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REVIEW ARTICLE

Narrative review on ethical and psychological issues raised by genetic and genomic testing in pediatric oncology care

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Abstract

In pediatric oncology, genetic and genomic tests are proposed throughout the care pathway for many reasons (e.g., cancer characterization, identification of the most appropriate treatment, patient selection for clinical trials, identification of tissue/organ donors, or risk of relapse prediction). Despite the many different approaches (somatic or germline testing, targeted gene or genome sequencing), the implicated individuals are confronted with situations that may intersect and that are interesting to compare. No study has identified and analyzed the available works on these new practices in pediatric oncology. The aim of this narrative literature review was to describe the ethical and psychological perspectives of children with cancer, parents, and healthcare professionals when genetic or genomic testing is proposed as part of the cancer management. Eighteen articles met the inclusion criteria and were comprehensively coded using MAXQDA. Their analysis showed that concerning the subjective implications of genetic and genomic testing, the areas of ambivalence (desire of treatment, desire for knowledge, uncertainty, and guilt) reported by patients and their parents seem to mirror the healthcare professionals' concerns. The ethical and psychological issues about predisposition testing, long discussed in the context of hereditary retinoblastoma and Li-Fraumeni syndrome, represent a useful starting point for a wider discussion of a genetic and genomic testing pathway in pediatric oncology more broadly.

KEYWORDS

consent, ethics, genome sequencing, information, parents, pediatric oncology, psychology

1 | INTRODUCTION

Recent research showed that 10%–15% of children with cancer carry cancer predisposition genes (ICGC PedBrain-Seq Project et al., 2018; Kratz et al., 2021). Since the discovery of the first retinoblastoma predisposition gene by Knudson, 1971, clinical practice has

progressively incorporated the search for predisposition genes as and when they are discovered (Knudson, 1971). Such testing is now integrated into the childhood cancer care pathway to identify predisposition genes (targeted search, gene panel, or genome sequencing). Genetic testing is useful to characterize the patient's cancer type, to propose the most appropriate treatment, or to select

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patients for clinical trials. The currently available sequencing technology makes it possible to identify somatic and also germline variants, with potentially broader implications for the patient's family. Moreover, it may lead to the emergence of data that are not directly related to the initial question, such as incidental findings and secondary findings,¹ and variants of unknown significance (de Biomédecine, 2020; Kratz et al., 2021). These findings may provide relevant information for the present, future (such as actionable variants² in relation to the childhood cancer), or distant future (such as familial predisposition or risk of recurrence). Therefore, healthcare professionals must decide whether to disclose such information or not, and parents whether they want to receive it or not (Bertier et al., 2017; Delanne et al., 2019; Isidor et al., 2019; Pujol et al., 2018).

Regardless of the disease, many studies have shown that genetic testing is associated with psychological and ethical issues for children and their parents, as well as for the involved healthcare professionals (Bertier et al., 2017; Botkin et al., 2015; Chassagne et al., 2019; Clayton, 2015; Houdayer et al., 2019; Wade et al., 2013). Several guidelines have been published to offer guidance on genomic testing in adults (the content of the information and consent form, and incorporating genetic consultations and psychological support in the care pathway for somatic or germline testing), but they are poorly adapted to children and adolescents (Botkin et al., 2015; Matthijs et al., 2016; van El et al., 2013). Similarly, in the context of cancer, there is extensive literature on the ethical and psychological issues associated with genetic/genomic testing in adults, whereas data are more limited for children. It is important to stress that genetic testing is generally proposed at a particularly sensitive time when parents and children are already in distress following a cancer diagnosis. (Droin-Mollard et al., 2021). Due to this specific context, children and parents may find it difficult, due to mental overload, to take into account new information. Therefore, the decision of whether or not to undergo genetic testing is made in a background of fear of death. This fear may take a more or less conscious form and can be psychologically overwhelming for children and their parents, thus blinding them to the importance of some decisions. Moreover, childhood cancer is associated with a high degree of uncertainty about its causes, particularly for parents whom may be combined with feelings of guilt and the desire to find an explanation. It is then difficult to make rapid decisions on complex issues, such as when somatic testing or predisposition testing is proposed.

To the best of our knowledge, no study has analyzed the available literature on the ethical and psychological issues of genetic/genomic testing in pediatric oncology. Therefore, we carried out a

¹By incidental findings, we mean a "pathogenic variant with no direct relationship to the initial indication and discovered by chance". By secondary findings, we mean "a pathogenic variant with no direct relationship to the initial indication, but actively sought through the analysis of a pre-defined gene list" (Agence de Biomédecine, 2020). "Secondary findings differ from incidental findings because they are actively sought and systematically evaluated using a list of genes selected based on guidelines developed by professional societies in various jurisdictions" (Isidor et al., 2019). [The first quotation is our translation from French.]

²The definition and use of the concept of actionable variant are the subject of debate at the national and international level.

