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Parents' experiences with large-scale sequencing for genetic predisposition in pediatric renal cancer: A qualitative study

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Abstract

Objective: In pediatric oncology, large-scale genetic sequencing contributes to the identification of cancer predisposition, which can facilitate surveillance and family counseling. Our qualitative study explores families' motives, knowledge, and views regarding germline genetic sequencing to improve future counseling and support.

Methods: Semi-structured interviews were conducted with parents of children with renal tumors participating in a national center, germline sequencing study. An inductive thematic analysis approach was used. Twenty nine parents participated, 17 mothers and 12 fathers. The median age of the affected children was 4 years.

Results: Parents were generally positive about sequencing and reported a combination of individual and altruistic motives to participate. Some families counseled about sequencing shortly after cancer diagnosis felt overwhelmed. Many parents had difficulties distinguishing between panel and exome-wide analysis. Families in which no predisposition was identified felt reassured. Most families did not experience distress after a predisposition was disclosed, although sometimes stress following disclosure of a predisposition added to pre-existing (cancer-related) stress.

Conclusions: Even though families reported positive experiences with germline genetic sequencing to detect cancer predisposition, timing of consent for sequencing as well as parents' understanding of genetic concepts can be further improved.

KEYWORDS
cancer, cancer predisposition, genetic counseling, informed consent, oncology, pediatric oncology, psycho-oncology

1 | INTRODUCTION

Germline genomic sequencing is becoming increasingly important in pediatric oncology to identify genetic predisposing factors. It is estimated that about one in 10 children with cancer have an underlying genetic predisposition.[1–3] In some pediatric tumors, including Wilms tumor, predisposing conditions are even more common, as demonstrated by a recently published study performed at our center, which identified (epi)genetic predisposing factors in 33% of all patients.[4] Notably, these (epi)genetic predisposing factors

are not always accompanied by a recognizable phenotype and can remain undetected in absence of molecular testing. Advancement of sequencing techniques enables testing of large numbers of patients for an extensive range of genes associated with childhood (renal) cancer predisposition. For clinicians, patients and families, this large-scale sequencing in pediatric oncology offers many new possibilities, including enhanced family counseling and early detection of tumors by surveillance.

At the same time, it has long been recognized that large-scale germline sequencing of minors may give rise to psychological, social, and ethical challenges.[5] These challenges give rise to a number of unresolved questions, including whether patients' parents have sufficient knowledge about genetics to make well-informed decisions[6–9]; how counseling can facilitate adequate decision-making[7,10]; whether sequencing can induce substantial distress[9]; and how potential distress can be prevented or alleviated.[9] Despite these challenges, the general public, has mostly positive views and expectations of large-scale sequencing.[11,12] Preliminary evidence from previous research suggests this also holds true for parents of pediatric cancer patients.[7,13] Yet, empirical literature on the views and actual lived experiences of families with large-scale germline sequencing remains scarce.[13,14] Therefore, there is a need to further explore this topic in pediatric oncology.

For this purpose, we interviewed parents about their experiences in the context of the WES-KidTs study, a nationwide germline sequencing study in an unselected cohort of children with renal tumors in the Netherlands. In this article, we report the outcomes of this qualitative interview study, thus providing insight into parents' views and experiences related to several stages of sequencing. We describe considerations, motivations, and concerns as experienced by parents during recruitment and consent. Subsequently, we shed light on how parents made decisions on sequencing, also taking into account their knowledge and understanding of genetic concepts and procedures. Finally, we will address how parents perceive disclosure of sequencing results and how these results impact their lives.

2 | METHODS

2.1 | Study population

This interview study was conducted in families recruited for the WES-KidTs study. A total of 80 families were recruited for this sequencing study of whom 57 consented and 23 declined. Below the relevant aspects of the WES-KidTs study will be illustrated; a detailed description of the methodology and results of the sequencing study have been published elsewhere.[4]

WES-KidTs used a two-step approach to analyze germline exome sequencing data. The first step consisted of a gene panel analysis of known pediatric renal cancer predisposition genes. If no predisposition was identified an exome-wide trio-analysis was performed to search for pathogenic variants in potential novel renal cancer predisposition genes. In this second step, incidental findings (a pathogenic variant causing other diseases than renal cancer) could be identified. During consent for WES-KidTs, parents made an upfront choice for either step 1 (gene panel only) or step 1 followed by step 2 (with step 2 only being performed if step 1 was negative).

