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Treatment Decision-Making Involvement in Adolescents and Young Adults With Cancer

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Abstract

Problem Identification: Involvement in treatment decision making (TDM) is considered a key element of patient- and family-centered care and positively impacts outcomes. However, for adolescents and young adults (AYAs) with cancer, little is known about the current state of knowledge about their perspective on and involvement in TDM or the factors influencing the AYAs' TDM involvement.

Literature Search: Integrative review focused on AYAs aged 15–21 years, their involvement in TDM, and factors influencing their involvement using the MEDLINE[®], PsycINFO[®], CINAHL[®], and Web of Science databases.

Data Evaluation: 4047 articles were identified; 21 articles met inclusion criteria.

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Pyke-Grimm, Franck, Halpern-Felsher, Goldsby, and Rehm contributed to the conceptualization and design. Pyke-Grimm, Kelly and Kleiman completed the data collection. Pyke-Grimm provided statistical support. Pyke-Grimm, Franck, Kelly, Halpern-Felsher and Rehm provided the analysis. All authors contributed to the manuscript preparation.

Synthesis: Five factors were identified: AYAs' preferred, actual, and perceived involvement; age and cognitive maturity; disease and illness factors; information and communication; and relationships, roles, and perspectives of parents and healthcare providers. AYA involvement in TDM varies depending on the magnitude and timing of the decision. AYAs' role in decision making is situational and may evolve with time.

Implications for Research: Research is needed to understand AYA's preferences for TDM, the type and degree of their involvement, and the interactions between factors that contribute to or impede TDM.

Keywords

adolescent and young adult; pediatric oncology; decision making; patient participation

Introduction

Although cancer survival rates have generally improved for pediatric and adult patients, survival rates for adolescents and young adults (AYAs) with cancer have not kept pace. This lack of progress is most evident for those between 15–25 years (Albritton et al., 2006, Bleyer, Ulrich, & Martin, 2012) who are confronted not just by cancer, but also by dealing with normal developmental challenges, the progression of which is affected by dependence on family and healthcare providers (HCPs), and by parental protectiveness (Coyne & Gallagher, 2011).

An important reason for the poorer outcomes in this age group is nonadherence to cancer treatment. Nonadherence rates as high as 60% have been reported (Alsous et al., 2017; Bhatia et al., 2014; Smith, Rosen, Trueworthy, & Lowman, 1979), which can lead to cancer relapse (Bhatia et al., 2015; Butow et al., 2010). Participation of AYAs in treatment decision making (TDM) may support adherence to medical treatment (Butow et al., 2010).

A core principle of patient- and family-centered care is empowering patients and families and build their confidence so they can make decisions about their health care (American Academy of Pediatrics, 2012). AYAs with cancer are increasingly encouraged to be involved in TDM by organizations such as the National Comprehensive Cancer Network (Coccia et al., 2014). Professional organizations, government agencies, and ethical and legal perspectives promote the inclusion of children and adolescents in TDM. However, guidelines about when and how to involve children and adolescents in TDM are mostly opinion-based with little empirical support, and evidence suggests they are infrequently followed (Unguru, Sill, & Kamani, 2010).

Understanding AYAs' preferences is key to changing healthcare delivery to improve participation in decision making, satisfaction with the process of decision making, adherence to the therapeutic plan, and ultimately, outcomes. To understand AYAs' involvement in TDM and factors that influence how they approach TDM, the authors conducted an integrative review of the literature. Because most pediatric oncology units primarily care for patients aged 21, years or younger, in this review, the authors focused on the AYA age group (15–21years). The aims of the review were to determine the following:

- The current state of knowledge about AYAs aged 15–21-years with cancer and their perspective on and involvement in TDM
- Factors influencing the AYAs' TDM involvement, such as their age, developmental stage, and phase in the continuum of care
- Their TDM involvement within the context of their family and with their HCPs.