What is known about this topic

Genetic/genomic testing is increasingly integrated into the pediatric cancer care pathway to identify predisposition genes, characterize the cancer type, determine the most appropriate treatment, or select patients for clinical trials. Yet, no study has analyzed the available literature on the ethical and psychological issues of genetic/genomic testing in pediatric oncology.

What this paper adds to the topic

This narrative review shows that in the context of pediatric cancer, healthcare professionals need to be better trained on the temporal and psychological issues encountered by patients and parents involved in genetic and genomic testing. Ethical benchmarks must be established to better support children with cancer and their parents during the genetic/genomic testing pathway throughout the care process.

narrative review of the literature on the perspectives of children with cancer, parents, and/or healthcare professionals to bring insights into the complex issues raised by genetic/genomic testing as part of the childhood cancer care pathway. This review allowed us to identify important psychological and ethical issues about these tests and about the timing, the information, and the support needed by children with cancer and their parents.

2 | METHODS

This narrative review of the literature follows the guidelines proposed by Green et al. (2006). Compared with systematic reviews or a scoping review (Munn et al., 2018; Petticrew & Roberts, 2006), the choice to conduct a narrative review was motivated by the need for qualitative interpretive richness to highlight complex and innovative research subjects by challenging the existing literature and by relying on the literature to develop new research projects: it could be qualified as a hermeneutic narrative review a process of creating an interpretative understanding of an emerging topic (Breslin & Gatrell, 2023; Green et al., 2006; Greenhalgh et al., 2018). To identify relevant articles, two databases were searched: PubMed and Cairn (a search engine for humanities and social sciences), followed by snowballing to find additional references in the bibliography of the identified articles. This broad search strategy was used to identify articles in a specialist field for which initial keyword-based searches produced limited results. The four authors (KLL, MDM, SDM, LH) worked together to define the relevant keywords in French and English to be included in the search equations. Searches were performed using Medical Subject

Headings (MeSH), combined with non-MeSH descriptors (for example, next-generation sequencing [NGS] is not a MeSH descriptor). The complete search string in English is in [Table 1](#).

Searches were performed using 2006 as the lower publication date limit (Strahm & Malkin, 2006; i.e., the year when genetic testing started to be routinely proposed for pediatric cancers) and May 2021 as the upper limit. The PubMed database was used for articles in English and the Cairn database for articles in French. This strategy identified 703 references in PubMed and 21 in Cairn. Moreover, 13 works were identified by snowballing. After reading the titles and abstracts to confirm that psychological and/or ethical

and/or sociological issues in pediatric oncology were the subject of the study, 318 articles in PubMed and 20 articles in Cairn were included in the review as well as the 13 “snowball” articles. They were imported into the bibliographic management tool Zotero. This work was performed by two researchers involved in the project (LH, MDM) who jointly discussed to ensure the relevance of the retained articles. During this discussion, they agreed on the final selection.

Then, these two researchers (LH, MDM) independently screened the retained articles using four inclusion criteria: articles written in French or English, inclusion of a description of the methodology, original research, and involving a child in the index case. Articles that were translations of already identified original articles, that did not concern pediatric oncology, or that were literature reviews, editorials, expert commentaries, or clinical cases were excluded from the literature review. However, some of them were read to develop the Background and Discussion sections. After removing duplicate records, the corpus consisted of 18 articles on the ethical, psychological, and/or sociological issues of genetic testing in the context of pediatric cancer. [Figure 1](#) summarizes the search process and includes studies.

The narrative analysis was performed by encoding each article inductively and comprehensively. Thanks to MAXQDA2020 software, this thematic analysis enabled us to develop a codebook (Campbell et al., 2021; Paillé & Mucchielli, 2012). The coding of each

TABLE 1 List of the complete search used using the words “AND” or “OR.”

oncology [All Fields] OR neoplasms [MeSH Terms] and [All Fields] OR oncogenetic [All Fields] AND next generation sequencing [All Fields] OR high-throughput nucleotide sequencing [MeSH Terms] OR genetic testing [All Fields] and [MeSH Terms] AND child [MeSH Terms] and [All Fields] OR children [All Fields] OR minors [MeSH Terms] and [All Fields] OR AYA [All Fields] OR young adult [MeSH Terms] and [All Fields] OR adolescent [MeSH Terms] and ethics [MeSH Terms] and [All Fields] OR ethical issues [All Fields] OR psychology [MeSH Terms] OR sociological issues [All Fields] OR psychological factors [All Fields] OR psychological impact [All Fields] OR incidental findings [MeSH Terms] and [All Fields] OR unsolicited findings [All Fields]

