counted as responsible behavior in the context of GT was framed by socially negotiated norms not only between health professionals and laypeople but also within families.

The guiding moral principles underlying these norms included "respect for autonomy" and "the obligation not to cause harm." Both principles were expected to hold for any person involved in the handling of GI regardless of their role as the affected individual, a relative, or a healthcare professional. However, these principles were frequently ambivalent and conflicting in the context of GT, as their appropriate realization varied with the genetic disorder under investigation and the specific intrafamilial relationships of the affected people (cf. Table 1 in Supplementary Material).

Ethical conflicts often discussed included the question of whether a duty to warn people at risk existed, and whether and when this duty could override the principle of respect for autonomy. Does one have a right not to know about one's genetic risk, if accurate risk information is available and could be used to prevent future harm? In a similar vein, questions about not harming at-risk people arose. Does sharing information increase autonomy or cause harm because it induces risks of social or psychological harm? Hallowell et al. (2003) formulated this conflict in terms of competing responsibilities: "At one and the same time, they [the affected women] saw themselves as having the responsibility of providing their kin with GI that would foster autonomous decision making about their health risk management and as having a responsibility to protect their relatives from the harms that this information might cause (p. 78)."

How the affected individuals or those at risk could or actually did make a responsible decision based on the conflicting principles of respect for autonomy and not harming others was the main concern of all 75 studies. This question of responsible conduct was, however, addressed from various points of view. The underlying social relationship was approached in different ways: some studies, for instance, looked at these ethical conflicts from a gender perspective (Hallowell, 1999; d'Agincourt-Canning, 2001, 2006; Foster et al., 2002; Hallowell et al., 2002, 2006; Forrest et al., 2003; Rowley, 2007), whereas others focused on intercultural differences (Raz and Schicktanz, 2009; Shaw and Hurst, 2009; Raz et al., 2014) or on specific familial relationships, such as that between parents and their underaged children (Steinberg et al., 2007; Arribas-Ayllon et al., 2008b).

Moral Approaches to GR: Rational/Principle-Oriented vs. Affective/Relational

The material displayed two moral approaches to GR. Although they were not mutually exclusive, their focus differed in the way the moral quality of responsibility was interpreted. Some studies understood taking responsibility as the reaction of an autonomous agent in rationally guiding her behavior toward herself and others according to formal, universal moral norms. This involved taking a detached, reflective attitude toward the interests and moral requirements affected by a specific course of action (a *rational/principle-oriented* approach). In other studies, the concept was approached as the reaction of a socially embedded agent guiding her behavior in accordance with the moral feelings and affective responses implied by her social role and relational identity. We labeled this as an *affective/relational* approach to responsibility, because it referred to close relationships built on emotions such as love, care, and trust.[5]

Interestingly, this difference resembles a classical distinction introduced by moral psychologists in the 1960s and 1970s to explain moral development (Kohlberg et al., 1983; Gilligan, 1987; Blum, 1993; Nunner-Winkler and Nikele, 2001). This debate has also influenced many discourse-based and feminist-oriented ethical approaches (Benhabib and Cornell, 1987; Held, 2006). We suggest using this basic distinction (between a *rational/principle-oriented* and an *affective/relational* approach to responsibility) for further categorizing concepts of GR.

The basic rational/principle-oriented approach defined GR as "an integral factor in influencing an individuals' desire whether to know and to act on the knowledge of their genetic risk, obligation of disclosure, reproductive decision-making, and genetic testing decisions" (Ross, 2015, p. 37). In this sense, GR comprised the moral obligations guiding a person's behavior with regard to herself and others. Here, GR as a moral responsibility centered on the moral attitudes of an autonomous agent, who was capable of perceiving and understanding the obligations and of deciding whether to act upon them. A responsible agent in the rational/principle-oriented sense was capable of justifying her actions and could be held accountable for them.

Notably, there was a subtype in the rational/principle-oriented approach to GR in the material, which primarily prevailed in the work of Arribas-Ayllon et al. This concept of GR, which we have described as "reactive," should perhaps be conceived of as a "discoursal articulation [of a macro-political concept] in the research interview setting" (Arribas-Ayllon et al., 2008a, p. 268),

[5]We do not intend to imply that the affective/relational approach to responsibility is less rational than the rational/principle-oriented approach. Emotional reactions toward close and distant others can be rational, for instance, if they reliably track the morally salient features of their relationships with others.

which indirectly mirrors the social and biopolitical norms to which people react. For instance, certain rhetorical strategies such as self-blame or the blaming of others could be interpreted as attempts by the affected people to justify their management of the genetic risks and to appear responsible for themselves and others against the prevalent social pressures.

The understanding of GR as an affective/relational responsibility was in opposition to these two rationalistic concepts and drew on the social identities of the affected or at-risk people, taking place in the close relationships that were shaped by strong emotions such as love and personal trust. Nonetheless, this can be still understood as a moral responsibility, but one which mainly occurs in close relationships such as families. Burgess and d'Agincourt-Canning (2001) defined this kind of responsibility in the context of GT as "a sense of moral self that is largely defined by the responsibilities personally acknowledged as arising from particular relationships. […] this notion is characterized by who an individual wants to be in relation to others. Relational responsibility draws attention to the everyday morality in which people live their lives, support loved ones, and struggle with their own sense of self and future. It acknowledges that a person's actions (and even desires) are shaped by responsibilities to others. […] relational responsibility is bound up with the meanings people construct around their sense of self and moral identity (p. 363)."

This affective/relational understanding is important to the extent that people factually often *feel* obliged to act and thereby to take responsibility in accordance with their social role and identity, but cannot always rationalize or justify these moral commitments. Women are especially often reported to feel a duty to act responsibly by influencing the health behavior of partners or other family members (d'Agincourt-Canning, 2001; Hallowell et al., 2002). In these cases, affective reactions toward children and the wider family are linked with and potentially caused by a conscious or unconscious endorsement of a specific social identity. Such endorsements present themselves, for example, when those affected make distinctions about their obligations to inform at-risk relatives that depend on the kind of relationship they feel they have with them.

Categories of GR Studies

Thematic, narrative analysis of the corpus allowed us to detect four major categories of studies that differed with regard to the type of agent of responsibility. This distinction reflects the finding that conceptualizations of GR generally differed primarily with respect to the agents and objects of responsibility. The agents of responsibility are those who make decisions related to GI and act upon them by targeting an object, i.e., themselves or another person (cf. Part 1: General Description of the Literature Corpus)]. We have classified the literature corpus into

(A) studies focusing on the responsibility for action based on knowledge of one's own genetic condition;
(B) studies addressing the responsibilities of (potentially) affected people in the context of family;
(C) studies focusing on the responsibilities of healthcare professionals in genetic clinics and laboratories; and
(D) studies focusing on the responsibilities of institutions such as health insurance companies and the state.

In the 62 studies classified as (A) or (B), the agents were the affected or at-risk individuals. By affected we mean that they wanted to undergo or had already undergone GT for themselves to learn about their genetic conditions and/or were seeking information about the genetic risks of their (unborn) children. Some studies reflected explicitly how GI has sparked a new form of "being affected"[6], for instance by detecting carriers for recessive genetic disorders (Reed, 2009; Atkin et al., 2015; Hoeltje and Liebsch, 2015).

The studies revealed a broad spectrum of agents as the objects of responsibility, ranging from the individual him/herself to the offspring of the individual, a distant relative and society at large. Studies in which the agent (the acting person) and the object of responsibility were identical ("self-responsibility") ($n = 3$) were grouped as type (A). Studies in which the agent was not identical to the object of responsibility ($n = 59$) were grouped as type (B) (responsibility for others) (cf. **Figure 4**). Studies on the responsibilities of the affected people in a family context formed by far the largest group (B) in our literature corpus. To make our results more accessible, we subdivided this large group into four thematic subcategories: decision making for personal GT (B1), disclosure of genetic risk (B2), decision making and disclosure intertwined (B3), and GT in reproductive decision making (B4). The "objects" of responsible action in all these studies were potentially at-risk children, relatives, and next of kin,[7] whereas the moral norms most often referred to were the obligation to promote the welfare of relatives and next of kin and the obligation to respect their autonomy if they preferred not to know about the potential risk or were unable to handle this information. However, the subgroup B4 ($n = 11$) differed from the other three subgroups in two important respects: first, the agents of GR in studies focusing on the context of reproduction were primarily women or couples with a family history of genetic diseases or with a proven genetic risk of having children with a genetic illness. Second, the studies in subgroup B4 only involved communication of genetic risks between the future parents of a child or between the parents and a genetic counselor and focused exclusively on parental responsibilities toward the embryo.