Background

Cancer treatment for AYAs requires families to make challenging decisions throughout the disease trajectory, including at the time of diagnosis, during disease recurrence, during therapeutic changes, and at the time of end-of-life care (Stewart, Pyke-Grimm, & Kelly, 2012). Current recommendations are that children should be involved in decisions when they are able to do so and choose to participate (Joffe et al., 2006; Masera et al., 1997; Spinetta et al., 2003).

A key aspect of the phenomenon of AYA TDM is the triangular relationship among the patient, HCP, and parents. Parents sometimes assume a gatekeeper role, deciding what the child may be told to protect him or her from upsetting information (Young, Dixon-Woods, Windridge, & Heney, 2003). Stewart et al. (2012) reported variability in the degree to which parents involved their child in TDM. Parents focused on whether participation in TDM was in their child's best interests, allowing more involvement in older children and less involvement when they prioritized sparing the child from distress.

Older children and AYAs commonly defer to parents or physicians for TDM for multiple reasons, including feeling pressured, wishing to avoid conflict, and deferring to others' knowledge or experience (Knopf, Hornung, Slap, DeVellis, & Britto, 2008). HCPs may not be able to accurately estimate the cognitive development of AYAs and, therefore, their ability to participate in or understand issues of TDM (Coyne & Gallagher, 2011). The imbalance of power between AYAs and parents and clinicians means their participation in TDM is likely under external control. In a study of the attitudes of pediatric oncologists, (de Vries, Wit, Engberts, Kaspers and van Leeuwen 2010) found that some physicians do not believe adolescents are capable of meaningful participation, do not always provide adolescents with necessary information and believe that proxy consent from parents is sufficient. However, the American Academy of Pediatrics (2012) policy statement advocating patient- and family-centered care makes recommendations to support effective partnerships among children, parents and families, and HCPs.

When children and AYAs participate in TDM, they may benefit in multiple ways, including improved sense of control and autonomy and improved adherence to medical treatment (Barakat, Schwartz, Reilly, Deatrck, & Balis, 2014; Butow et al., 2010; Coyne, Amory, Kiernan, & Gibson, 2014; Coyne & Gallagher, 2011; Kelly, Mowbray, Pyke-Grimm, & Hinds, 2017; Ruhe, Badarau et al., 2016; Snethen, Broome, Knafl, Deatrck, & Angst, 2006). Participation in TDM could be overly demanding (Ruhe, Badarau et al., 2016). However, research results about why, how and under what circumstances AYAs make cancer-related treatment decisions are unavailable. There are no reviews to the authors'

knowledge that examine the research literature as a whole to guide practice and further research on AYAs' preferences and involvement in TDM or influencing factors.

Methods

Whittemore and Knafl's (2005) methodology for conducting an integrative review served as the framework for this article. TDM was defined as decisions surrounding treatment or research aimed at curing or delaying cancer progression, or a decision about diagnostic, therapeutic, procedure or supportive care choices. Older children were defined as ages 7–14 years.

A systemic search of the research literature was undertaken using the following databases: MEDLINE®, CINAHL®, PsycINFO®, and Web of Science. All databases were searched from their inception (PsycINFO 1880; Web of Science 1900; Medline 1946; CINAHL 1981) through June 8, 2017. An initial scoping search led to the exclusion of Sociological Abstracts as it did not yield any unique, relevant articles. Reference lists of relevant articles were examined to find additional references. A combination of database index terms and keywords were used to ensure maximum recall of relevant articles, targeting three main concepts of the search: TDM, AYA age group, and childhood cancer. Non-English articles were excluded. The search strategy had three main concepts: cancer and related diseases; TDM, including patient participation and informed consent; and the AYA age group. Each concept was searched using a combination of database index terms and extensive keywords.