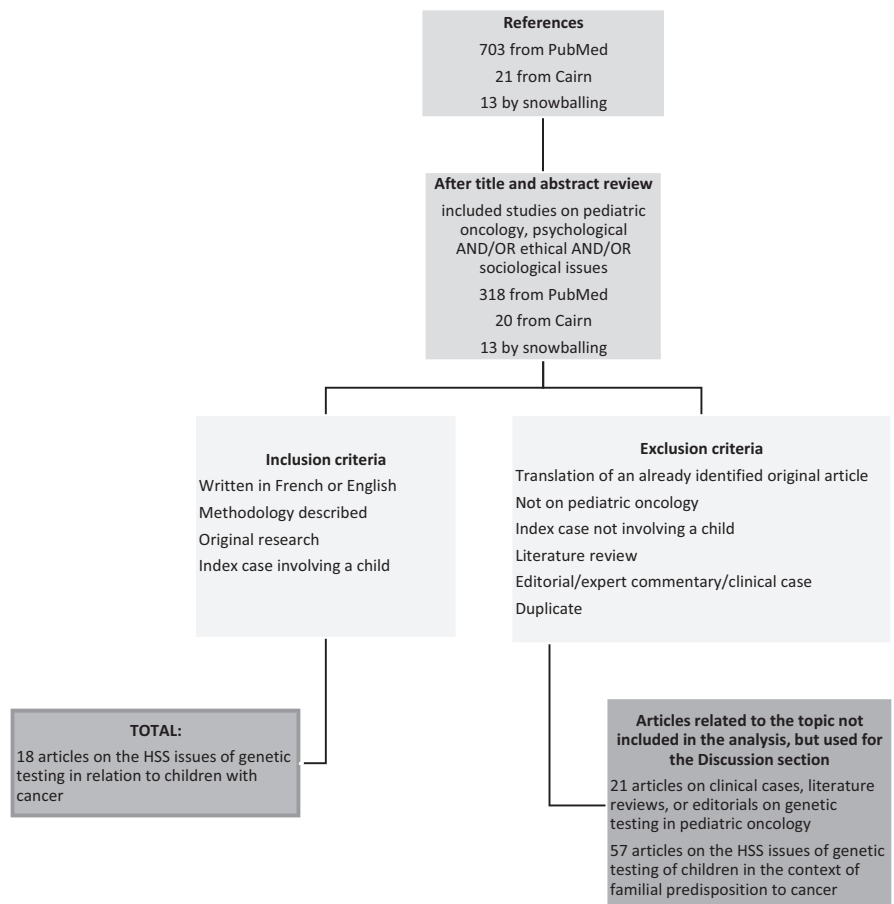


FIGURE 1 Flow diagram of the search strategy.

article and the codebook were refined iteratively through discussion by the four authors (SDM, LH, MDM, KLL; the codebook is accessible in Data S1).

This narrative review focused on the psychological, sociological, and ethical issues of genetic/genomic testing in children with cancer, and not on the medical issues (e.g., relevance and indication of genetic testing) that were also discussed in the included articles.

3 | RESULTS

3.1 | Description of the articles included in the review

The first authors of the 18 articles included in the review were from a wide variety of research fields ($n=11$ from humanities and social sciences). The authors used different methodologies: quantitative methods ($n=4$), qualitative methods ($n=8$), and mixed methods ($n=7$). Most studies were performed in the authors' country of residence (8 USA, 4 Canada, 1 France, 1 Germany, 1 United Kingdom, and 3 Australia), with the exception of one study carried out in Kenya, but with a Canadian lead author. Articles most often explored the parents' perspective (15/18), followed by the children's perspective (7/18) and the healthcare professionals' perspective (3/18). Eight articles focused on the possibility of discovering incidental or secondary findings. Table 2 presents an overview of the studies included in the narrative review.

3.2 | Thematic analysis

3.2.1 | Defining the right role for the child: A shared concern for healthcare professionals, parents, and children

Genetic testing in pediatric settings raises specific questions about the child's right role, with a tension between their vulnerability, interests, and desire for autonomy (Forbes Shepherd et al., 2019). This tension was expressed by parents, who wanted their children to play a greater role in the return of genetic/genomic test results as they became older (McGill et al., 2019). Healthcare professionals also found it difficult to define the roles of children and parents. To protect the child's interests, they cannot rely only on parental responsibility because the decisions made by parents may be well-meaning, but they can have unintended consequences and result in revelations that will affect the patient as an adult (Esquivel-Sada et al., 2017). The situation is particularly complex for adolescents and young adults because adolescence is a period of transition between autonomy and dependence (Forbes Shepherd et al., 2019; Weber et al., 2019). Healthcare professionals are aware of the need to include patients in the decision-making process, but in practice, patients are sometimes absent from such discussions or made invisible when present. For instance, Claret et al. showed

that the consultation loses the protective function it could provide if the child is forgotten and they raised the question of the child's place in the consultation, as done also by Weber et al. (Claret et al., 2018; Weber et al., 2019).

In response to these challenges and by taking into account the parents' expectations, the best practices identified in the included articles emphasize the importance of involving the children in the process and taking their age and emotional maturity into consideration. It is also crucial to determine the adolescents' understanding of genetic issues in relation to themselves and their close family, and their level of involvement (active or less active) in the decision-making process (Forbes Shepherd et al., 2019). However, it can be challenging to thoroughly adolescent comprehension and decision-making capability. These authors also suggested that once adolescents are sufficiently mature, their decisions should be given priority over the parental wishes, for example, by postponing predisposition testing if they are not ready to take part in the decision-making process. This is consistent with the position statement published by the Human Genetics Society of Australasia in 2020 (Vears et al., 2020).