In the 11 studies comprising group C, the agents were healthcare professionals working in the context of GT: genetic counselors, physicians, nurses or geneticists, and laboratory workers performing genetic tests. In group D, only two cases, institutions such as a state legislative (Bickerstaff et al., 2008) or insurance companies (van Hoyweghen et al., 2006), acted as agents of GR.

In all thematic categories, usage of the rational/principle-oriented or affective/relational concepts of GR was identified (see also **Figure 4**); these are discussed in the following sections.

[6] for a general discussion of the term, cf. Schicktanz et al. (2008).

[7] We have distinguished between "relatives" and "next of kin." The first includes not only people who are genetically related but also those who are affected by the GI who do not share the ancestry relevant for the genetic risk, such as partners or spouses or the family of the spouse.

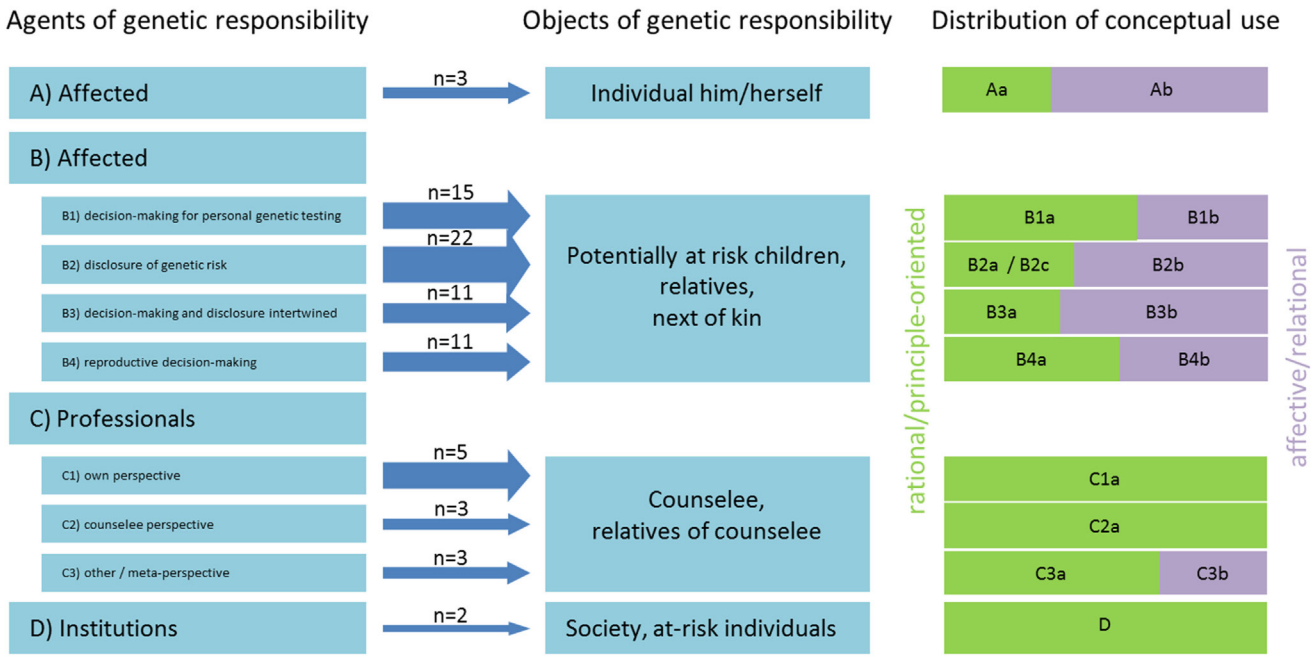

FIGURE 4 | Relations between agents and objects of genetic responsibility and distribution of conceptual use of genetic responsibility. The left column and mid-column show agents and objects of responsibility and the thematic focus of the studies dealing with them, including the number of studies for each category. Right column shows the relative distribution of conceptual use of genetic responsibility in each category: a = rational/principle-oriented type, b = affective/relational type, and c = reactive type.

GR for Agency as a Form of Self-Responsibility (A)

An affected or at-risk person, who is acting in favor of their own health interests by taking knowledge of their own genetic condition as a reason, can be described as displaying GR for agency. This form of GR represents a rather general approach to responsibility in the context of the "new genetics." Although it is frequently evoked in theoretical approaches to GR (Fletcher, 1974; Novas and Rose, 2000; Lemke, 2006; Buchanan et al., 2007), it was found in only three studies (code A) (Kerr et al., 1998; Shipman et al., 2013; Meisel and Wardle, 2014). Moreover, this concept is exceptional insofar as the agent and object of responsibility are identical. To possess agency in the context of GI means to be the cause of the actions taken with regard to that information. Therefore, GR for agency presupposes an autonomous agent who is capable of choosing which GI she wants to obtain, of guiding her own actions, and of setting self-chosen goals and life-plans according to this information.

Framed by a genetic condition, GI is construed as an appeal. The subject is called upon—be it internally by her conscience or externally through social expectations—to acknowledge her genetic condition and to act on this knowledge. However, given the huge gap between prediction and diagnosis on the one side and therapy or effective prevention on the other, the meaning of being genetically responsible for agency is ambivalent. If the agent cannot control her genetic predispositions anyway, what does it mean for her to act responsibly with regard to her knowledge about these predispositions? Hence, acting includes forms of discursive strategies such as "blaming" or "actively ignoring" or practical strategies involving life-style changes, increased healthcare checks, or application of selective technologies such as abortion or embryo selection in cases of reproductive decision making.

Conceptualizations of GR centering around the pattern of "responsibility for agency" were used in the three studies to question whether and to which extent the perception of the ability to make decisions and to act changed with the accessibility of GI. They also pertained to the question of whether increasing knowledge of GI is perceived as a means for improving decision making or rather as a reason for fatalism with regard to one's own future conditions (Shipman et al., 2013; Meisel and Wardle, 2014). For example, Meisel and Wardle found that receiving genetic test results for a multifactorial condition such as obesity enforced narratives of personal responsibility for weight gain. Individuals who tested positively for a genetic predisposition to weight gain had a tendency to interpret this information as confirming the need to actively control their eating behavior. Meisel and Wardle (2014) concluded that:

> Acknowledging an underlying genetic contribution to the susceptibility to weight gain by offering genetic testing may relieve some of the stigma, guilt and self-blame attached to problems with weight control. The 'scientifically objective' personal result appeared to enable some people to recognize that they are 'battling against their biology.' Viewing problems with eating behavior as a 'condition' and not a personal

> shortcoming appeared to increase confidence about managing and overcoming weight problems [Meisel and Wardle, 2014, (p. 183)].

In contrast, another study found that, in cases of GT of tumor tissues for Lynch syndrome diagnostics, those affected did not reflect on (and often did not understand) the implications of the genetic knowledge that would come with testing (Shipman et al., 2013). As a consequence, they did not make any considered choices for managing their condition. Instead they gave accounts of their responsibility with respect to tumor GT only after being subtly directed to the issue by the interviewer and, even then, they still tended to justify their inactivity in gathering knowledge and giving consent for the testing procedure by either reframing or downplaying their role in the actions taken. Hence, the interviewees did not show any responsibility for the issue, even though the outcome of the testing could have had a significant impact on their lives. Shipman et al. (2013) identified two alternative rhetorical strategies used by the participants to justify their inactivity to the interviewer:

> Generalised responsibility is commonly other-oriented and involves strategies that excuse a lack of knowledge or understanding of tumor testing by reframing consent as a hypothetical moral orientation […], civic duty […], and 'blanket consent' […]. Strategies that performed diminished responsibility were self-oriented and justified a lack of knowledge about tumor testing and consent by downgrading its significance […] [Shipman et al., 2013, (p. 238)].

