Return and Disclosure of Research Results: Parental Attitudes and Needs Over Time in Pediatric Oncology

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Objectives To explore parental attitudes regarding the return and disclosure of research findings in pediatric cancer trials over time.

Study design Two surveys were set up to evaluate the stability of parental attitudes. One survey was carried out among 581 parents whose child was diagnosed recently (response rate, 53.5%). A second, population-based survey was set up with a time interval of 4 years between first cancer diagnosis and survey in which 1465 parents were included (response rate, 55.1%).

Results Almost all surveyed parents stated a parental right to receive aggregate research results. Fifty-five percent of the parents who recently participated in trials and 62% of those asked after a multiyear time interval thought that disclosure of individual findings is in any case necessary ($P = .0034$). The respondents wanted to restrict the duty to disclose study results to the child according to their notion of the child's well-being, composed of child's maturity, impairment of the parent–child relationship, and the quality of the results.

Conclusions Attitudes of parents regarding the return of research findings change over time. Shortly after diagnosis, parents are mainly interested in aggregate findings. Interest in individual findings appeared to increase as more time elapsed between cancer diagnosis and survey. *(J Pediatr 2017;191:232-7).*

In recent ethical debates, returning aggregate and individual study results in a research context has been broadly discussed, particularly with adult participants in mind. The major questions considered are what results need to be returned, when is the best time to return them, and what is the best way to communicate them to research participants? In responses, reasoning is usually based on ethical principles, such as individual autonomy, respect for the patient, beneficence, and the acknowledgement that research cannot progress without the engagement of participants.[1-6] Most proponents of disclosure consider it a principal duty to return study results to research participants. Some authors have restricted this obligation to the communication of aggregate results,[7-11] whereas others want to realize the routine disclosure of individual research results.[12-16] However, many advocates of the latter want to return individual study results only if certain criteria are met, especially if they can be used to inform medical care.

In pediatrics, additional questions regarding the disclosure of research results arise that need further investigation. Some of these questions are based on the ethical principle of individual autonomy referring to the child and the parents. However, that concept must be balanced with other ethical principles, such as beneficence and not harming the child, and last but not least, the cost effectiveness of the whole procedure.[17] In the following study, we focus on 2 ethical questions concerning the inclusion of children in research: first, do parents as legal guardians have a right to be informed about research results referring to their child and, more specifically, do they have a right to decide what kind of information they want to receive about their child? Second, are parents more or less inclined to receive research results about their individual child at some point in time or may they select what kind of information they want to disclose?

Few empirical studies on parents' views of returning study results in pediatric clinical research exist; most work comes from the field of molecular genetics and genomics. In this context, parents often express a strong interest in receiving results, especially if they have a meaning for the individual.[18-21] It was the aim of our survey to examine parents' preferences and considerations concerning the return of study results that were generated in a pediatric oncology therapy optimization clinical trial in which their child participated in Germany. Our study is based on 2 surveys because we also wanted to assess, as time elapsed after the child's diagnosis, whether the attitudes and preferences of the surveyed parents changed.

Owing to the direct reference to therapy optimization in both surveys, we assumed that parents would be interested in receiving all the results of the study in which their child is participating, but that parents' interest in individual results are stable and higher than interest in aggregate findings. Our second hypothesis was that

GCCR German Childhood Cancer Registry

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parents want to base their decision on whether or not they disclose research results to their child on their own understanding of the child's well-being.

Methods

We carried out a standardized survey among 2 study samples. Both groups were parents whose children were diagnosed with a malignant disease or a central nervous system tumor defined in the International Classification of Childhood Cancer.[22] Almost all of these children were treated in a therapy-optimization clinical trial appropriate to the cancer diagnosis and only a few had been treated by a nontrial therapy protocol. The presented results are part of a larger survey of which a detailed description of the participants and other survey results regarding parental informed consent and child's assent have been published elsewhere.[23,24]

The 2 samples differ in terms of the time lag between the child's cancer diagnosis and study participation and the survey. The first study sample was approached in a rehabilitation clinic right after their child's study participation was completed. In Germany, after oncologic treatment, children receive rehabilitation that starts directly after hospitalization. About 70% of the patients stay about 4 weeks together with their parents and siblings in a rehabilitation clinic.[18] That survey was conducted from December 1, 2008, to November 31, 2009, in cooperation with 3 German rehabilitation clinics. In total, 581 questionnaires were distributed to the mail boxes of the families in the rehabilitation units and 311 of them were returned, all of which could be analyzed (response rate, 53.5%). In the following, we refer to this survey as the rehab-clinic survey.