Independently of the children's age, some authors proposed "a child-led approach" in which age-appropriate language is used, tailored to the patient's maturity level (Mandrell et al., 2021; McGill et al., 2019). This includes an appropriate choice of words and the use of "childlike terms" (Hill et al., 2018; McGill et al., 2019).

3.2.2 | Bringing up genetic testing at the right time in the care pathway and repeatedly

The family's emotional status is not always correlated to the time elapsed since diagnosis (Howard Sharp et al., 2020). Healthcare professionals should present genetic information in such a way that families "[are] given sufficient time to process the information, especially in the context of their emotional state, before making a decision." At least 1 h should be planned for a genetic testing consent consultation (Scollon et al., 2014). The authors of three articles (Brozou et al., 2018; Howard Sharp et al., 2020; Johnson et al., 2019) considered the preliminary stage of obtaining consent as a time when many questions can arise, and therefore, they proposed a two-stage consent process. Some authors suggested providing information about genetic testing earlier in the care pathway, at different stages, and repeating such information over time. This should allow healthcare providers to adapt the information and decision-making process to the emotional availability of patients and parents (Brozou et al., 2018; Claret et al., 2018; Hill et al., 2018). For instance, Claret et al. observed that in consultations with children, parents did not allow themselves to have a critical view on the healthcare professional's recommendations. Therefore, they tested a two-stage approach for oncogenetic consultations: an initial parent/physician consultation followed by a parent/physician/child consultation (Claret et al., 2018), as first described for presymptomatic genetic testing by Lahlou-Laforêt (Lahlou-Laforêt et al., 2012). This two-stage approach might allow

TABLE 2 Overview of the studies included in the narrative review.

First author and date	Methodology	Perspective of parents, children or health professionals	Germline and/or somatic testing Direct testing, panel, or genome sequencing IF, SF or VUS considered	1st author discipline & work location
Brozou et al. (2018)	Quantitative N = 94 0-18-year-old children	Parents	Germline Genome sequencing IF and VUS in the consent form	Pediatrics, Germany
Claret et al. (2018)	Qualitative Clinical observations of consultations with 3-12-year-old children	Parents and child	Germline testing	Psychology, France
Desrosiers et al. (2019)	Mixed methods N = 42 parents of 0-18-year-old children <i>Hypothetical proposition</i> Statistical and thematic analysis	Parents	Germline & somatic genome sequencing IF	Genetic counseling, United States
Esquivel-Sada et al. (2017)	Qualitative N = 11 parents, semi-structured interviews with 4 IF hypothetical scenarios. Thematic and phenomenological analysis No information on children age	Parents	Germline & somatic genome sequencing IF: main focus of the research, hypothetical scenarios	Sociology, Canada
Forbes Shepherd et al. (2019)	Mixed methods Questionnaire: quantitative and qualitative data (hypothetical clinical scenarios with 15-year-old children) N = 43 Statistical and thematic analysis	Healthcare professionals	Germline testing for Li-Fraumeni syndrome	Psychology, Australia
Foster et al. (2017)	Quantitative N = 292 patients Questionnaire completed by >16-year-old patients or parents No information on children age	Children, parents, adults	Germline testing for retinoblastoma	Genetics, UK
Gedleh et al. (2018)	Mixed methods Qualitative: 31 individuals across 5 focus groups, thematic analysis Quantitative: questionnaires No information on children age	Adult survivors of pediatric retinoblastoma or parents of children with retinoblastoma	Germline testing for retinoblastoma	Anthropology, Canada
Hill et al. (2018)	Qualitative, focus group: adult survivors and parents of children with retinoblastoma N = 15 No information on children age	Adult survivors of pediatric retinoblastoma or parents of children with retinoblastoma	Germline testing for retinoblastoma	Molecular genetics, Canada
Howard Sharp et al. (2020)	Mixed methods N = 53 families who declined genome sequencing during clinical care. Statistical and thematic analysis 0-21-year-old children	Parents	Germline & somatic genome sequencing IF, SF, VUS included in the consent and information to families	Psychology, United States
Johnson et al. (2017)	Mixed methods N = 52 pediatric hematologists/oncologists Questionnaires No information on children age	Healthcare professionals	Germline & somatic genome sequencing VUS	Pediatric oncology and bioethics, United States

(Continues)

TABLE 2 (Continued)