In the latter study (Shipman et al., 2013), GR for agency was presented as a form of "reactive" moral responsibility aimed against the interviewer's subtle expectation that the participant would make considered choices. Instead in the former study (Meisel and Wardle, 2014), GR for agency was more a case of rational moral responsibility. Here, even when results of GT confirmed a genetic predisposition to obesity, the affected people still considered themselves responsible for the maintenance of their health by controlling their eating behavior. According to Meisel and Wardle (2014), this could be explained by the prevailing moral norm in Western societies that health maintenance should be an individual task.

Hence, the exceptional case of responsibility for agency encompasses both moral stances of GR. By focusing on strategies applied to deny responsibility for action rather than an opposing strategy, the study by Shipman et al. represents reactions toward a feeling of being called on to act by a very local social norm. The interviewee apparently felt an obligation to meet the expectation of the interviewer. This expectation, however, did not presuppose responsibility for a general and widely accepted social norm such as "health maintenance is the responsibility of the individual," but the assumption that GT results represent an important moral issue for concern and reflection. This shows that the normative uses of the term GR are independent of its agents and that a person can be understood to be self-responsible in a rational/principle-oriented sense and in a reactive sense.

Both approaches have in common that the primary focus was the subject's capacity to act and choose for herself. This was highlighted in a focus group study (Kerr et al., 1998), in which laypeople were asked to discuss the current and purported future practice of GT. The study reported that laypeople typically stressed the importance of autonomous decision making regarding GT, implying the need to take responsibility for the consequences of one's individual choices. On the other hand, these laypersons struggled with the relevance and validity of the social and moral constraints. The discussions revealed that the simplistic imperative for taking responsibility by making autonomous choices is often undermined by doubts about the actual capacities and possibilities of acting autonomously. For example, the authors observed that "[…] individual choice was clearly valued, but not uncritically, because there was a recognition of the social pressures that compromise an individual's autonomy. In general, participants' accounts of the choices with which clients of genetic testing are faced involved such sophisticated discussion. Several groups talked about cultural difference and the normative nature of our value judgments about medical and behavioral characteristics […]" [Kerr et al., 1998, (p. 120)]. This highlights that self-responsible choices can be shaped by responsibilities toward various cultural, social, and moral norms.

In summary, evoking self-responsibility by GI is yet a rarely addressed topic in qualitative studies, but occurs in contexts of where the lifestyle of an individual is already socially accepted as causality for bad health.

GR of At-Risk People in the Context of the Family (B)

The thematic framework of GR in the context of the family differed from the previous framework with regard to the objects and norms of responsible behavior. Here, the self was only one possible object of responsibility,[8] and the use of GR usually lacked a strong appellative character. Likewise, the norms to which agents were expected to be responsible were more concrete and other oriented and included "respect for autonomy," "the duty not to harm others," and "contributing to the wellbeing of future descendants."[9] These other-oriented norms informed the variety of GR in four ways. First, there were the studies (group B1) that addressed GR by examining individual decision-making processes with respect to GT ($n = 15$). These studies focused primarily on the perceived other-oriented responsibilities of the affected individuals who were currently facing a genetic test or who had faced the decision to undergo GT in the past. A second group of studies (B2) ($n = 22$) focused on the process of communicating genetic risks to relatives and next of kin. Third, the studies in group B3 ($n = 11$) presented both issues together. In this latter group of studies, experiencing the situation of facing a decision to

[8] Transmitting genetic risk information responsibly can theoretically involve a feeling of obligations toward oneself, such as resisting familial pressures to disclose information or preventing harm to oneself, which might otherwise be caused by the reactions of others to the disclosed information. In practice, however, the self is rarely seen as the object of responsible agency in the context of informational genetic responsibility.

[9] These other-oriented norms were obviously drawn from a Western or European cultural context. This reflects the structure of our literature sample.

undergo GT was often associated with considerations about the responsibility of (not) disclosing genetic risks to others. Finally, studies referring exclusively to the context of reproductive decision making, such as carrier testing, prenatal diagnostics of the fetus, or preimplantation screening of the embryo, were included in group B4 ($n = 11$).

Decision Making (B1). Studies in group B1 referred to both rational/principle-oriented and affective/relational approaches to GR. The studies related to the experiences of the participants on receiving test results and the ways in which receiving the GI affected the perceived responsibilities of those interviewed toward others and their reliance on moral and local social norms.

In the rational/principle-oriented approach (code B1a), GR was associated with familiar moral norms such as not harming one's children or society's "gene pool," and the need to comply with these norms or the failure to do so was framed as a burden in these studies (Taylor, 2004; Meiser et al., 2005; O'Doherty, 2009; Smith et al., 2013). For example, Smith et al. used the term "responsibility" to describe the frequent motivation of the participants to do "the right thing for one's children's reproductive decision-making (p. 418)" in GT for HD. For the affected individuals, testing became an issue of moral deliberation involving the norms of not harming offspring and respect for autonomy. Moral reasoning also played a role in decision making in less well-understood hereditary dispositions, such as bipolar disorder. Meiser et al. (2005) reported that participants tended to believe that a genetic explanation would alleviate their personal responsibility for the condition as it reduces the stigma of personal failure. The study by Taylor (2004) is particularly noteworthy because it frames GR toward others as a moral responsibility for the well-being not only of family members but also of others (or society at large).

In contrast to these prospective, future-oriented actions, GR for close others also occurred retrospectively in some studies (Bostrom and Ahlstrom, 2005; Steinberg et al., 2007; Lehmann et al., 2011). In these cases, blaming and feelings of guilt were associated not only with the parental duty to prevent harm but also with interviewees' previous decisions to reproduce.

The ways in which moral responsibilities to the family form the moral justifications of individuals seem, however, to vary depending on the disease context. Hallowell (1999), in discussing women's decisions to undergo GT for HBOC, suggested that responsibilities toward family members interfere with individual choice and agency. Observations from several genetic counseling sessions revealed that, besides the rational/principle-oriented moral decision making and the partly biased information provided by the counselor, the obligation to respect the autonomy of the next of kin and their right (not) to know often constrained the personal interests of those being counseled. In accordance with this, Smart (2010) reported that moral responsibilities toward the family often provide motivation for the affected persons to undergo GT for long QT cardiomyopathies, a set of multifactorial conditions where test results are highly uncertain.

We found the use of affective/relational concepts of GR in only a small number of studies on decision-making processes ($n = 6$; code B1b). One very prominent example was the study by Boenink (2011) showing that expectations about the participant's family relationships that were implicit in the testing procedure significantly influenced the outcomes of the tests. This may be explained by the fact that in the Netherlands, the protocols for the testing procedure imply that the participants live in familial settings that allow them to involve their family in the decision-making process, whereas procedures in the United States assume a much less socially embedded individual. Hence, differing structural expectations shape the strength of other-oriented responsibilities, as perceived by the test participants. Social roles and identities were also emphasized in several studies, which concentrated on the perceived responsibilities of individuals within their personal family relationships (Burgess and d'Agincourt-Canning, 2001; d'Agincourt-Canning, 2001, 2006; Hallowell et al., 2002, 2006; Etchegary, 2006; Felt and Müller, 2011; Etchegary et al., 2015). Challenging the claim that taking a genetic test is an issue of independent deliberation and choice, Etchegary (2006) writes:

> Clinical guidelines for offering the genetic test for HD stress autonomy in decision-making, and genetic counseling embraces the principle of autonomy […]. Yet, narratives of constrained decisions make it difficult to maintain seriously that test decisions are purely rational—self-interested, self-directed, and proceeding in a cost-benefit analysis. Rather, test decisions are taken in the context of one's family [Etchegary (2006), p. 65].

The obligations to others were perceived as inherent in one's role in the family. d'Agincourt-Canning made this clear in a study investigating women's decision-making procedures for undergoing GT for HBOC. Here, gender (among other features such as embodiment, family relationships, and civic self-understanding) intersected with social obligations insofar as being a gendered person implies specific role-related responsibilities for the well-being and health of the kin (d'Agincourt-Canning, 2006), a point we also make in the context of reproductive decision making (cf. below). Decisions (not) to undergo GT were, thus, shaped by the roles and contextual self-understandings of the people to be tested. This was also observed when considering the motivations of women undergoing GT for HBOC. As Hallowell et al. (2002) reported, altruism and the facilitation of risk management for other family members were important motivators. This reflected a specific other-oriented approach to responsibility for women, which was often implied by their familial position. A good formulation of this kind of affective/relational GR was expressed in a study by Manuel and Brunger (2014) on GT for ARVC "[…] relational responsibility reinforces that individual choices are socially embedded within the nexus of our social relationships and the meanings that we assign to these interactions. It also reaffirms that decisions about genetic testing do not happen in silo but are discussed amongst family members, particularly between spouses" (p. 1050).