Articles identified by the search strategy underwent further screening according to the inclusion and exclusion criteria. The following inclusion criteria were used:

- Original quantitative, qualitative, and mixed methods studies, systematic reviews, and meta-analyses that included AYAs aged 15–21 years with cancer
- Studies that had a majority of participants with cancer if they included other chronic illnesses
- Procedures, methods, and analysis clearly described
- Studies that examined AYAs' involvement in or perspective of TDM regarding their cancer treatment

The exclusion criteria were as follows:

- Studies limited to survivors or studies focused solely on fertility preservation
- Non-systematic reviews, editorials, or case reports

Data extraction included reviewing the articles for the purpose statement and description of design, sample characteristics, variables measured, data collection, analysis, main findings, and limitations (see Table 1). PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analysis) was used as a guide for reporting the studies (Moher, Liberati, Tetzlaff, & Altman, 2009). Factors were identified to synthesize the findings. The Mixed Methods Appraisal Tool (MMAT) was used to assess the methodological quality of the articles (Pace et al., 2012; Pluye, Gagnon, Griffiths, & Johnson-Lafleur, 2009). This tool allows analysis of

multiple study types, including mixed methods; qualitative; randomized controlled; non-randomized; and observational descriptive. Articles are scored from 0–4, with 0 indicating no criteria met and 4 indicating all criteria met. Two of three authors (K.P.G., K.P.K., R.R.) scored each article using the MMAT. Differences of opinion were discussed until consensus was achieved. Data analysis entailed developing categories of findings and identifying important factors by collapsing similar results and comparing findings between studies.

Results

The search identified 4047 possible studies. After removal of duplicates and further review of abstracts and full study reports in accordance with the inclusion and exclusion criteria, 21 articles met the inclusion criteria and were selected for critical review: 15 were qualitative, and 6 were quantitative. Figure 1 shows the PRISMA flowchart. Two articles were secondary data analyses. The years of publication ranged from 1993–2017. Two articles exclusively reported on AYAs (age 15–24 years), and 19 articles included children aged 14 years and younger in addition to the target group of AYAs. Across the 21 studies, age ranged from 7–24 years. The studies took place in the United Kingdom (n = 2), the Netherlands (n = 3), Switzerland (n = 2), Australia (n = 2), Canada (n = 1) and the United States (n = 13). Two of the 21 studies were multisite and conducted in two different countries. The 21 studies included participants who were receiving treatment for cancer, no longer receiving therapy (survivors), or diagnosed with a different chronic illness. The types of decisions described in the studies were primarily related to research participation (phase 1, 2 or 3 trials) (n = 8), end-of-life decisions in which there may have been an option to participate in a phase 1 clinical trial (n = 2), and TDM in general (i.e., at diagnosis, treatment, relapse and supportive care) (n = 11).

The MMAT study quality scores ranged from 2–4 (0–4 scale). Three studies were assessed to be of high quality (score = 4), 15 as moderate quality (score = 3) and 3 as lower quality (score = 2). All 21 studies were included so as to be comprehensive in examining the scope and depth of the phenomenon (Noblit & Hare, 1988).

The following five major factors were identified from the 21 studies, reflecting the AYAs' involvement in TDM or influencing factors:

- AYAs' preferred, actual, and perceived involvement
- Age and cognitive maturity
- Disease and illness factors
- Information and communication
- Relationships, roles, and perspectives of parents and HCPs

The factors relate to the degree of involvement in TDM (factor 1) and those influencing TDM (factors 2–5).

Preferred, Actual, and Perceived Involvement

Older children and AYAs with cancer did not always participate in TDM at their level of preference and comfort (Unguru, 2011; Zwaanswijk et al., 2007) and they varied in their preference for involvement in TDM, from none at one end of the spectrum, to making most of the decisions at the other. No matter the AYA's role in decision making, the family was usually involved to some extent.