First author and date	Methodology	Perspective of parents, children or health professionals	Germine and/or somatic testing Direct testing, panel, or genome sequencing IF, SF or VUS considered	1st author discipline & work location
Johnson et al. (2019)	Quantitative N = 121 Statistical analysis No information on children age	Parents	Germine & somatic genome sequencing	Pediatric oncology and bioethics, United States
Malek et al. (2017)	Qualitative, interviews longitudinal n = 33	Parents	Germine & somatic genome sequencing IF, VUS	Medical ethics and Health policy, United States
Mandrell et al. (2021)	Qualitative, Parents n = 43	Parents	Germine & somatic genome sequencing IF	Pediatric nurse research, United States
McCullough et al. (2016)	Qualitative, longitudinal Thematic interviews: 16 oncologists and 40 parents, before and after receiving the results No information on children age	Parents and health professionals	Germine & somatic genome sequencing IF: <i>central to the article</i>	Medical ethics, United States
McGill et al. (2019)	Mixed methods Qualitative: 35 semi-structured interviews, thematic analysis Quantitative: questionnaires 3–18-year-old children	Adult survivors (>16 years of age) or parents of children with cancer	Germine testing	Psychology, Australia
Scollon et al. (2014)	Quantitative N = 121 families 0–17-year-old children	Parents	Germine & somatic genome sequencing IF, SF, VUS in consent form	Genetic counseling, United States
Vetsch et al. (2020)	Mixed methods N = 622 questionnaires N = 87 semi-structured interviews 0–16 years at diagnosis + >5 years post-diagnosis	Adult survivors (>16 years of age) or parents of children with cancer	Germine testing	Health sciences and health policy, Australia
Weber et al. (2019)	Qualitative N = 7 semi-structured interviews	Adolescents with a pathogenic variant of a tumor predisposition gene	Germine testing	Genetic counseling, Canada
Total number of studies = 18	4 quantitative 7 qualitative 7 mixed methods	Parents: 15/18 Child: 7/18 Healthcare professionals: 3/18	Germine testing: 10 Germine & somatic testing: 9 Genome sequencing: 9 <i>IF, SF or VUS: 8 articles</i>	11 from Humanities and social sciences

Note: IF: by incidental findings, we mean a pathogenic variant with no direct relationship to the initial indication and discovered by chance. SF: by secondary findings, we mean a pathogenic variant with no direct relationship to the initial indication actively sought through analysis of a pre-defined gene list. In the second column, the age of the children included in each study is indicated in bold. In the 4th column in normal typing it is precised the category of genetic testing between somatic and germline, in bold we precise the genetic approach (direct testing, panel or genome sequencing), and in italic if the paper deals with IF, SF or VUS.

a better respecting the needs of all family members in terms of timing, expectations, and their role. Hill et al. recommend scheduling follow-up meetings during and after treatment to allow families to ask questions and incorporate feedback (Hill et al., 2018). Moreover, children with cancer have little understanding of the risk and treatment history. Genetic/genomic testing could have a major impact on their life decisions, for example, in relation to reproductive choices. Therefore, it is important to provide gradual, repeated, and personalized counseling to pediatric patients as they grow to give them adequate and age-relevant information to improve their adult life (Desrosiers et al., 2019; Foster et al., 2017; McGill et al., 2019; Vetsch et al., 2020; Weber et al., 2019).

3.2.3 | Supporting decision-making by considering the family context

The “baggage” brought by the families to the consultation influences their response to the offer of genetic testing. This baggage consists of various elements: the clinical context in which sequencing is suggested (Scollon et al., 2014), the family history (“some family histories may be particularly tragic, and the family tree a veritable tree of death”) (Claret et al., 2018), their past experiences of cancer, and personal beliefs about cancer heritability (McGill et al., 2019; Vetsch et al., 2020).

The decision-making process is informed by how and when genetic testing is offered, thus adding to the initial baggage:

- Parents' confidence in the professional's expertise (McGill et al., 2019).
- Parents' trust in the professional as a person (McGill et al., 2019).
- The perception that the advice is useful for the patient and patient's family (McGill et al., 2019).
- The opportunity for parents and patients to ask questions, although limited by the asymmetry of the patient-caregiver relationship (Gedleh et al., 2018).
- The perception of a risk of a breach of confidentiality (Claret et al., 2018), particularly in the United States (Howard Sharp et al., 2020).³
- The fear of the financial cost for families, already heavily burdened by the cost of cancer treatment (Gedleh et al., 2018).
- The patients' ethnic origin: Sharp et al. found that “only patient race/ethnicity significantly differentiated research participation refusal, with families of black children more likely to decline than families of non-Hispanic or Hispanic white children.” This is consistent with the general underrepresentation of this ethnic group in research protocols (Howard Sharp et al., 2020).
- The risk–benefit ratio (see the following section).

Lastly, the way in which the offer of genetic testing and the results are received is influenced by the many difficulties in understanding genetic concepts. Some patients find it difficult to distinguish between the terms “genetic” and “hereditary” (Gedleh et al., 2018), and between somatic and germline mutations/testing (Johnson et al., 2019; Scollon et al., 2014). In the two-step consent model, parents better understood the difference between somatic and germline variants, although 30% of them were still confused (Johnson et al., 2019). Moreover, some parents found it difficult to change their mind about the perception that cancer ran in their family, even after receiving a negative genetic result (Vetsch et al., 2020).