Similar considerations were also raised by Raz et al. (2014) in an Israeli-German study, who framed their responsibilities toward their future children as motivation for having a genetic

test: "lt was common amongst Israeli respondents, but not German respondents, to speak about their motivation for taking the HD test in the context of their children, as one Israeli respondent summarised it: '[Taking the test] is my responsibility as a mother'" [Raz et al. (2014), p. 187].

In sum, a common understanding of hereditary conditions provokes a meaning of GR as family decision making and by this enlarges the radius of action from the individual to his/her family.

Disclosure (B2). Generally, explicit notions of GR in the context of family communication and disclosure occurred frequently in the qualitative literature (n = 22). To be responsible with regard to disclosure of GI was usually taken to mean acting sensibly with regard to the effects this information might have on children, relatives, and next of kin. Gaff et al. (2005) gave a comprehensive definition of GR in the disclosure context:

> Taking responsibility for transmitting information implies acting responsibly towards fellow family members. To act responsibly is to assess the potential consequence of sharing or withholding information and is embedded in a moral economy consisting of (mutual) assessments of competence and maturity, as exemplified by the deliberative communication strategy. Communication of genetic risk information is the result of intricate, existing ties of differing strengths [Gaff et al. (2005), p. 1003].

In line with this definition, studies in the B2 group frequently investigated the different strategies of communicating, transmitting, and managing genetic risk information in a very close social context.

In the given context of disclosure, GR was predominantly construed as a form of affective/relational responsibility ($n = 13$, code B2b) and, slightly less often, as a rational/principle-oriented responsibility ($n = 9$). Among these rational/principle-oriented approaches, we found descriptions of GR according to the "standard" subtype ($n = 5$, code B2a) and the "non-standard," reactive subtype ($n = 4$, code B2c). These patterns are discussed in this section, starting with the prevailing affective/relational responsibility framework.

The affective/relational approach to GR first appeared in studies with a feminist theoretical background that focused on women at risk for HBOC. However, studies using other theoretical backgrounds, such as the sociology of science (Felt and Müller, 2011), and other genetic conditions as models, such as HD (Etchegary and Fowler, 2008) or blood clotting predisposition (Parrott et al., 2015), also confirmed this approach. Dancyger et al. (2010) exposed an understanding of an affective/relational informational approach to responsibility when they wrote:

> All 'committed' family groups described their primary motivations for testing as arising from an obligation to others, while also acknowledging some, but less strong, motivations to be tested for their own benefit. […]. This obligation to others can be considered in terms of genetic responsibility: to do what is morally right for the family [Dancyger et al. (2010), p. 1294].

Some studies considered familial roles other than that of women managing health in the family, such as those of male partners and fathers (Hallowell et al., 2006) or those of parents toward children (Forrest et al., 2003; Arribas-Ayllon et al., 2008b; Manuel and Brunger, 2014; Vavolizza et al., 2015) or of adults toward distant relatives (Etchegary and Fowler, 2008; Etchegary et al., 2009).

The application of an affective/relational approach to GR in the disclosure context also involved the description of constraints and facilitators with respect to family communication. Several studies stressed that transmission of genetic risk information depends on a variety of individual, social, and situational factors (Forrest et al., 2003; Gaff et al., 2005; Keenan et al., 2005; Mesters et al., 2005; Ratnayake et al., 2011). "Feelings" of being responsible for the well-being of others or even anticipated feelings of regret for causing harm were strong facilitators for disclosure. The severity of the genetic condition also motivated sharing the information. Disrupted or overly tense family relationships, on the other hand, were barriers for disclosure. Studies generally reported that the affected people viewed disclosing genetic risk information as a great responsibility or even as a "burden" (Bonadona et al., 2002).

Besides gender and family roles (i.e., being a woman, being a parent, being part of the nuclear family) (d'Agincourt-Canning, 2001; Mesters et al., 2005), the studies named the severity of the disease (Weiner, 2011), emotional ties between the informant and the hearer (Gaff et al., 2005; Carlsson and Nilbert, 2007), family dynamics (Forrest et al., 2003), and the spatial distance between the relatives (Etchegary and Fowler, 2008) as factors influencing bonding and responsibilities. Mostly, these bonds were also reported to influence the wish to balance other-oriented responsibility and autonomy (Hallowell et al., 2003; Foster et al., 2004).

In some contexts, biological kinship can make a difference to the approach to responsibility with respect to disclosure. For example, a Turkish study by Akpinar and Ersoy (2014), which asked patients and physicians about their responsibilities in the process of disclosing genetic risks, revealed a difference in the ascription of responsibilities toward spouses and siblings. Only a few physicians and affected individuals saw it as the responsibility of the individual to inform a spouse, while there was strong consensus that it was the affected individual's responsibility to inform a sibling. This shows the importance of specific relationships for the perception and conception of GR.

A notable exception to this seeming pervasiveness of the affective/relational approach to GR was seen in a study by Weiner (2011). This study investigated responsibility in the context of familial hypercholesterolemia (FH), the symptoms of which can be controlled comparatively easily. The author reported that felt obligations to disclose the disease to children or next of kin were only of marginal importance to the affected individuals. This is surprising, because most forms of FH are caused by a dominant single gene mutation, with a 50% chance of inheritance. Obviously, the severity and treatability of a genetic disorder plays a major role in the felt obligations and responsibilities [cf. also Raz and Schicktanz (2009) and Boardman (2014b) for similar findings].

The standard subtype of the rational/principle-oriented approach to GR was referred to when non-disclosure of genetic risk was discussed as a problem resulting from conflict between moral norms. Here, the conflicting norms were again "respect for autonomy" vs. "the obligation not to harm." Respect for autonomy justified non-disclosure if the recipient did not want to know his genetic status. The "do-no-harm" principle was mostly used to justify an obligation to disclose GI in order to allow the recipients of the information to take action to prevent possible future harm caused by their genetic condition. It was, however, also used as an argument against disclosure, if the tested person concluded that the recipient needed to be protected from information with which she would be psychologically incapable of coping.

An early study adopting a standard view of rational/principle-oriented responsibility based on the ethical dilemma between respect for one's own interests and the no-harm principle was carried out by Hallowell (1999). The author reported that women affected by HBOC tended to relinquish their right not to know about their genetic makeup—sometimes to the extent that they seemed to dispense with their respect for their own autonomy in favor of the effect that others were not harmed—because of perceiving a strong moral obligation to disclose genetic risk information to reduce harm. This view about moral obligations toward others was not restricted to women at risk for HBOC. As Vavolizza et al. (2015) reported for the case of cardiac arrhythmias leading to unexpected death, disclosure was perceived as a moral obligation by the affected people in the majority of cases. This obligation seemed the stronger, the more pressing, the more severe and uncontrollable the consequences of the genetic condition were. For cardiac arrhythmias, which cause no pain and are usually only discovered after a death in the family, disclosure was widely acknowledged as a moral obligation. Where consequences seemed more controllable and not necessarily fatal, the obligation was thought to be weaker. For instance, Dancyger et al. (2011) reported that information was edited or withheld in HBOC families if those who were tested considered the recipient's capacity to cope with the information as diminished.

Finally, four studies (Hallowell et al., 2006; Arribas-Ayllon et al., 2008a,b, 2011) analyzed GR by focusing on communicative strategies during interviews and counseling sessions, drawing on the "reactive" subtype of the rational/principle-oriented approach to GR. This notion was particularly noteworthy in the work by Arribas-Ayllon et al. (2008a,b, 2009, 2011). These studies described a variety of rhetorical strategies used by the interviewees to argue that their behavior was genetically responsible in family negotiations. Despite not having disclosed genetic risk, affected or at-risk individuals used blaming others or themselves to appear responsible. These accounts of GR are interpreted as resistance against a common sense moral framework that framed only well-considered and autonomous management of genetic risk information as responsible behavior (Arribas-Ayllon et al., 2011). The authors did not interpret feelings of guilt and blame as signifying responsibility but rather as artifacts evoked by the interview situation. By giving accounts of GR in terms of feelings of guilt, interviewees were reacting to a dominant moral framework underlying the social discourse about GT. In line with the sociological analyzes by Novas and Rose (2000), taking *individual* responsibility for the management of genetic risk was morally required. Drawing on this framework, the authors concluded that non-disclosure of genetic risk to family members and the associated affect of guilt must be interpreted as expressions of the "obligation to manage GI." This means that the concept of GR in these studies was very similar to the concept in group A studies. This is not because responsibility would be necessarily directed to the self as the object, but rather because accounts of responsibility expressed the feeling of being called to engage in the management of genetic risk. The following quote from Arribas-Ayllon et al. (2011) illustrates this idea.