A pathogenic or likely pathogenic variant in a known renal cancer predisposition gene was always disclosed. Incidental findings were communicated with the families if approved by a multidisciplinary committee. If parental DNA was sequenced, parents could opt for disclosure of incidental findings in their own exome. Families were informed that they may be contacted again after initial disclosure of individual results if future re-analysis reveals additional (previously undetected) pathogenic findings or if future scientific evidence indicates that certain variants are pathogenic. Parents could opt-out from this re-contacting option, which may take place long after the study closes. At the age of 16, children would be contacted to give them the option to learn about incidental findings predisposing to adult-onset conditions. Individual results were communicated by a clinical geneticist.

2.2 | Recruitment, sampling and informed consent for the interviews

The WES-KidTs sequencing study started in 2018; from February 2019 until July 2021, parents were briefly informed about the interview study as part of the consent or disclosure procedure of the sequencing study. Families were not eligible if their Dutch was insufficient and/or their treating physicians had objections to an interview (exclusion criteria).

To ensure a wide range of experiences was covered, purposive sampling was used.[15] Among other factors, purposive sampling was aimed at interviewing a sufficient number of parents who had received a positive sequencing result (i.e. cancer predisposition or incidental finding). Furthermore, we also purposively sampled parents who declined participation in the sequencing study to include their perspectives in the scope of this article.

2.3 | Interviews

If parents consented to the interview study, an appointment was made for the actual interview, either at the outpatient clinic or at a location at their convenience. After the Covid-19 pandemic (March 2020) had started, interviews were also conducted using a secure videoconferencing platform (Skype for Business).

During the semi-structured interviews, a guide based on literature and clinical experience was used to make sure all relevant topics where covered (Supporting Information S1).

One researcher (S.B.) conducted the interviews with one parent from each family. Sometimes, at the request of the interviewee, the other parent was also present in the room but was not actively

involved in the interview. The interviews lasted 42 min on average (range 16–75 min). Interviews were audio-recorded and transcribed verbatim.

2.4 | Data analysis

We used an inductive thematic approach to analyze the interview data.[16] This qualitative method has the advantage that it does not build on a predefined theoretical framework. Therefore, inductive thematic analysis is well-suited to explore the full range of parents' experiences through an open lens. First, the transcripts of six interviews were independently coded by two authors (±20% of the total number) (S.B., R.W.). These two authors discussed differences in coding until consensus was reached about the provisional codebook. The provisional codebook was then used by one author (S.B.) to analyze the subsequent interviews, and the coding of each consecutive interview was subsequently checked by another author (R.W.), upon which they (S.B., R.W.) discussed the differences in coding to further refine the codebook. Using an iterative strategy, previous interviews were recoded if changes to the codebook were made, going back and forth between the codebook and the underlying data. In line with the general standards for qualitative research, inclusion and data collection continued until data saturation was reached, that is, until additional interviews did not yield further changes to the codebook.[17]

All coding was performed using NVivo (QSR International Pty Ltd. Version 12, 2018).

The process resulted in an elaborate code tree, which generated a broad overview of the topics and content of what participants had discussed. The code tree was subsequently discussed by all authors to identify emerging themes. Provisional themes were discussed until consensus was reached.

The study was approved by the University Medical Centre Utrecht Institutional Review Board (MEC-18-033/M, 08-02-2019).

3 | RESULTS

We conducted 33 interviews with 29 parents of 29 different pediatric cancer patients; four parents were interviewed twice, both before and after they received the sequencing results.

The willingness to be interviewed was high; out of 33 families invited (27 who participated in the sequencing study and six who declined) only four families did not want to be interviewed (two participating families and two families who declined).

Sixteen families declined participation in the sequencing study during the inclusion period of the interview study, 10 of these families were not eligible for the interview study. Recruitment of the remaining six families resulted in four interviews. The experiences of the families that declined participation in the sequencing study are incorporated in the results below, unless otherwise stated (one paragraph focusses specifically on the findings in this group). An overview of characteristics of patients whose parents were interviewed as well as of the interviewed parents are provided in Table 1.

3.1 | Parents' considerations

3.1.1 | Motivation to participate

Most parents had more than one reason to participate, often both altruistic and individual motives.

> [We participated] for you [i.e., researchers], mainly so that we, well, you could get more information about the course of a certain disease. But also for us of course, to know whether it is hereditary.
>
> (mother family no. 16)

The majority stated helping others was at least one of the main reasons to participate. In almost half of the parents, this was actually the most important reason. Parents felt that they should contribute to the well-being of future patients, like previous families had contributed to the care for their child. In addition, several parents articulated they wanted to advance science in general. Altruistic tendencies appeared to be even stronger among parents who had already received their child's sequencing results (whether a predisposition was identified or not), several of whom explicitly stated possible benefits for their child did not play a role in their decisions.