In response to these challenges and to anticipate the family's emotional reactions, several authors stressed the need to explore the following issues when offering genetic testing and when discussing the results: personal views, belief systems, and particularly personal cancer causation theories, family expectations, previous knowledge of genetics, and the family's social and cultural environment (Forbes Shepherd et al., 2019; Gedleh et al., 2018; McCullough et al., 2016). Several studies reported the patients' and parents' keen desire for methods to improve communication and for information that must be comprehensive, repeated over time, and accessible (Gedleh et al., 2018; Hill et al., 2018; Scollon et al., 2014; Vetsch et al., 2020). Adapting information to the cultural and linguistic context also might improve the patients and their families' understanding (Gedleh et al., 2018).

These studies also included specific recommendations on the information content and format. For example, Scollon et al. suggested that participants should receive “an explanation of the different types of potential tumor and germline whole-exome sequencing results, with specific examples of mutations in each category provided; [and] an emphasis on the fact that germline variant results have potential implications for other family members” (Scollon et al., 2014). McGill et al. emphasized that written information could help to support the information communicated orally, which may be forgotten (McGill et al., 2019). Hill et al. proposed various printed and video educational materials (cartoon booklets adapted for children, and a YouTube channel with videos on genetics), using everyday language, to be used as a long-term source of information. In their study, these new educational resources “were widely viewed and had positive patient feedback” (Hill et al., 2018). They also proposed strengthening the provision of information through peer support groups to ensure “patient-oriented approaches to deliver comprehensive genetic healthcare.”

3.2.4 | Regaining a feeling of control: A perceived benefit for families

Genetic testing could enable individuals to put themselves back in a position of anticipation, a proactive approach that can be reassuring, compared with the powerlessness generated by the disease (Malek et al., 2017; Mandrell et al., 2021; McGill et al., 2019).

³The ability of insurance companies to use genetic data to calculate premiums varies in different countries, with a consequent impact on families. In addition, study participants are not always safe from the risk of data breach.

Parents may see genetic/genomic testing as a source of additional knowledge (e.g., cancer cause, or choice of treatment type; Mandrell et al., 2021; McCullough et al., 2016). The idea of being able to identify something that may be of benefit to their child's care prevails over the anxiety that may be associated with genetic testing (Scollon et al., 2014). In adolescents, the awareness of tumor predisposition could increase knowledge, satisfaction, and fulfillment of expectations (Weber et al., 2019).

Parents are comforted by the feeling of having done as much as they could for their children (McCullough et al., 2016), including having found all the information that could be needed for their care (Scollon et al., 2014). This might be seen as a moral duty, resulting in a form of non-choice that outweighs the identified risks (Mandrell et al., 2021; McCullough et al., 2016). Another reason for the parents' interest in genetic testing is that this exam could give information on the potential impact for the broader family with the hope of being able to estimate the risk, preventing cancer in other children through surveillance and healthier lifestyles (Brozou et al., 2018; Mandrell et al., 2021; Scollon et al., 2014). Families even reported that genetic testing might influence their desire to have more children (Hill et al., 2018; Malek et al., 2017; McCullough et al., 2016).

3.2.5 | Family fears add more complexity to a highly sensitive situation

Among the factors for refusing NGS testing for their child, parents cited feeling overwhelmed by the current situation and thus unavailable to engage with a new proposition, particularly if they feared a possible psychological effect, such as guilt, linked to the result nature (Howard Sharp et al., 2020; Mandrell et al., 2021; Scollon et al., 2014). Tensions may arise within families, such as marital conflicts that may lead to divorce or abandonment (Gedleh et al., 2018). Moreover, not knowing the level of genetic risk might limit the desire to have other children due to fear of passing on the disease (Foster et al., 2017).

3.2.6 | Identifying the child's interests: A professional dilemma

If a cancer predisposition is identified, it represents a way to anticipate potential recurrence through surveillance, but also a burden of knowledge that can have a major detrimental impact. In this context, the potential advantages and drawbacks as well as personal autonomy regarding the right not to know must be discussed before testing (Brozou et al., 2018). This is particularly important when the findings concern the child's future rather than the child's present. Indeed, many national and international guidelines agree that "in general, having this kind of information about the child's future health is contrary to the child's best interests and children should only undergo predictive genetic testing if

preventive treatment and measures can be initiated during childhood" (Esquivel-Sada et al., 2017). This produces a kind of subjectivity in the involved individuals because healthcare professionals and parents "will pull upon different beliefs to make decisions regarding genetic testing" (Desrosiers et al., 2019). Healthcare professionals highlighted the issue concerning the disclosure of results obtained by somatic testing, which currently does not always require rigorous consent like germline genomic testing (Desrosiers et al., 2019). Another ethical question concerns the return of results to families in the event of the patient's death (Desrosiers et al., 2019). These issues stress the need for adequate training for healthcare professionals.

The need for training for healthcare professionals

The implicated healthcare professionals must have adequate training in genetics. This will allow them to properly weigh up the ethical and psychosocial issues raised, such as the effect on the family of a predisposition risk (Forbes Shepherd et al., 2019). Johnson et al. proposed that the lack of training among healthcare professionals may have a negative impact on the patients through excessive or inadequate screening, treatment based on inappropriately interpreted results, and ignorance of the implications of sequencing findings, thus violating the principle of nonmaleficence (Johnson et al., 2017). Desrosiers et al. (2019) noted that professionals wanted support from genetic counselors when returning the genetic testing results because of their lack of confidence in ordering, interpreting, and discussing the results of somatic and germline genomic and exome sequencing.