> […] not speaking or sharing the truth of genetic risk is itself a form of expression not captured by structures of signification. Nevertheless, non-disclosure warrants an account because our participants implicitly recognised that they live in a moral order where genetic risk should be communicated to kin. And yet, the explanations they gave for this were redundant forms of moral communication (that is, post-hoc justifications and excuses). […] we suggest that people resist the onerous obligation of managing genetic risk and disclosing bad news to kin to escape the moral/discursive confinement of autonomy [Arribas-Ayllon et al. (2011), p. 19].

Overall, disclosure was not solely seen as an implication of GR. Some cases also revealed a complex form of non-disclosure as a *moral* implication of GR. However, the justification of whether to share or to hide GI was based on underlying moral principles (e.g., autonomy) and their concrete interpretation with respect to the family situation or illness.

Decision Making and Disclosure Intertwined (B3). Some studies (n = 11, code B3) revealed that, from an affected person's point of view, the decision for a test and the disclosure of GI were inextricably related to each other (Burgess, 1994; Bostrom and Ahlstrom, 2005; Etchegary and Fowler, 2008; Etchegary et al., 2009; Raz and Schicktanz, 2009; Dancyger et al., 2010; Smart, 2010; Vears et al., 2015). This approach was, however, often indirect, as studies often focused on the psychological and social changes in an individual resulting from the disclosure of a test result. With respect to responsibility, the changes in the tested person's perceptions of their moral obligations toward others are of interest. Changes of this kind were, for example, reported from a study using hereditary epilepsy predisposition as a model (Vears et al., 2015). The study showed that receiving a positive test result affected the perceived moral responsibilities toward others in various possible ways:

> Interestingly, some parents felt identification of the causative gene meant the responsibility of passing on the epilepsy had been removed. Yet other parents were grateful the genetic information had not been available when they were having children, removing the need for decision-making in family planning […]. Feelings of 'genetic responsibility' were expressed by some of our participants, who described a desire to

> prevent epilepsy in future generations […]. Some family members were apprehensive for those who had had children despite the risk of passing on epilepsy [Vears et al. (2015), p. 69].

Other studies noted a conflict between self-responsibility for health and the responsibility to disclose genetic risk to the next of kin. While in Western societies the individual is considered obliged to make decisions with regard to her body and health—such as the decision to undergo GT or not, knowing one's genetic status is perceived as implying the responsibility for disclosing genetic risk information to prevent potential harm to others. In a comparative analysis of affected individuals' and laypeople's attitudes toward GT in Israel and Germany (Raz and Schicktanz, 2009) revealed this conflict between the self-responsibility of knowing one's genetic status and the responsibility toward others that comes with this knowledge:

> The moral conflict was described as consisting of, on the one hand, the moral responsibility towards kin, but on the other hand the recognition that how to handle such information is a genuinely individual decision – especially about health and the body, which everybody has to care for on their own [Raz and Schicktanz (2009), p. 437–438].

However, depending on the disease context, the decision to have GT did not necessarily conflict with the responsibility for disclosing the genetic risk. As Dancyger et al. (2010) observed, the expectation of disclosure of genetic risk information to the next of kin, in order to take responsibility for their health behavior, sometimes motivated the decision to have GT in the first place. This was particularly true for mothers at risk for HBOC, who perceived their daughters' future options in healthcare decision making as a reason to have GT: "She told her daughters about her mutation the day she received her result and was keen for her daughters to 'get on with it' and be tested themselves. One daughter was aware of her mother's motivation for testing and interpreted her communication about the test result and genetic risk as nagging" [Dancyger et al. (2010), p. 1291].

This mother clearly expressed a rational/principle-oriented approach to the responsibility to have GT and to disclose the test result, motivated by her perceived obligation to reduce harm for her daughters. At the same time, one might argue that she disrespected her daughters' right to decide whether they wanted to know this information. Responsibility for others played a significant role in the motivation to have GT, especially for preventable or treatable diseases. In particular, if the testees were already affected by the disease, the decision to have a test to determine whether the disease was heritable was shaped by the expectation that the result would be disclosed to help the next of kin. As Etchegary et al. (2009) reported in a study on GT for HBOC and HNPPC:

> Study participants described their responsibility to their families particularly children to provide risk information that could be used in cancer-risk management decisions. […] Indeed, many suggested the primary motivation for testing was for relatives, more than for themselves [Etchegary et al. (2009), p. 259].

In contrast to the case in Dancyger et al. (2010), the authors were referring to an affective/relational understanding of GR. The decision to take the test was interpreted as suggesting that "some people at risk for inherited cancer did not construct as independent, making choices that only affected themselves. Rather, in decisions about inherited cancer risk the self was constructed as interdependent" [Etchegary et al. (2009), p. 259]. Here, the obligations appeared to be localized in the specific context of the family and were not framed as a general moral obligation to disclose the genetic risk. Such localized responsibilities may affect the testing decision in the opposite way. As d'Agincourt-Canning (2006) reported, declining a test can be motivated by felt responsibilities toward others, as well, when disclosure of genetic risk status to kin might be considered to be doing more harm than good.

It was an important point of many qualitative studies on GT that local, affective/relational responsibilities undermined the picture of the autonomous agent underlying the ethical principles prevailing in counseling guidelines for GT (Hallowell et al., 2003; Etchegary and Fowler, 2008). This point was displayed in an extreme way in a study by Foster et al. (2002), who discussed a case in which affective/relational responsibility for taking a genetic test expressed as the wish "to keep everybody else happy" (p. 479) was not primarily perceived as a responsibility toward family and kin, but as a reaction to the physician's invitation to take the test.

From an intercultural perspective, few studies reported differences in the responsibilities perceived between different Western societies (Raz and Schicktanz, 2009; Raz et al., 2014). While Miller et al. (2014) observed an acknowledgment of the importance of disclosing genetic risk information in interviews with Westerners about genomic sequencing for late-stage cancer care, disclosing GI to the wider family was perceived as less morally important in some groups with a non-western cultural background. Similarly, Shaw and Hurst (2009) reported that disclosing genetic risk to the wider family was not perceived as a moral responsibility in Pakistani communities in Great Britain. Rather the information was understood as belonging exclusively to the couple or the tested person.

In sum, moral conflicts occurred regularly on two levels of decision making: first whether and when to make a GT and afterward, whether and when to disclose/not to disclose the results of such a GT within the family.

Reproductive GR (B4). Studies discussing GR mainly in the context of reproduction involved GT for hereditary diseases of the fetus or embryo, carrier testing, and prenatal blood screening for genetic anomalies of the fetus. The concept of GR in this respect differed from the situations above in that it focused neither on the self-agency nor on communication, but on the decision to have children or the decision to have GT during pregnancy or to select embryos by preimplantation genetic diagnosis before the pregnancy. Importantly, the studies in our data set differed

significantly from the theoretical approach to reproductive GR of the 1970 s and classical eugenics (Lipkin and Rowley, 1974). None of these studies suggested "classical eugenics," conceptualizing genetically responsible reproductive behavior as decision making in favor of optimizing the gene pool of society or a population.

As Hoeltje and Liebsch (2015) argued in their German interview study with those affected in the context of CF, this "new" paradigm of GR stands in contrast to classical eugenics as an imperative for those affected to self-determinedly "select" and understand their decision as an individual choice [cf. also Habermas (2005)]. Rather than the imperative to impact a populations' gene pool, it is to avoid individual suffering. However, as the authors concluded, the result might be the same.