For most parents, individual motives, that is, reasons geared toward their own families, also played an important role. The majority reported possible benefits for their child (the patient) as a motive to pursue sequencing. In addition, many mentioned potential benefits for siblings, themselves, or other family members. A small subset of the parents considered possible benefits for future grandchildren. When asked to specify the benefits, parents referred to early detection and treatment of disease and/or preventive strategies including lifestyle-adjustments and surveillance. Most parents could not give examples of what these strategies or adjustments exactly would be. In general, parents expressed the attitude that more knowledge about health and genetics is preferable to less knowledge. In addition, several parents stated curiosity to learn why their child had developed a tumor was one of the reasons to participate. Notably, all parents acknowledged that the WES-KidTs study was a research project that was not primarily aimed at gaining individual clinical benefits.

3.1.2 | Concerns

Parents rarely mentioned drawbacks of genetic testing. If they did, these drawbacks appeared to be relatively minor issues that had not

TABLE 1 Patient and interviewee characteristics

Patient characteristics	*N* = 29
Sex	
Males	10 (34%)
Females	19 (66%)
Age	
Median age	4.0 years (range 0–11)
Mean age	4.6 years (SD 3.1)
Tumor type	
Wilms tumor	27 (93%)
Non-Wilms renal tumor	2 (7%)
Treatment status at the time of the interview[a]	
During treatment	9 (31%)
After treatment	24 (69%) mean 1.7 years (range 0–4,1)
Decisions on sequencing participation and analysis	
Renal gene panel analysis only	3 (10%)
Exome-wide trio analysis	22 (76%)
Declined exome sequencing study	4 (14%)
Genetic results at time of the interview	
Pending	12 (36%)
Negative result (no predisposition identified)	10 (30%)
Incidental finding (predisposition not related to renal cancer)	3 (9%)
Positive result (cause for renal cancer identified)	4 (12%)
Characteristics of interviewed parents	
Sex	
Males	12 (41%)
Females	17 (59%)
Educational level[b]	
Low	0 (0%)
Middle	10 (34%)
High	19 (66%)
Nationality	
Dutch[c]	25 (86%)
Other European	4 (14%)

[a]Four families were interviewed twice, both before and after they had received the sequencing results.

[b]Educational level defined according to Statistics Netherlands (CBS, Centraal Bureau voor de Statistiek), 2016: low educational level = no education, primary school, lower secondary education; middle educational level = upper secondary education, pre-university education, intermediate vocational education; high educational level = higher vocational education, university.

[c]Including Caribbean Netherlands and double nationalities.

influenced their decision-making. When being directly queried about downsides to sequencing, parents typically stated drawbacks (if any) were outweighed by potential benefits.

The burden of sequencing was perceived as minimal. A minority indicated knowledge of having a genetic condition might induce stress. Nevertheless, parents thought the (potential) benefits of knowing the test result would be more important.

Two out of three families who opted for gene panel analysis only, stated that they declined exome-wide analysis because they could not cope with the pressure related to making that decision at the time of consent. All parents who participated expected to be able to handle stress resulting from a potential genetic predisposition.

> It may be tough now, but still, it is better to know you have something than not to know you have it
>
> (father family no. 11)

> It can never turn out negatively I think, there can only be positive sides to it
>
> (mother family no. 23)

Some parents raised concerns about privacy but stated they trusted the hospital to keep their data safe. When specifically asked whether they had thought about insurance implications, parents answered (future) insurability had not influenced their decision-making and was not an issue they were particularly concerned about.

3.1.3 | Families who did not participate in sequencing

Only four families who declined participation in the sequencing study were willing to be interviewed. The first family indicated that they experienced high levels of distress related to the child's cancer at the time of recruitment, and therefore wanted to avoid any additional stress. The second family was worried about the potential psychological impact of knowing about an increased risk for a disease. In a third family, religiously motivated reservations toward DNA technology in general and the expectation that genetic testing may lead to an excessive burden (e.g., additional hospital visits) made them decide to not participate. A fourth family did not participate because of privacy concerns. Three of the families who did not participate displayed doubts regarding their decision, for example, by speculating whether it was still possible to change their minds.

> I don't know if this is a wise decision [...] At this moment this is a good one, but it can really be that I think differently about that in six months, but yeah then the discussion is over
>
> (mother family no. 29)

3.2 | Recruitment and consent

Families were recruited for sequencing in different phases of treatment and follow-up. Some of the families that were approached within the first months after diagnosis indicated the timing was not optimal and that they had a lot on their minds at that time.

