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How to support cancer genetics counselees in informing at-risk relatives? Lessons from a randomized controlled trial

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ABSTRACT

Objective: In hereditary and familial cancer, counselees are requested to inform their at-risk relatives. We developed an intervention to support counselees in this task.
Methods: A randomized controlled trial was conducted aimed at improving cancer genetic counselees' i) knowledge, ii) motivation to disclose information, and ii) self-efficacy in this regard. Eligible participants were randomized to telephonic counseling (n = 148), or standard care (n = 157) and assessed at baseline, 1 week post-intervention, and 4 months after study enrolment.
Results: No between-group differences were found in participants' knowledge, motivation, and self-efficacy. Knowledge concerning which second-degree relatives to inform was lower compared to first-degree relatives. About 60% of the participants was of the opinion that they needed to inform more relatives than stated in their summary letter and only about 50% were correctly aware of which information to disclose. Of note, at baseline, almost 80% of the participants had already correctly informed their at-risk relatives.
Conclusions: Since, unexpectedly, counselees already informed most of their relatives before the intervention was offered, efficacy of the intervention could not convincingly be determined. Counselees' knowledge about whom to inform about what is suboptimal.
Practice Implications: Future interventions should target a more homogeneous sample and address counselees' understanding and recall.

1. Introduction

In early-onset breast, ovarian or colorectal cancer or in families with multiple family members affected by these cancer types pedigree analysis and DNA testing can lead to a diagnosis of hereditary or familial cancer. This diagnosis implies a high or increased cancer risk for the index patient, i.e., the first in a family to request cancer genetic counseling, which then can lead to appropriate surveillance or preventive measures. In general, the diagnosis of an increased cancer risk also has implications for multiple at-risk relatives. In hereditary syndromes, the cancer risks are high and involve not only the nuclear family but also distant relatives. In familial cancer, risks are generally limited to close relatives. In both cases, family members at risk should be informed. Therefore, as part at of the genetic counseling process, counselees are requested to disclose the relevant information to their at-risk relatives [1–3].

However, in practice, counselees often do not inform their relatives or convey the information correctly [4–7]. Approximately 40% of counselees report experiencing family-related problems after the cancer genetic counseling process [8]. As a result, many relatives lack correct information and, thus, the opportunity to make a well-informed decision about dealing with their possibly heightened cancer risk [1,9].

Barriers that counselees may have related to informing their relatives include lack of knowledge, lack of motivation, and lack of self-efficacy [4,6,10,11]. Lack of knowledge regarding *which relatives* to inform may lead to not informing the right relatives, whereas lack of knowledge about *what information* to disclose may result in giving their relatives incorrect or insufficient information

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[11,12]. In addition, a counselee may lack the motivation to disclose the information. For example, the information may be considered too burdensome for a given relative, or the relative might be considered not mature enough to understand the information, or counselees want to protect themselves from (expected) negative reactions from relatives [13,14]. Moreover, a substantial proportion of counselees experiences burden informing relatives about a genetic condition [15]. Finally, counselees may feel unable to inform their relatives due to being insecure about their own ability to correctly disclose the information, i.e., a lack of self-efficacy. To overcome these barriers, counselees have reported a need for enhanced information and support [16,17] and they may benefit from professional backup [18,19].

Indeed, supporting counselees to effectively communicate risk information with their relatives is considered essential to obtaining the full benefits of genetic services [20]. Interventions developed to improve family communication about genetic testing include: the provision of enhanced information to counselees, giving them communication skills training and, more recently, the provision of telephonic counseling [21–26]. Effects were found on counselees' satisfaction with the process of family communication [26], but not on the information giving process itself. Only one non-randomized cohort study investigating an extensive intervention (providing counselees with a pedigree chart explicitly identifying relatives at-risk, a follow-up letter, and two consecutive telephone calls guiding counselees to inform their relatives) found an increase in uptake of genetic services by relatives in the intervention group versus the control group (61% vs. 36%) [21].

Our group developed an intervention to support counselees to inform at-risk relatives based on motivational interviewing (MI) [27] (see section 2.3 and Table 1 for more details). MI is a directive, client-centered counseling style for eliciting behavior change by helping clients to explore and resolve ambivalence [27]. In our case, ambivalence between feeling responsible to inform relatives while at the same time wishing to protect oneself and/or relatives form negative emotions or from feeling unable to correctly inform relatives. The principles of MI take into account the healthcare professionals' challenge of stimulating counselees to provide correct genetic cancer information to at-risk relatives, while at the same time respecting counselees' possible wish not to inform, i.e. their autonomy [28]. The intervention comprises a telephonic counseling session performed after counselees' have undergone the regular process of genetic counseling and testing. In this intervention trained psychosocial workers address issues regarding information disclosure and possible barriers experienced by the counselees. A pretest indicated the intervention to be feasible and appreciated by counselees [29].

In this multicenter randomized trial, we examined the efficacy of this additional telephone counseling intervention. Specifically, we hypothesized that, compared with a control group, counselees in the intervention group will have: i) improved knowledge of which relatives to inform, ii) improved knowledge on what information to disclose, iii) increased positive motivation, iv) decreased negative motivation, and v) increased self-efficacy.

2. Methods

2.1. Procedure

The study protocol was approved by the institutional review boards of the three participating hospitals. Full details of the trial design have been published elsewhere [30]. The original trial design also included data collection from participants' relatives to assess the implication of the intervention for their knowledge of their cancer risk and preventive measures, and their intention to engage in genetic counseling. However, as a result of different

Table 1
Content of training based on principles of Motivational Interviewing.*.

Phase	Step	
1	**1**	**Agenda setting** to introduce the subject of family communication about hereditary cancer risks without evoking resistance Skills: Asking for consent to discuss the issue of dissemination of risk information within the family Rolling with resistance
	2	**Exploring** counselees' current and planned pattern of informing relatives Skills: Systematically assessing *knowledge* about which relatives need to be informed, and which information needs to be conveyed (using pedigree and summary letter) Exploration of counselees' *motives and (possible) resistance* to inform relatives.
Verification of whether or not the counselee has informed all at-risk relatives. If so, the counseling session is ended, if not, the psychosocial worker proceeds to phase 2.		
2	**3**	**Providing additional or corrective information**, if needed. Skills: The "elicit-provide-elicit model"; first eliciting the person's understanding and information needs, then provide this information neutrally, followed by inviting the counselee to interpret the information.
	4	**Build motivation and strengthen self-efficacy** Skills: Inviting the counselee to speak out arguments in favor of informing relatives to reinforce these arguments and thus strengthen the counselees' motivation. Likewise, inviting counselees' to discuss strategies they deem feasible to inform relatives.
	5	**Discuss possible solutions** Skills: Brainstorm about possible solutions for their experienced barriers in informing at-risk relatives. To motivate and enlarge counselees self-efficacy, counselees are encouraged to list possible solution themselves.
To be avoided: Confronting; advising; over stressing the importance of informing relatives **Recommended:** respect counselees' autonomy; do not 'go faster' than the counselee; be aware that the counseling has not failed if the counselee makes a well-informed decision to not inform certain relatives		

* A more detailed description of the training is provided in [29].

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