Disclosure of research results to research participants: A pilot study of the needs and attitudes of adolescents and parents

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BACKGROUND: Researchers have a moral responsibility to offer to return research results to participants, but the needs and attitudes of parents and adolescents with cancer in paediatric oncology regarding the issue are relatively unknown.

OBJECTIVES: To explore the needs of potential research participants or their guardians with respect to the offer of a return of research results.

METHODS: A questionnaire was used in a focus group and in telephone interviews with eight adolescents and 12 parents of children with cancer. The participants were asked to respond to the questions and to comment on the inclusiveness of the questionnaire.

RESULTS: The majority of participants (18 of 20) wished to receive research results. Two somewhat unexpected findings are described. First, all participants in the present study felt that it was the primary responsibility of the participant to retain contact with the researchers for the purpose of obtaining research results. Second, few participants (n=2) indicated that the Internet would be a satisfactory way of transmitting these results. One-half of the participants wished to have face-to-face communication of results.

CONCLUSIONS: These results provide preliminary guidance for the return of research results to participants and validate the use of the questionnaire in a larger study of this issue.

Key Words: *Ethics; Focus group; Participants; Questionnaire; Research results*

Divulgation des résultats de recherches aux participants à la recherche : Un projet pilote des besoins et des attitudes des adolescents et des parents

HISTORIQUE : Les chercheurs ont la responsabilité morale d'offrir de transmettre les résultats de leurs recherches aux participants, mais les besoins et les attitudes des parents et des adolescents cancéreux en oncologie pédiatrique à ce sujet sont relativement inconnus.

OBJECTIFS : Explorer les besoins de participants potentiels à des recherches ou de leur tuteur à l'égard de l'offre de leur transmettre les résultats de ces recherches.

MÉTHODOLOGIE : Un questionnaire a été utilisé dans le cadre d'un groupe de travail et d'entrevues téléphoniques auprès de huit adolescents et 12 parents d'enfants cancéreux. Les participants ont été invités à répondre aux questions et à commenter l'inclusivité du questionnaire.

RÉSULTATS : La majorité des participants (18 sur 20) désiraient recevoir les résultats des recherches. Deux observations quelque peu inattendues sont décrites. D'abord, tous les participants à l'étude pensaient qu'ils étaient eux-mêmes responsables de maintenir le contact avec les chercheurs pour obtenir les résultats de l'étude. Ensuite, peu de participants (n=2) ont indiqué qu'Internet représenterait un moyen satisfaisant de transmettre ces résultats. La moitié des participants désiraient qu'on leur communique les résultats en personne.

CONCLUSIONS : Ces observations fournissent une orientation préliminaire à l'égard de la transmission des résultats de recherches aux participants et valident le recours à un questionnaire dans une étude plus vaste de cet enjeu.

The return of research results to participants has recently been increasingly recognized as a moral obligation of researchers based on the principle of respect for individuals (1-3). There are many potential benefits for the participants, including individual benefits such as the feeling of being acknowledged for one's contribution to science (1,4). There are also larger, global benefits, including diminishing a sense of secrecy by researchers and increasing public understanding of the impact of research. Despite the benefits described above, universal offerings of research results seldom exist (5). In part, this may be due to the logistics involved or it may be related to barriers perceived by investigators, such as possible harm to participants (6). Possible harms may include distress on hearing results that may increase the risk for late effects, distress about being on the inferior arm and distress for family members of research participants who have since died. There may also be harms associated with receiving results that are uncertain or evolving, or for which no screening is available.

Children with cancer are usually offered enrollment in clinical research trials. Upwards of 70% of children with cancer in North America participate in a trial sponsored by the Children's Oncology Group (COG). These research studies include clinical trials (phases I to III) lasting several

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months to several years; collection of biological specimens for banking and future investigation with potentially indefinite timelines and epidemiology; and long-term follow-up studies, which may last for decades. In a previous study (6), we surveyed all the principal investigators in the COG to ascertain their attitudes and practices with respect to the return of research results. While the majority of respondents supported the concept of developing guidelines for the mandatory return of research results, there was strong opposition by 27%. Written comments included concern about the ability to track participants and provide lay summaries, and the development of yet another regulatory hurdle.

There is little known about the needs and attitudes of research participants or their guardians (7). Partridge et al (8) demonstrated that adults participating in a phase II oncology trial wished to receive research results. However, they did not explore issues such as what to do with potentially disturbing information, the issue of informed consent before receiving this information, or how best to retain contact and communicate with participants. Schultz et al (9) published a study that provides further information regarding the return of research results. Of note, while most individuals wished the return of research results, a significant proportion described a high degree of fear, anxiety or sadness.

Because minimal information exists with respect to the needs of research participants in general, and paediatric oncology in specific (4), we conducted a pilot study to develop and validate a questionnaire in this population. We intend to corroborate the findings reported here with a larger, multi-institutional project. This information can then be used to guide researchers about the needs of participants with respect to the important issue of disclosure of research results.

METHODS

All research participants gave written, informed consent, and the study was approved by the Research Ethics Board of the IWK Health Centre before its start. Participants were recruited by word of mouth or poster presentations. Inclusion criteria included being a paediatric oncology patient (age 12 years and older) or a parent of a paediatric oncology patient (of any age). Participants had to be able to speak English or have access to a translator. Participation was either through a focus group or a phone interview.

The questionnaire was developed after careful review of the English-language literature and incorporated important issues such as mode of return of results, desirability for return of results and need for follow-up contact. The questionnaire included a section on demographics and a section on the major identified themes relevant to the return of research results. The full questionnaire is available at <www.pulsus.com/home2.htm>.