> For me a bit later would have been better, then I would understand it better and I would memorize it better. There is so much coming at you, you get phone calls from doctors and then something like this comes along and you think okay. [laughs]. It is okay, but for me, at that moment, it was too much. I didn't have enough headspace for it
>
> (mother family no. 22)

Most parents had quickly reached a decision on participation in the sequencing study. Some decided on the spot, while others discussed it shortly at home. Although most parents had briefly mentioned the study to at least one family member (most often the child's grandparents), they did not widely involve others in the decision-making. When asked how they felt about making decisions on behalf of their child, the typical reaction was that parents did not perceive this as problematic. Participants suggested such decisions on sequencing are in line with numerous other smaller and bigger choices that parents make. One parent stated it is difficult to make such a decision for a child and therefore consented to gene panel analysis only.

Some parents who opted for gene panel testing only indicated they might want to pursue wider analysis later. In other words, parents did not reject broad genetic analysis indefinitely or on principle. This is in line with the observations with respect to parents who declined sequencing overall (as described above).

3.3 | Receiving results

Seventeen parents were interviewed after they had received the sequencing results. All parents who were informed that a genetic predisposition had not been identified felt primarily relieved. A few parents reported mixed feelings because it did not answer the questions they had about the cause of their child's tumor. Parents often concluded that their child's tumor was most likely the result of bad luck. Several parents understood that not finding a genetic predisposition does not completely exclude its presence. These parents were aware of the possibility that re-analysis may reveal a previously undetected predisposition in the future. While these parents found the possibility of eventual re-contacting unsettling, they did not seem to be opposed to such future updates.

> Science keeps developing, so maybe in 5 years we will be called again: 'well [child] has ever had a tumor. We've thought of something new, are we allowed to look again.' So then it's never finished, is it?
>
> (father family no. 21)

The parents were not explicitly inquired about their views regarding the policy to postpone disclosure of incidental findings related to adult-onset diseases until the child is (legally) able to provide consent at the age of 16. Among the few parents who raised this topic themselves, only one expressed discontent at this policy; this mother articulated that she felt somewhat uncomfortable about the idea that the researchers withheld information from her.

The predisposing conditions and incidental findings that were identified were diverse, ranging from well-established and highly penetrant pathogenic variants to recently discovered or low-penetrance variants. The parents of patients with an identified germline pathogenic variant, generally reflected optimistically on this particular outcome. They focused primarily on the advantages of having this knowledge, for example, by pointing towards opportunities for surveillance or for the use of reproductive technologies such as pre-implantation genetic diagnostics. Parents acknowledged that having a genetic predisposition to childhood cancer does not necessarily lead to actual malignancy; they stressed that bad luck was still a major factor. The test result itself was not perceived as particularly burdensome by most parents. Yet, two parents who described that they already felt stressed before sequencing said that the sequencing process and its result added to their distress. None of the parents articulated feelings of guilt.

> It is an accumulation of situations […] the information from this study is like an extra brick in your backpack
>
> (father family no. 25)

All parents of school-aged children had briefly mentioned the results to their child, but this had not led to elaborate conversations about the topic of genetic susceptibility.

> [we told her] that it was not hereditary and that her sister did not have to get tested for all that kind of stuff. She knows that, yes, yes […] It's funny because those children, this is not interesting to them. They think 'oh okay' and then continue playing.
>
> (father family no. 20)

Almost half of the parents of the families in which no predisposition was identified shared their test results with family members. Families in which a predisposition was identified that could have consequences for other family members all shared this within the family.

> [The clinical geneticist] called while I was on the road […] Of course I first called [my wife]. I think I waited for one, or one and a half hour, and then we just shared it in the family WhatsApp group.
>
> (father family no. 13)

3.4 | Knowledge

Most parents did not search for any additional information about genomic sequencing. They expressed a high level of trust in the information provided by the hospital and typically described this information as clear and sufficient. Nevertheless, during the interviews it became apparent that parents faced difficulties grasping genetic concepts and information. Throughout virtually all interviews, various misconceptions were encountered. These misconceptions ranged from mixing up the concepts of germline and tumor DNA to stating that their child's disease could not possibly be hereditary because initial diagnostic testing was negative.

The differences between panel and exome-wide analysis proved especially challenging for parents to understand, at least in retrospect, as almost all interviewed parents struggled to explain the difference between the two approaches. Several parents could not recall that they had a choice between cancer panel only and extending to exome-wide analysis. Moreover, many of the other parents had difficulties discerning between the implications of both options regarding incidental findings.