Therefore, the studies focused on private reproductive decisions, which were guided by different individual norms. The decisions and actions described were more negative-selective (e.g., deciding for abortion or for relinquishment) than positive-selective (e.g., actively choosing one's children's genetic make-up). Some studies examined explicitly how these private decisions by the parents were influenced indirectly by public discourse and not—as in the case of classic eugenics—explicitly enforced by state authorities (Boardman, 2014b; Yau and Zayts, 2014; Hoeltje and Liebsch, 2015). Another group of studies focused on the role of "experienced knowledge" of the illness within the family and how this influenced reproductive decisions.

The studies in the field of reproductive GR fell into two subcategories: studies focusing on the process of making a decision about having a child if the risk of having a baby with a genetic disease was considerable ($n = 7$) (Downing, 2005; Dekeuwer and Bateman, 2013; Donnelly et al., 2013; Boardman, 2014a,b; Yau and Zayts, 2014; Hoeltje and Liebsch, 2015) and studies using gender as a category to investigate the role of future parents in the context of prenatal genetic screening procedures ($n = 3$) (van Berkel and Klinge, 1997; Reed, 2009; Atkin et al., 2015). In addition, one study approached reproductive GR indirectly *via* the perceptions and opinions of genetic counselors (Ekberg, 2007).

Similar to the context of disclosing genetic risk in the family, some studies focused on a rational/principle-oriented approach to responsibility ($n = 6$, code B4a) (Ekberg, 2007; Dekeuwer and Bateman, 2013; Donnelly et al., 2013; Boardman, 2014a,b; Yau and Zayts, 2014) and some on an affective/relational approach ($n = 5$, code B4b) (van Berkel and Klinge, 1997; Downing, 2005; Reed, 2009; Atkin et al., 2015; Hoeltje and Liebsch, 2015).

The focus on GR in the rational/principle-oriented approach might have been invoked by the fact that, in many studies, parents or affected persons were interviewed about the hypothetical or rather abstract scenario of whether to test for a genetic disorder. This was 'hypothetical' insofar as most studies were not conducted within the setting for prenatal testing, but approached the affected people long before or after they made their reproductive decision. Two interview studies in the context of BRCA mutation revealed 'a dual dimension' to how women perceived it to be a moral dilemma when they were to be tested for their own health, but anticipated in advance that a positive result might negatively impact on their reproductive decisions later (Dekeuwer and Bateman, 2013; Donnelly et al., 2013). Overall, they prioritized their own health; reproductive decisions were a less relevant motivation for GT. Receiving GI resulted in attempts to 'manage' the risk in various ways (closer surveillance, prophylactic mastectomy, or timing of conception in cases of radiochemotherapy). It was always deeply embedded in the lived experience of the disease and depended on whether the wish for their own children was made explicit before the testing (Dekeuwer and Bateman, 2013). Parental responsibility meant "to justify this choice [to have a child and to potentially transmit the BRCA mutation] to her children" [Dekeuwer and Bateman (2013), p. 240]. The non-standard reactive meaning of rational/principle-oriented GR occurred in the sense that the interviewees argued that they would be held accountable for decisions that affected themselves and their children. This dilemma could also be seen as a choice between two types of risk: the risk of knowing vs. the risk of occurrence (to have a child suffering from a disease). As an observational study in a genetic counseling setting in a Hong Kong hospital revealed, the risk of knowing was mainly put forward as an argument when clients wanted to signal their responsible accountability not to test (Yau and Zayts, 2014). A particular implication of reproductive GR was evoked in families with children who had a serious, untreatable recessive disease such as spinal muscular atrophy (Boardman, 2014b). In these families, the so-called expressive objection, a critique of GT saying that testing and abortion not only express a negative validation of the fetus tested but also of the lives of people living with the disability, framed the understanding of "responsibility." The interviewees were aware of how their reproductive decision could be interpreted by others, especially by existing children with the disease. Hence, accountability for decisions taken became crucial, but depended on the social relationships in which the actors were embedded. Here, the notion of responsibility was associated with morally embedded relationships, in contrast to the idea of an individualized agency or personal choice. Furthermore, stressing responsibility as increased awareness or practice of justification was supported by socioempirical findings that responsibility or related understandings were used for both, even contrary outcomes of decisions, namely, to test as well as not to test, to abort as well as not to abort.

The affective/relational approach to GR was, instead, one of the main motives in studies focusing on gender issues. Reed (2009) claimed it was the "gendered nature of genetic responsibility (p. 343)" by which gender stereotypes formed expectations and performed in their parental roles as a mother or a father. In the context of reproduction, the idea of an embodied responsibility occurred as (essentialist) feelings of maternal responsibility for the well-being of the fetus (van Berkel and Klinge, 1997). However, when it came to high-risk screening results, women's articulation of maternal responsibility varied in extent with respect to men's involvement. If men were involved in carrier testing, women were more relaxed about their own responsibility. The study by Atkin et al. (2015) also pointed to an increased masculine responsibility during carrier screening. Genetic screening provided men with a chance to feel bodily engaged and to demonstrate responsible "fatherhood." Finally, one small but detailed case study by Downing (2005) on HD

families illustrated how responsibility was negotiated by family members independently of whether they accepted or rejected GT.

In summary, GR in the context of reproduction did not provide a simplistic model of eugenic attempts, but implied a complex process of moral interpretation and justification. Furthermore, the review has revealed that GR is associated with different outcomes of decisions, including opposing ones. However, only a few studies comprehensively explained their conceptual understanding of terms such as responsibility or GR (Downing, 2005; Reed, 2009; Hoeltje and Liebsch, 2015). Most seemed to assume what is meant by responsibility in the context of GT as self-evident.

Professional GR (C)

In the studies in group C, physicians, nurses, geneticists, and laboratory workers associated with performing the genetic tests were the agents of GR. These studies primarily investigated the roles and obligations of healthcare professionals in GT in general, as well as the obligations of genetic counselors to patients and their families. The norms of responsible behavior included respect for the autonomy of the counselee (expressed in an appeal not to direct their decision), the obligation to secure informed consent, and a duty to prevent harm to the counselee's family members (sometimes against the will of the counselee). There were three subtypes in group C. Subgroup C1 comprised studies that investigated professional GR by directly drawing on the views of healthcare professionals about their daily work and practice in genetic counseling or GT ($n = 5$, code C1) (Arribas-Ayllon et al., 2009; Bredenoord et al., 2010; Hines et al., 2010; Townsend et al., 2012; Hens et al., 2013). These studies investigated the professionals' subjective assessments of the distribution of responsibility in clinical practice. The second subgroup (C2) addressed professional GR indirectly *via* the expectations of people, who were genetically at risk, or unaffected people confronted with hypothetical ethical dilemmas ($n = 3$, code C2) (Burgess, 1994; Haga et al., 2012; Daack-Hirsch et al., 2013). The third subgroup ($n = 3$, code C3) approached professional GR from a third-party perspective using interview material with physicians involved in genetic medicine to describe paradigm shifts in genetic counseling that affected professional duties and responsibilities (Bogner, 2013), or to argue that healthcare professionals in genetics tended to refer to legal or professional regulations to escape confrontation with their personal responsibilities (Wieser, 2011; D'Audiffret Van Haecke and de Montgolfier, 2015). These studies did not directly address professional GR but indirectly drew conclusions about GR by describing and analyzing the subtext of interview material about physicians' daily practice of genetic medicine.

The studies often took ethical dilemmas as a starting point to discuss professional responsibilities. In subgroup C1, professional GR was primarily construed as a form of rational/principle-oriented responsibility. With respect to the management of the uncertainties implied in genetic test results, genetic counselors saw it as their responsibility to secure informed consent by providing as much relevant, balanced information as possible (Arribas-Ayllon et al., 2009; Bredenoord et al., 2010; Hines et al., 2010), to protect the autonomy of the patient (Hines et al., 2010), and (if relevant) to prevent harm to the patient's relatives by supporting or even intervening in disclosure to the family. As Bredenoord et al. (2010) put it with regard to the specific uncertainties of GT for mitochondrial DNA disorders, the moral impetus of securing informed, autonomous decision making is central to professional GR: "In our view, it belongs to the responsibility of professionals working in this field to try to help affected couples to make a well-informed decision, even if this cannot always consist in providing them with precise data about remaining risks" [Bredenoord et al. (2010), p. 16].