A second survey was set up with a much longer time interval between first diagnosis and survey with the help of the German Childhood Cancer Registry (GCCR). As a national population-based registry, it aims to collect data on all cancer cases for children under 15 years of age (and since 2009, under 18 years of age) in Germany as reported by pediatric oncology units.[19] With the consent of parents or legal guardians, about 95% of all German children subject to these conditions are registered in the GCCR by name. In our survey, families with a child under the age of 15 who was first diagnosed with such a disease defined in the International Classification of Childhood Cancer-3 between January 1 and December 31, 2005, were eligible for inclusion. The survey was conducted from March 1 to July 15, 2009, about 4 years after the children were first diagnosed. Because nearly all German children diagnosed with childhood cancer are registered in the GCCR, most of the rehab-clinic survey had been registered as well. However, there is no intersection between the surveys as we did not receive questionnaires from parents of the rehab-clinic survey specifying 2005 as date of first diagnosis (otherwise, we would have excluded these cases).

A total of 1624 patients registered in the GCCR fulfilled the inclusion criteria. Children who had died were also included, with the exception of children who had died within 6 months before the planned contact. In addition, the hospitals that had treated the children were given the opportunity to exclude individual patients from the survey by request (eg, owing to anticipated emotional distress in the family). Few hospitals made use of this offer, and only 27 families were excluded. Some families had moved to an unknown address and could not be traced (n = 29). Finally, 1465 questionnaires were distributed successfully. If no response was received by 4 to 6 weeks, the GCCR sent a single written reminder. Of the questionnaires sent, 807 were returned and could be included in the survey (response rate, 55.1%). In the following, we refer to this sample as the registry survey.

Survey and Analysis

We asked the respondents whether parents should have access to aggregate and/or individual study findings resulting from the study in which their child took part. We further queried how they want to receive the requested results and how they evaluated the disclosure of study results from the parents to their child. The questionnaire was assessed by pediatricians and piloted at the Department of Pediatric Hematology and Oncology at the University Medical Centre Hamburg-Eppendorf. The surveys were approved by the ethics committee of the Medical Association of Hamburg, Germany.

Data were analyzed using SPSS 23.0 (IBM, Somers, New York). The results from the registry group and the rehab-clinic group were pooled and the calculated response percentages are based on the total number of responses to each question. Responses to survey questions are presented as descriptive statistics for each survey (registry and rehab-clinic). The groups were compared using the Pearson χ^2 test (2-tailed). *P* values for these analyses were considered significant if below the .05 level.

Results

In the rehab-clinic survey, all families staying in a rehabilitation clinic during the chosen time period were included. In contrast with the registry survey, we did not have basic clinical information about this sample. Instead, parents were asked about their child's diagnosis, date of first diagnosis, trial participation, and the child's age. The short time between first diagnosis and the date of the survey is the defining feature of the rehab-clinic sample. This defining feature was demonstrated by the fact that most parents (78.5%) of those who answered the question stated that the child was first diagnosed after the year 2007 (n = 228). According to the parents who gave an unambiguous response, 95.7% of the children participated in a clinical trial during treatment and 4.33% were treated outside a trial (n = 277).

The GCCR holds basic information of all patients to whom questionnaires were sent (such as International Classification of Childhood Cancer-3 diagnosis, age at diagnosis, residence at diagnosis, sex, relapse status, vital status at the time of survey, and clinical trial participation). We used these data to compare survey participants and nonparticipants of the registry sample. The multiple analyses showed no significant or