> Yeah, I didn't know we had a choice. I thought it was standard, that if they couldn't find anything in [child], they would then switch to sorting out our DNA
>
> (mother family no. 12)

4 | DISCUSSION

This qualitative study examined the experiences of parents of children with renal tumors regarding their considerations in decision making, knowledge, and views on (obtaining results of) germline genetic sequencing in their child. The overall impression from the interviews is that parents are very motivated to participate in this germline sequencing study. Parents made their decision for sequencing quickly and with conviction, generally describing few hesitations or concerns about potential drawbacks. Our study shows that parents are typically motivated by a mix of individual reasons (i.e., learning about or promoting their children's or their families' health) and altruistic reasons (i.e., aiding future patients or medical science). Consistent with previous literature, the principal attitude toward genomic sequencing observed in this study is that learning more about their children's health is considered better than knowing less.[11,13,18]

Our study identified religious beliefs, privacy concerns and feeling overwhelmed as reasons not to participate, in line with previous studies.[19–21] What our study adds is that the decision to decline (exome-wide) sequencing seems temporary rather than indefinite in the sense that this choice is contingent on specific circumstances at the time of consent. Over time, as a family's situation changes, they might want to change their decision on sequencing accordingly.

The results of our study also draw attention to a number of (partly) unresolved issues and challenges. One of these issues concerns the optimal timing of germline sequencing to detect an underlying predisposition. Upfront routine germline sequencing as part of the cancer diagnostic process is gaining support in the literature.[22–24] Yet, in our study sample, several parents who were informed about the sequencing study shortly after diagnosis felt overwhelmed by the rapid cascade of events and choices. This suggests that genetic counseling would ideally take place after treatment has been initiated, which is also supported by studies into patients' and parents' preferences.[7,25]

Despite the generally positive attitudes, the psychosocial burden of germline sequencing continues to warrant careful consideration in pediatric oncology. In this study, some parents indicated that deciding on testing, thinking of possible outcomes and pondering potential re-contacting induced feelings of stress and worry. This is in line with other qualitative studies, that have reported positive overall experiences but also sequencing-related anxiety and worry in at least a subset of participants.[26,27] Reviews of quantitative adult and childhood cancer studies suggests long-term psychological impact of germline genetic testing is limited or even negligible.[28,29] A possible explanation for this discrepancy is that questionnaires used in these studies are not appropriate to capture the impact caused by the worries families have in this particular context.[28,30] Finally, most quantitative research on the psychosocial impact focusses on the consequences of learning about a genetic test result, while our study highlights that deciding whether to take the test can also be stressful for at-least a subset of families. This underlines the need for more empirical evidence on the psychosocial impact (including quality of life outcomes) of sequencing in pediatric oncology.[31,32]

4.1 | Study limitations

This study has some limitations. We were able to interview only four families who declined genomic sequencing, during the inclusion period of the interview study 16 families declined sequencing, however 10 families were not eligible based on the exclusion criteria. The study sample predominantly consisted of highly educated parents. In addition, because of our focus on children with renal tumors, who are often young (median age 4 years) and have a relatively favorable prognosis, future research in other tumor types is needed to assess to what extent our findings are generalizable.

4.2 | Clinical implications

Our study shows that parents generally hold favorable views towards large-scale sequencing and reflect positively on their own experiences regarding sequencing. Hence, parents appear to be supportive of wider translation of genetic sequencing to daily clinical practice in pediatric oncology. Nevertheless, our study also highlights the need to further improve counseling strategies. During the interviews, misconceptions about genetics were frequently encountered. This finding is consistent with previous studies assessing genetic knowledge and underscores the urgency of promoting genetic literacy

among patients and families.[6–9] Furthermore, professionals should ensure that parents are mindful of psychosocial implications of sequencing further down the road, especially for the subset of parents who already face considerable levels of (cancer-related) distress. To avoid confusion regarding different approaches to analyzing large-scale genetic datasets, counseling for gene panel-analysis should preferably be separated from counseling for more extensive (e.g., exome or genome-wide) analyses. Finally, to optimally support families in making well-deliberated decisions, counseling should ideally occur at a relatively stable phase of treatment.

4.3 | Conclusions

Most parents were very motivated to participate in germline genetic sequencing. Considering the extensive literature on to the drawbacks of germline genetic sequencing in children, it is remarkable that parents identified only a few disadvantages. Our study did identify several challenges, such as timing of consent and knowledge of genetic concepts. Furthermore, it suggests that the sequencing process can have an impact on families even if a predisposition is not identified. Further research into the experience of families with older children affected by cancer and children affected by other types of cancer is warranted.

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CONFLICT OF INTEREST

All authors declare to have no competing interests.

DATA AVAILABILITY STATEMENT

Individual data cannot be shared due to Institutional Review Board restrictions.

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SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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