Responses ranged from completely deferring decision making to others, to independently making decisions. In 3 of the 21 studies, older children and AYAs described how they preferred to have their physician or parents decide about their treatment (Ellis & Leventhal, 1993; Stegenga & Ward-Smith, 2008). Ellis and Leventhal (1993) reported that most wanted the physician to make all of the decisions. Some older children and AYAs believed there was no real decision to make because the only choice is to proceed with life-saving therapy (Coyne et al., 2014). Similarly, Barakat et al. (2014) found that the majority of AYAs had little or no role in TDM about phase 3 clinical trials. Unguru et al. (2010) also reported that older children and AYAs perceived they had little or no role in the decision to participate in research. Five studies identified older children and AYAs' preference to collaborate with their parents and providers (Dunsmore & Quine, 1996; Kelly et al., 2017; Ruhe, Badarau et al., 2016; Unguru et al., 2010; Zwaanswijk et al., 2011); most wanted to be informed (Zwaanswijk et al., 2007) but did not want the sole responsibility for making decisions (Dunsmore & Quine, 1996; Unguru et al., 2010).

Some older children and AYAs preferred to be or were fully engaged in the decision (Weaver et al., 2015) or made the final decision with parental assistance (Zwaanswijk et al., 2007). Two studies reported that older children and AYAs wanted to be either more informed or more involved than they actually were in decision making (Dunsmore & Quine, 1996; Unguru et al., 2010). Having autonomy in decision making related to clinical trials was very important to most AYAs in a study conducted by Pearce et al. (2016). In seven articles, altruistic motives were a reason for participating in clinical research (Barakat et al., 2014; Broome, Richards, & Hall, 2001; Hinds et al., 2005; Miller et al., 2013; Pearce et al., 2016; Read et al., 2009; Unguru et al., 2010).

Investigators reported situations when older children and AYAs did not want to assume responsibility for making the decision, such as at or close to the time of diagnosis (Stegenga & Ward-Smith, 2008; Weaver et al., 2015). At diagnosis, there was often an urgency to start treatment while older children and AYAs were too ill to participate in discussions or decision making (Barakat et al., 2014; Zwaanswijk et al., 2007). There were also situations when they did not want to surrender the control of TDM, such as at the end-of-life (Hinds et al., 2005; Miller et al., 2013). Barakat et al. (2014) reported the AYAs recalled minimal or no role in clinical trial TDM, although their parents recalled substantial efforts to involve the AYA. AYAs and parents had markedly different memories of the same event, and AYAs experienced regret for not being involved as they would have preferred.

Age and Cognitive Maturity

Eight of the studies reviewed showed that age and cognitive maturity were factors influencing whether older children and AYAs were involved in TDM. Five studies reported that increased age and maturity was associated with increased decision involvement (Barakat et al., 2014; Coyne et al., 2014; Ellis & Leventhal, 1993; Unguru et al., 2010; Zwaanswijk et al., 2011). Unguru et al. (2010) reported age was associated with the types of decisions, roles performed, and physicians' willingness to discuss decision making, with older children being more likely to be involved in TDM. In the study by Zwaanswijk et al. (2011), parents, patients, and survivors uniformly recommended that children (aged younger than 12 years) not be involved in TDM (odds ratio [OR] = 0.1, confidence interval [CI] [0.0, 0.2]), but preferred that adolescents (older than 12 years) be involved (OR=18.2, CI [6.8, 48.4]). Patient age was the main factor associated with information and preferences to participate in decision making. Ellis and Leventhal (1993) found that older children and AYAs believed the patient should be at least 16 years to participate in making minor treatment decisions. Barakat et al. (2014) reported that cognitive and emotional maturity facilitated involvement in phase 3 clinical enrollment decisions.

In contrast, several studies showed that age was not associated with family decision-making patterns, such as exclusionary, informative, collaborative and delegation (Snethen et al., 2006). Weaver et al. (2015) also found no correlation between age, time on treatment, new or relapsed disease, and decision-making preference, although their study consisted of only 40 participants. In their study of 51 older children and AYAs with cancer, Dunsmore and Quine (1996) found that age was not correlated with decision making.

It was not possible to determine from the studies if preferences for TDM were based on age, experience with cancer and cancer treatment, or other factors. Younger patients may have been diagnosed with cancer for a longer period of time than older, recently diagnosed AYAs, so may acquire experience beyond their years.