When involving geneticists and genetic counselors

Genetic counselors might be better equipped than oncologists for supporting the families' decision-making about genetic/genomic testing (Desrosiers et al., 2019), for identifying the families' needs in terms of psychological support (Forbes Shepherd et al., 2019), and for proposing an age-appropriate dialogue, particularly appreciated by adolescents (Weber et al., 2019). Support from a genetic counselor reduced the patients' anxiety and improved their understanding of genetic risk (Gedleh et al., 2018). Genetics health professionals play a specific symbolic role for patients because they take the family as a whole into consideration (McGill et al., 2019). Some authors recommended working with an experienced team, trained in genomic testing consent and in active patient communication tailored to the patients' health literacy and numeracy (Johnson et al., 2019; McGill et al., 2019).

Psychological and peer support

A study highlighted the need to provide psychological care to patients and families in parallel to the offer of genetic testing, "given the high levels of distress widely known to characterize this time" (Esquivel-Sada et al., 2017). Psychological consultations could be systematically offered rather than only after the request by the patients/families, in a context where families are highly vulnerable but cannot easily identify their immediate needs. Besides psychosocial support for families, peer support could be encouraged by the clinical team (Hill et al., 2018; Weber et al., 2019).

3.2.7 | Incidental findings and variants of unknown significance: The need for professional guidelines

Healthcare professionals should weigh the potential medical benefits of genomic testing against its current limitations: understanding genotype–phenotype relationships, variability in the expression and penetrance of a disease, potential risks of unnecessary testing, negative psychological effects, and incidental findings with uncertain predictive value (Esquivel-Sada et al., 2017; Mandrell et al., 2021). Indeed, incidental findings bring additional challenges to healthcare professionals: lack of consensus on the disclosure criteria (scientific validity, clinical utility, actionability, surveillance in childhood), subjective practices, lack of training for healthcare professionals, respect for individuals (consent and information), and issues regarding the timeline of the return of results compared with the initial testing (Johnson et al., 2017). For example, the question of actionability is particularly important because it has to be related to the patient's clinical and family history, but testing laboratories do not always have access to this information (Johnson et al., 2017). Some parents have a positive representation of incidental findings, such as clinical utility, psychological utility (relief of guilt), satisfaction of curiosity, and pragmatic utility (plan for future and reproductive decisions). However, some parents and children raised an ethical dilemma concerning “the autonomy of parents to make this kind of choice, as opposed to the child's right to an “open-ended future” (...), the right to have open-ended choices until they are able to take their own decisions regarding their own lives” (Esquivel-Sada et al., 2017; Malek et al., 2017; Mandrell et al., 2021).

Incidental findings also complicate the genetic testing interpretation. Moreover, healthcare professionals may have concerns about how parents will receive them in an already painful situation (Desrosiers et al., 2019; Scollon et al., 2014). However, a study showed that whole-exome sequencing data were not more complex to explain to parents than other data, as long as they serve a clinical purpose, or make parents feel they have done everything they can (McCullough et al., 2016). Therefore, it could be pertinent to incorporate the parents' expectations and conception of utility in the discussion about incidental findings (Esquivel-Sada et al., 2017).

To address all these uncertainties, some authors suggested several best practices: incidental findings and variants of unknown significance must be discussed when obtaining consent (Brozou et al., 2018). Ideally, this should be done in a dedicated pre-testing consultation (Desrosiers et al., 2019) the objective of which is that “the participants understand the purpose of the research, the type of test(s) to be performed, the various types of results that they will receive (and not receive) through participation and in which situations they have a choice about receipt of results in these categories, as well as the potential risks and benefits of participation” (Scollon et al., 2014). In line with this, “presymptomatic testing results should be communicated by genetic specialists in a multidisciplinary team, and appointments spread out over time with follow-up support” (Esquivel-Sada et al., 2017; Johnson et al., 2017).