A mixed focus group of parents and adolescents was conducted in July 2003. The focus group had two objectives: first, to discuss the question of the return of research results in a format that follows a fixed template and, second, to review in detail the draft of the questionnaire for readability, content and completeness.

The phone interviews, which used the same template as the focus group, also explored the issue of return of research results and the draft questionnaire. The focus group and telephone interviews were conducted by one investigator (ST). They were recorded, transcribed and analyzed for major themes and individual responses to questions within the questionnaire. Responses from the focus group and the telephone interviews were collapsed for the purpose of analysis. Descriptive statistics are used to report the results.

RESULTS

Eight participants completed the questionnaire in a focus group and 12 completed the questionnaire in telephone interviews for a total of 20 participants. Adolescents with cancer represented eight of the respondents. The median time between the diagnosis of cancer and the focus group or interview was one year (range 0.5 to 11 years). Fourteen participants were female and six were male. The age range of the adolescents was 12 to 18 years. The age range of the parents was 35 to 45 years. Eight parents recalled being part of a research study and three recalled providing consent for their child to be part of a COG study.

None of those who had participated in a research study had been offered the return of research results after study completion. When asked to describe whether they would wish a copy of the results of a research study in which they or their child had participated, 18 of 20 indicated that they would.

Satisfactory methods by which to return research results were described as by mail (n=12), face to face (n=5), in a group setting (n=2) and through the Internet (two adolescents). However, when asked how they personally would like to receive research results, a larger number (n=10) indicated that they would wish face-to-face contact. Most felt that the primary oncologist or a research nurse was the most appropriate person to provide these results. The desired timing for return of research results to participants varied. Of those who responded to this question, 10 participants wished the results to be shared when accrual was complete, four wanted the results only when the study had been published, and three wanted the results after data analysis but before publication.

When asked who should be responsible for retaining contact between the researcher and the research participant, all 20 indicated that they felt that the updating of contact information was the primary responsibility of the participant.

Most felt that the return of research results would be helpful (n=16), while a few felt that it would create both benefits and harms (n=3). One participant felt that it would only be harmful. Approximately one-half of the participants (six parents and five adolescents) wished to receive only a summary of results while the remainder (six parents and three adolescents) wanted more specific information.

The draft questionnaire was rated highly in terms of readability and completeness, and only minor wording changes were suggested.

DISCUSSION

Our study's adolescent patients and parents from paediatric oncology showed that these individuals (representing potential and actual participants in research) strongly wished to receive research results of studies in which they had participated. Despite the potential benefits and the compelling moral reason (1) to do so, none of our participants had been offered research results. A possible reason was that some were still on therapy, but even those who had completed study participation for several years had not received any results. The lack of a universal offer of results was expected because a very low rate was documented in two other COG studies (5,6) that examined consent forms and institutional practice. The strong desire of participants to receive results parallel the findings of Partridge et al (8), who described a high interest of adult oncology patients participating in a phase II study to receive research results.

In the present study, only two individuals expressed concern about the potential for adverse effects or events. This is of note because a study by Schulz et al (9) suggested that substantial harm (severe fear, anger or sadness) occurred in up to 25% of participants who received research results regarding risk of cancer following retinoblastoma; these findings by Schulz et al may even have been an underestimate. Several issues potentially biased the study by Schulz et al (9): there was a long variation in the time between provision of results and survey of response and an under-representation of those deemed at highest risk for bad news (and, thus, possible adverse effects), and the results document may not have been clear. We believe that potential adverse effects must be anticipated in designing return of research results programs.

One area of surprise was the response regarding ownership of responsibility to retain long-term contact. In a previous study (6), we examined the attitudes and needs of principal investigators within the COG. While, in general, there was support or strong support for the concept, a vocal minority of principal investigators raised concerns about feasibility, which was linked, at least in part, to the difficulty in retaining contact with participants. The present study suggests that the responsibility for this contact is willingly assumed by the participants, rather than falling to the researcher. While it remains incumbent on the researcher to describe the balance of benefits and harms of receiving results (and, thus, of retaining contact), our study shows that the issue of long-term contact is not likely to result in a substantial burden to researchers.

Another surprise is the relatively low frequency of request for receiving results through the Internet. As both respondents who supported Internet dissemination were adolescents, it may be a higher priority for this age group. The low frequency was a surprise because older individuals in other settings have demonstrated a reasonably high Internet usage as a means to obtain health care information (10). Also of interest is that although by mail was said to be a satisfactory way to receive results, most participants stated that they would prefer face-to-face contact. The findings should be confirmed by a larger study. If representative, these results will have substantial impact on how best to return results to patients who wish to receive them.

The limitations of our findings include the fact that this was a pilot study at one site. The number of participants is small, and participants were self-selected for participation. They may not have been representative of the larger paediatric oncology population. We plan to address these issues through a multicentre, multiethnic study with consecutive patient sampling to reduce potential bias. In addition, we used mixed methodologies to obtain our results. It is possible that respondents in the focus group were influenced by others and, therefore, did not express divergent opinions. Despite these limitations, confirmation in a different population by Partridge et al (8) is encouraging and indicates that the trend of patients indicating a desire to receive results is likely real.

CONCLUSIONS

Overall, this study emphasized the importance of offering research results as a sign of respect to the participants, and confirmed that participants really do want to be informed. As outlined previously, this may have many positive ramifications. Further work needs to be done to determine the optimum method to return results, retain contact and provide information that will not harm subjects but respect their role in the research process.

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