Disease and Illness Factors

Fifteen articles reported findings relating older children and AYAs' disease and illness factors to TDM involvement. Their symptoms, the seriousness of the condition, and the urgency of the decision were barriers to their participation in TDM (Barakat et al., 2014; Broome et al., 2001; Zwaanswijk et al., 2007). Prognosis also determined whether older children and AYAs were included in TDM. Parents did not want them to participate if they had a moderate (OR = 0.5) or unclear (OR = 0.2) prognosis (Zwaanswijk et al., 2011). Barakat et al. (2014) highlighted acute emotional or physical stress as a limiter of AYAs' involvement and participation in decision making. These studies demonstrate that the emotional or physical state of older children and AYAs can influence the actual TDM role they assume.

Additional factors influenced older children and AYAs' choice or actual participation in TDM. They classified decisions as minor (delivery of care, decisions about pain management and antiemetics) or major (decisions about treatment protocols) (Coyne et al.,

2014; Ruhe, Wangmo, et al., 2016; Tenniglo et al., 2017). They also considered major decisions not to be decisions at all because there was only one obvious choice and refusal was not an option (Coyne et al., 2014). Decision making involvement was situational; older children and AYAs preferred to participate in minor or supportive care decisions, if they felt well but left most major decisions to the HCP (Ruhe, Wangmo et al., 2016; Weaver et al., 2015). Kelly et al. (2017) also reported that most older children and AYAs did not want to make big decisions.

TDM involvement was often dependent on the stage in the disease trajectory. In one study by Miller et al. (2013) 85% of older children and AYAs reported they made the final decision about a phase 1 cancer study. They had all experienced the standard diagnosis and management, so they would be classified as experienced, no matter their chronological age. Other researchers found decision making shifted from the parents or HCP to older children and AYAs later in the disease trajectory (Hinds et al., 2005).

Information and Communication

In 17 studies, older children, AYAs and families referred to information seeking, information exchange, and communication with the healthcare team in the context of decision making. Limited knowledge of the disease and treatment were important barriers to participation in decision making (Coyne et al., 2014; Stegenga & Ward-Smith, 2008; Zwaanswijk et al., 2007). Ellis and Levantahl (1993) found that although most older children and AYAs with cancer wanted the doctor to make all decisions, those who had less understanding of their diagnosis were less likely to want to be involved in making decisions than those with more understanding ($p = 0.039$). Alternatively, Coyne et al. (2014) reported that adolescents identified receiving information as an important determinant of their participation in shared decision making, claiming they were limited by parents and HCPs. Ruhe, Wangmo, et al. (2016) found that parents actively limited information that could be upsetting or related to prognosis, therefore limiting the knowledge older children and AYAs needed to participate in TDM.

In Zwaanswijk et al.'s (2011) study, 60% of older children and AYAs felt it was important to receive information even if they did not want it at the time. They also thought the patient should participate in medical decision making. Pearce et al. (2016) reported that AYAs thought effective communication, information availability, and the opportunity to ask questions was important for them to participate in trials. A trusting relationship with their HCPs allowed for good communication and encouraged trial participation.

In contrast, Stegenga and Ward-Smith (2008) found that receiving information and being informed was important to older children and AYAs, but not necessarily linked to decision making. They studied adolescents within six months of diagnosis, finding that their desire for information was not a desire for control but simply a desire to understand; they believed the healthcare team should make the decisions.

Relationships, Roles, and Perspectives

Eighteen studies discussed how the relationship or role of the parent or HCP influenced older children and AYAs's involvement in TDM. Ruhe, Wangmo et al. (2016) reported that physicians and parents regulate older children and AYAs's participation in decision making. They sometimes controlled the amount, type and timing of information given to the child.

The most frequently reported factor by older children and AYAs, parents, and physicians in making decisions was consideration of and sensitivity to others (Hinds et al., 2005). They relied on their parents to protect them and trusted them to make decisions in their best interests (Broome & Richards, 2003). They trusted HCPs because they assumed they were experts (Coyne et al., 2014). They wanted support or preferred shared decision making with family and clinicians (Coyne & Gallagher, 2011; Zwaanswijk et al., 2007).