4 | DISCUSSION

In this narrative review, we analyzed the existing psychological, ethical, and sociological studies on genetic and/or genomic testing in pediatric oncology. We identified a small number of articles (18) on this topic written mainly by researchers in biomedical humanities and social sciences who investigated the perspectives of children with cancer, their parents, and/or healthcare professionals. Various points of agreement emerged from these articles. From the perspective of children and their parents genetic or genomic testing appeared to be secondary relative to the emotional upheaval caused by cancer, making it difficult to propose it (McGill et al., 2019; Scollon et al., 2014). Families had insufficient genetic information in the context of their child's cancer (Foster et al., 2017; Gedleh et al., 2018; Hill et al., 2018; Johnson et al., 2019; Vetsch et al., 2020). The child's role should be better considered, by differentiating it from the parents' role, through separate consultations and appropriate language and timing (Claret et al., 2018; Weber et al., 2019). One particular area of research concerned the role of adolescents and young adults, and their capacity to consent and be informed directly, or reformed as they grow (Forbes Shepherd et al., 2019; Foster et al., 2017; Gedleh et al., 2018). In another vein, three articles analyzed the motivations of parents who are in favor of incidental finding disclosure. They found that these parents wanted to take a proactive, anticipatory approach. This revealed a broad understanding of the clinical utility of incidental findings (Malek et al., 2017; Mandrell et al., 2021; McCullough et al., 2016). From the point of view of healthcare professionals, specific training in genetics should be introduced and regularly updated for those implicated in genetic and genomic testing. This will allow them to develop more appropriate skills and help them to address the complex ethical questions raised by these tests (Desrosiers et al., 2019; Forbes Shepherd et al., 2019; Hill et al., 2018; Johnson et al., 2017). Thus, the ethical issues raised by genetic/genomic testing in pediatric oncology are mostly similar to those raised in adult oncology, or even in other disease contexts (Isidor et al., 2019). However, some are specific to children: the right to an “open-ended” future of knowing or not knowing; consideration of the child's opinion, particularly for adolescents who are developing their autonomy; respecting their parents' opinion; and the difficulty in oncogenomics to comply with ethical guidelines for genetic investigations for diseases that develop later in life (presymptomatic testing).

4.1 | Study limitations

Our narrative review has several limitations. First, articles published from 2006 were included, but sequencing methods, and thus the associated issues, have greatly evolved since then. We were aware of this point, but we wanted to examine the inclusion of genetic and then genomic testing in pediatric cancer care in a comprehensive way. Second, from a methodological point of view, all included articles concerned children with cancer (or history of cancer), but some considered the question of predisposition for the child and/or the child's family (e.g., hereditary retinoblastoma, Li-Fraumeni

syndrome), while others involved fictional scenarios of possible genetic results. Third, as some studies were retrospective and others prospective, participants were consulted at different points during the care pathway and thought processes. Moreover, the potentially different legal contexts in different countries may have influenced the interpretations by the authors. Fourth, the search terms used were perhaps not exhaustive enough, thus explaining why some studies were overlooked.

4.2 | Clinical implications

Conventional oncogenetic consultations, designed to investigate the existence of an individual or familial predisposition, take into account the timelines and subjective implications of potential results for patients, according to their age, and for their parents (Dattilo et al., 2021; Druker et al., 2017; Lahlou-Laforet & Gimenez-Roqueplo, 2019). However, in the context of the pediatric oncology care pathway, genetic/genomic testing is often offered at very specific times (diagnosis, relapse) for children and their parents, and frequently in an urgent context. Furthermore, there are areas of ambivalence (desire for treatment and of knowledge, uncertainty, and guilt) in relation to the subjective implications of genomic testing for patients and their parents that in various ways seem to mirror the healthcare professionals' concerns. The ethical and psychological issues surrounding predisposition testing for childhood cancers, much discussed in the context of hereditary retinoblastoma or Li-Fraumeni syndrome, should be taken into account also in genetic and genomic testing in pediatric oncology, particularly for incidental genomic data.

This review of the literature complements a previous existing review on genomic testing in children (Gereis et al., 2022) by focusing on cancer and exploring the challenges for parents, children, and healthcare professionals. It raises the question of whether new procedures are needed in the genomic testing pathway for pediatric oncology. For example, somatic and germline genomic sequencing could be proposed in multistep consultations that give each individual the space and time to think and prepare questions in advance. Moreover, it might be sensible to keep the "future open" until the child is old enough to understand or even be committed to the decision (Houdayer et al., 2019). In the context of the growing use of genomics in the clinic, ambitious human and social sciences research projects need to be carried out to identify/analyze the various players involved by starting from the ideas and guidelines developed in the field of pediatric oncogenetics in the last 15 years (Bertier et al., 2017; Botkin et al., 2015).

AUTHOR CONTRIBUTIONS

MDM: Design of the review and selection of articles, then discussion with LH to ensure the relevance of the articles selected at this stage. Active participation in the writing of the article and its subsequent versions. LH: Design of review with MDM, selection of articles, then discussion with MDM to ensure the relevance of the articles selected at this stage. Iterative refinement of the analysis plan through

discussion among the four authors. Review of articles before submission. SDM: Validation of the design of the review, iterative refinement of the analysis plan through discussion among the authors, active participation in the writing of the article and its subsequent versions, and submission of the article. KLL: Validation of the design of the review, iterative refinement of the analysis plan through discussion among the four authors, revision of the article in its various versions. Authors confirm that they had full access to all the data in the study and take responsibility for the integrity of the data and the accuracy of the data analysis. All authors gave their final approval of this version to be published and agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

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CONFLICT OF INTEREST STATEMENT

Author Marion Droin Mollard, Author Sandrine de Montgolfier, Author Lucile Hervouet, Author Khadija Lahlou-Laforet declare that they have no conflict of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

ETHICS STATEMENT

Human Studies and Informed Consent: No human participants in this study.

Animal Studies: Not applicable.

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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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