Other responsibilities of healthcare professionals were more context dependent. Studies mentioned that healthcare professionals involved in genetics/genomics were also aware of being responsible for providing psychosocial support to patients (Hines et al., 2010), controlling uncertainties in testing of complex diseases by referring patients to more experienced colleagues (Bredenoord et al., 2010) and actively supporting the patients' reproductive decision making in terms of helping them to figure out the relevant factors and values involved in the decision (Hines et al., 2010). In cases involving the GT of underaged children, genetic professionals were also reported to be responsible for actively balancing the parents' avoidance of responsibility not to cause harm to the children (Arribas-Ayllon et al., 2009). In all these context-dependent cases, however, professional GR was also construed as a rational/principle-oriented concept.

Studies in subgroup C2 reported that, from the perspective of the counselees, the professional responsibilities of the genetic counselors and physicians sometimes extended further than believed by the genetic counselors and physicians themselves. For example, Haga et al. (2012) reported that laypeople expected professional geneticists not only to explain pharmacogenetic information in an understandable manner but also to manage the disclosure of ancillary genetic risk information. Daack-Hirsch et al. (2013) reported that, during whole-genome sequencing, laypersons attributed responsibility to the genetic counselor to disclose incidental findings and to suggest adequate follow-ups.

These expectations also indicate that professional GR is a form of rational/principle-oriented responsibility, because the underlying norm to which professionals are expected to conform is the protection of the patient's autonomy and action with their consent. Patients expected to make their own choices but also expected professionals to disclose any GI that might be relevant for their decisions about how to act.

Two studies in subgroup C3 focused on the GR of professionals in relation to (novel) legal regulations associated with genetic counseling. A French study (D'Audiffret Van Haecke and de Montgolfier, 2015) used a rational/principle-oriented approach to professional GR. The authors observed that legislation requiring genetic counselors to disclose the genetic risks associated with fatal genetic diseases to affected family members incited conflict between the counselor's responsibilities to protect the patient and the patient's next of kin and their responsibility to respect the interests of the healthcare system and society at large. The authors concluded that the new legislation brought about a situation where:

> We are at the crux of a debate opposing individual and collective responsibility, with the healthcare professional—as actor in disease prevention and in matters of public health—at the center, caught in the crossfire between acting in the interests of the general population and acting in the interests of their patient and family [D'Audiffret Van Haecke and de Montgolfier (2015), p. 10].

To handle this conflict, genetic counselors applied different strategies resulting in different outcomes with respect to their moral obligations. For example, in the same study, genetic counselors "recognize that the law gives them something to fall back on in 'those rare cases' where patients refuse to warn at-risk relatives (I1), to 'counter refusals when the reasons are as much to do with family feuds, geographic separation, or fear of genetics and the allied denial as anything else' (I7)" [D'Audiffret Van Haecke and de Montgolfier (2015), p. 6]. Other professionals "took the stance that there was no real need to legislate since informing patients of risks to relatives is part of professional duty, and developing disclosure support is part of professional practice (I2)" [D'Audiffret Van Haecke and de Montgolfier (2015), p. 6].

Non-performance of professional GR was also an important issue in an Austrian study, which found that reference to legal regulations or to the medical code of conduct served as a rhetorical strategy for genetic professionals to repudiate GR in ethical dilemmas (Wieser, 2011). Hinting at the fact that genetic professionals, who are the experts for the issues under legal regulation, contributed to existing legal regulation and hence were at least partly responsible for it, the study revealed their denial of responsibility as morally flawed:

> The interviewees [medical geneticists] repeatedly drew on the law in order to convince the interviewer that ethical issues of genetic medicine were sufficiently cared for. […] Essentially, in such a way the interviewed medical experts provided answers with reduced accountability [Wieser (2011), p. 178].

The only example of an affective/relational approach to GR was provided by a study focusing on a paradigm shift with respect to genetic counseling (Bogner, 2013). The study argued that the concept of genetic counseling had shifted from a paternalistic, knowledge-based approach to a participatory model. Instead of providing information, the new participatory approach implied that genetic counselors have responsibility for coaching the patient during her decision-making process. This shift could best be explained by the idea that the person at risk had to take increased individual responsibility for her health, while the genetic counselor's responsibility was reduced to adhering to the moral ideal of "neutrality" and non-directive coaching.

In summary, professional GR was discussed mainly as community skill- and capacity-based awareness of professionals to be sensitive to the various moral dimensions and conflicts. It is embedded in a general understanding of an asymmetric professional-lay relationship where the asymmetry relies not only to factual knowledge but also to the social implications of sharing GI later on.

The GR of Institutions (D)

Only two papers (code D) discussed the GR of institutions (van Hoyweghen et al., 2006; Bickerstaff et al., 2008). van Hoyweghen et al. (2006) analyzed processes of responsibilization in life insurance companies and their policies, showing how they constructed different notions of being at risk. While, from the institutional point of view, genetically at-risk people were identified as victims of circumstance who deserved solidarity or "collective responsibility" [van Hoyweghen et al. (2006), p. 1232], people with risky lifestyles and behaviors were identified as being responsible for their own health risks and were thus held accountable for themselves. According to the authors, life insurance companies functioned as a normative "technology and practice" that shaped ideas and notions of responsibility. Conversely, this implied the responsibilization of society in general as carers for genetically at-risk people who were found worthy of solidarity, while voluntary risk-takers were dismissed as being unworthy of solidarity. Thus, this article introduced two possible agents of responsibility: society or the insurance company as agent and the (explicitly non-genetic) lifestyle risk-taking individual taking responsibility for his/her own actions, as opposed to the genetically at-risk individual who was not responsible for his/her condition.

Bickerstaff et al. (2008) discussed GR in the context of citizens' perceptions of "responsibility in relation to a number of risk issues" [Bickerstaff et al. (2008), p. 1312], of which GT was only one of six. Strikingly, laypeople participating in the study discussed an institutional responsibility for application of GT technologies. GR was described as a "responsibility for the management of environmental and technological risks" and was ascribed to institutional actors, such as "government" and "science," because of their specific competence to resolve those issues. Hence, responsibility resulted from the institutions' unique agency, i.e., "knowledge and power to take meaningful action" (all quotes p. 1321). In this model, society was the object of a social responsibility attributed to institutions that had a duty of care, deriving from their more or less exclusive abilities. It was noteworthy that this type of responsibility was not about genetic risk information as such, since the mentioned institutions were concerned with creating and shaping regulatory frameworks of GT technology and its usage. They were, on a meta-level, responsible for the prevention of harm to individuals or to society that may occur as a result of the application of GT technologies.

Both papers framed the GR of institutions as a rational/principle-oriented concept, since the agents were able to take on rational responsibility, basing their duty on their unique role as a factual authority that was trusted to fulfill its protective role toward individuals or society. Overall, the role of institutions was rarely addressed, but the few studies revealed the need for more comprehensive understanding of collective actions and collective responsibility in relation to GI.

DISCUSSION

Our findings revealed that, in empirical-qualitative studies, GR was seen as a subspecies of responsibility that was biased toward certain relata and conceptual dimensions. The basis of the GR concept was primarily the prospective responsibility of affected or at-risk individuals to their family and next of kin (including potential future children) but was also, albeit to a far lesser extent, the responsibilities of genetic counselors and medical doctors. Institutional responsibilities have been relatively neglected. The focus of the papers was primarily on the individual in their close, personal social environment. A wider social or political perspective was only visible in rare cases, where GR occurred as retrospective responsibility in a 'reactive' manner. This seemed to be the only context in which social norms and expectations beyond the individual family appeared on the conceptual horizon. Interestingly, this individualistic form of GR that we found in the material confirmed a common pattern in the current sociological and bioethical discourse. The problem of how to use the increasing possibility of obtaining and managing GI has induced a shift in the medical sociological and bioethical literature on GT from an optimistic rhetoric of autonomy and choice toward a more pessimistic, critical approach. GR no longer only stands for the promotion of the well-being of future generations, but has more and more become a notion that reflects a major social trend toward a proliferation of responsibilities, amounting to a general responsibilization of the individual in questions of health. The term responsibilization describes the phenomenon of the increasing transfer of responsibility for handling health risks from collective or institutional agents to the individual. In health care especially, with the new technologies and means for control, new risks and possibilities for acting are developing. In the process of responsibilization, individual agents are becoming the carriers of these new responsibilities (Ter Meulen and Jotterand, 2008; Young, 2011). In contrast to this mapping of the uses of GR in the qualitative research literature, this review also found an empirical and theoretical necessity to employ a more differentiated notion of GR in current bioethical and biopolitical discourses. In fact, responsibilization was rarely discussed in the qualitative studies we reviewed.

Our results indicated that, in the qualitative medical sociology literature, GR is a complex concept that is embedded in a web of dependencies, relationships, and expectations. GR can, hence, be approached from a variety of different angles. However, most studies in our corpus centered on the ascribed or projected responsibilities of at-risk or affected people and, therefore, favored a rather pragmatic approach to the ethics of GT. These responsibility ascriptions, however, varied in general terms along three major pathways.

First, there was a generalized assumption associated with GR that action should be taken with regard to the mere possibility of GI. These actions, however, should encompass various forms. Taking responsibility for one's own health status seemed widely perceived as the responsibility of the affected individual, whereas the application of GT is continuously enlarging the spectrum of affected agents. Hence, the prospect of using GI to obtain guidance for individual health maintenance has created a space for self-directed action. Other-oriented blame and self-justification by affected individuals who did not disclose GI, as observed by some studies, can be interpreted as a reaction to this general feeling of being called to use the information for one's own good, for the good of one's kin, or even for society's greater good. Because this responsibility is often not obvious or implicit, it was only very seldom directly revealed in interview studies but has instead appeared as part of the interpretation process. The apparent difficulties associated with showing this pervasive pattern directly by empirical investigation, however, lend doubt to the idea that an affected individual is able to consciously perceive these social constraints to their decision making or is aware of the social process of responsibilization.

Second, there were more mundane patterns to be seen in GR. We found both rational/principle-oriented and affective/relational approaches to GR in the context of the family with respect to GT. The quantitative prevalence of the affective/relational approach might be an effect of sociologists' interest in social roles and their sensitivity to interpersonal dependencies, which can shape an agent's responsibilities—a fact that is hardly discussed in bioethical or legal debates (Beier et al., 2016).

As supported by philosophical-theoretical approaches to responsibility (Hart, 1968; French, 1991; Yoder, 2002; Held, 2006; Young, 2011), these decisions were not undertaken by an autonomous deliberator, but were mutually shaped by social roles and expectations. However, the use of responsibility in many studies would benefit from more distinct reflections on the underlying relationships. The rational/principle-oriented concept of GR seemed, instead, to be used to describe conflicting obligations and processes of moral deliberation. Hence, insights into how laypeople address such moralities, and practical ways of naming the conflicts related to GT, would allow the underlying norms (such as a person's autonomy, the duty not to harm, and others) to be delineated.

Third, the contexts of reproductive or institutional decision making were both susceptible to implied obligations to society—whether now or in the future. This is not surprising, given the historical legacy of traditional eugenics which proposed a benefit for future society based on austerity of the individual. However, it is striking that most Anglo-American studies circumvented the context of eugenics and only one German and one German-Israeli study referred to it explicitly.

Only a small number of studies construed GR as the responsibility of genetic professionals. This is striking for two reasons. First, professional GR was almost exclusively construed as a rational/principle-oriented responsibility based on concrete professional norms like securing informed consent, respecting the client's autonomous decision making and preventing harm. However, studies on professional GR rarely investigated the concrete implementation of these norms, which would require methodological access to the practice and critical reflection on it, instead using observation and critical communication studies. It would, for instance, be interesting to know how the characterization of the responsibilities ascribed to (or self-ascribed by) genetic professionals maps on the current ideal of non-directive

genetic counseling as defined by the UNESCO (United Nations Educational, Scientific and Cultural Organization, 2004)[10] It is probable that the praxis of professional GR, before individual autonomy and—as a consequence—non-directive counseling became the prevailing paradigm in applied medical genetics, did not look the same as it does today. Second, perspectives on professional GR often differed between patients and professionals. More generally, there seemed to be a gap between the professional's perceived obligation to manage the conditions for informed and autonomous decision making by the patient and the patient's expectation to feel and act according to their own responsibility. This calls for a reconsideration of the roles and obligations in the relationship between genetic counselor (or physician) and patient.

This is supported by the observation that some studies implied that the availability of genetic tests did not necessarily correlate with the increase in the perceived responsibilities of the affected individuals toward their family or next of kin (Weiner, 2011; Meisel and Wardle, 2014). It was less likely that a study reported feelings of responsibility toward others where the etiology of a disease was more complex, its probability of transmission into future generations was lower, and the possibilities of controlling the medical consequences of the health condition were better. Patients typically did not think that it would be irresponsible not to disclose genetic conditions that do not significantly differ from acquired forms of the disease in terms of clinical symptoms (e.g., FH or obesity) to other family members. The studies on these kinds of diseases undermined the idea that at least this form of GR is proliferating since the advent of GT.

Conversely, as the focus of qualitative empirical research on GT was mainly on interpersonal family relationships, the influences of the increased possibilities for GI were not clearly revealed on a broader social and institutional level. This was supported by the blatant underrepresentation of empirical studies focusing on institutional responsibilities. The role of institutions such as insurance companies, state legislation, and professional associations has been insufficiently examined empirically (Heyman and Henriksen, 2001; Bogner, 2005; Hashiloni-Dolev, 2007).[11] The same holds for contexts beyond industrialized western countries. We find it concerning, for the further universalization of findings related to GR, that only a few studies attempted to reflect on the cultural and national situation of GT, counseling and healthcare provision (e.g., what is publicly funded or campaigned for) [cf. also Raz and Schicktanz (2009)]. There was a gap in the current literature on the ways that these discourses are shaped.

Our findings suggest that the socioempirical literature focused on the complexity of concrete practices and experiences of GT, whereas the philosophical and ethical discourses about GT focus more on the liberal challenge whether and when to intervene legally in the GT practice (Chadwick et al., 2014). The diversity of interpretations of the concept of responsibility in relation to GI restricted the opportunity to develop general sociological hypotheses about its psychological and social consequences. This was particularly obvious in a clinical genetics context, on which the majority of studies were focused. In almost all studies, the relationships of responsibility in genetic medicine held between genetic professionals, at-risk individuals, and their next of kin. These responsibilities concerned the management of hereditary diseases with different hereditary and clinical properties, but seemed rather unrelated to concerns about the medicalization of harmless genetic aberrations, the general genetization of society, or even genetic discrimination. This was highlighted not least by the fact that the concepts of GR for individual agency with regard to one's genetic make-up were only implicit or marginal in our text corpus.

CONCLUSION AND OUTLOOK

The focus of the qualitative studies in the examined social science literature was on the micropolitics of interpersonal relationships. State institutions, law givers, insurance companies, or even the healthcare system as a whole were regarded as the bearers of responsibility in only very few studies. This is striking because the claim of "responsibilization" of the individual in a neoliberal atmosphere is a common critique in more current sociological theories, but was only found to be indirectly approached in empirical studies (Arribas-Ayllon et al., 2011; Felt and Müller, 2011).

Moreover, the studies we reviewed indicated that the increasing availability of genetic tests for an increasing array of genetic conditions with ever more complex and multifactorial causes does not seem to have led to an increase in the individual burden of responsibility. This is primarily because the salience of GR in most socioempirical studies was restricted to the clinical context and did not shed light on non-clinical contexts such as direct-to-consumer applications.

As a result, the studies' general references to GR were rather pragmatic and undertheorized. This is remarkable if one takes into account that a significant part (two third) of the studies appeared in journals of medical sociology or medical ethics. However, they often helped to reveal communicative barriers and social or psychological issues in the management of GT practices or aimed to contribute to a solution to the pressing psychological, legal, and ethical conflicts, contributing to an improvement in the current situation. Therefore, it would be worthwhile for future studies to apply a more explicit, precise concept of GR to address and determine what exactly is at stake.

AUTHOR CONTRIBUTIONS

JL: literature search, summaries, detailed analysis, tables, article conception, and writing; MS: literature search, detailed

[10]For a recent discussion of this standard and its history, see also Louhiala and Launis (2013).

[11]This observation is not true for studies examining professional attitudes toward "prenatal testing" (Heyman and Henriksen, 2001; Bogner, 2005; Hashiloni-Dolev, 2007). However, as our search strategy focused on "genetic testing" we excluded studies mainly focusing on prenatal testing if "genetic testing" was not indicated in title, abstract, or keywords.

analysis, and writing. SS: study conception, detailed analysis, and writing.

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SUPPLEMENTARY MATERIAL

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