Snethen et al. (2006) described four family patterns of decision making in the context of clinical trials. Parents whose goal was to protect older children and AYAs and excluded them from participation in TDM were identified as exclusionary. Parents who preferred to have their older children and AYAs understand the issues and the meaning of the decisions were identified as informative. Parents whose children tended to be older compared to the members of other groups and who advocated active participation by AYAs were identified as collaborative. Finally, parents who approved the choice to be made but turned over the actual decision to the AYAs were identified as delegators.

Collectively, these findings highlight the complexity of the triadic relationship among the provider, parent, and AYAs. Their involvement in TDM was related to many variables, including age, experience with illness, and their progress along the continuum of care.

Discussion

The findings from these studies demonstrate that, for older children and AYAs, their actual TDM involvement varies based on their chronological and developmental stage, disease state, previous experience with disease, type and magnitude of the decision, and decisional and family context. Preferences for their level of involvement in TDM varies during cancer treatment for many reasons (e.g., stage of illness, seriousness of condition, type of decision); therefore, participation appears to be an ongoing process that varies with the situation (Coyne & Harder, 2011).

Even though the findings included older children, the discussion will focus on how they pertain to AYAs. In this integrative review, five factors were identified about AYA's involvement in or perspectives of TDM regarding their cancer treatment. Treatment decision making is situational because AYAs are comfortable making some decisions but not others; parents and HCPs have varying degrees of involvement. Decisions they are more comfortable with include minor decisions about care delivery (Coyne et al., 2014), whereas for decisions of tremendous consequence, such as those at the time of diagnosis, they are more likely to defer to their parents or providers.

Decision making is a process that evolves during an AYAs disease trajectory. AYAs enter the illness journey with little or no understanding of their disease, with their experiences providing a rapid education. Later in the course of the disease, when many of these AYAs experience complications and face a decision about alternative treatment or end-of-life care, they have become knowledgeable and more assertive about their involvement in decision making (Bluebond-Langner, Belasco, & DeMesquita Wander, 2010; Hinds et al., 2005; Lyon, McCabe, Patel, & D'Angelo, 2004; Miller, 2009; Miller et al., 2013; Miller & Harris, 2012; Miller, Reynolds, & Nelson, 2008).

Early in the disease trajectory, the degree of involvement by AYA's in their own TDM is determined primarily by the attitudes of the HCPs and parents, not by the AYA's ability (Coyne, 2008; Martenson & Fagerskiold, 2008). Even when AYAs are competent to make treatment decisions, they still want support and prefer shared decision making with family and clinicians (Coyne & Gallagher, 2011; Pearce et al., 2016; Zwaanswijk et al., 2007).

This review identified contradictory findings regarding the association of age with TDM. Bluebond-Langner et al. (2010) argued that relying on chronological age or developmental stage to determine how to involve young people with cancer in decision making is unreliable because of the marked inter-individual differences in rate and stage of development and treatment experience. Recently diagnosed patients—no matter what age— may still be in the passive, early state of decision making. If they are experiencing substantial suffering or stress, even the most mature adolescents are likely to rely on relatives or other proxy decision makers (de Vries et al., 2010). In a systematic review Day, Jones, Langner and Bluebond-Langner (2016) also reported that adolescents' ability to participate in TDM and discussions, as determined by their HCP, is determined by their maturity or disease experience and not their age. In the studies reviewed, researchers did not provide information about the distribution of experience of their samples, so this interaction cannot be identified. Future studies must assess the complexity of the patient experience in addition to the participants' chronological age.

How information is provided and the degree to which parents and HCP directly communicate with AYAs is an important factor in determining involvement in and experience of TDM (Stegenga & Ward-Smith, 2008; Zwaanswijk et al., 2007). For some, receiving information and communication defines their level of involvement in decision making. For adolescents, lack of information is a barrier to active decision making. Adolescents living with chronic illness value clear and straightforward technical information (Britto, Cote, Horning, & Slap, 2004; Britto et al., 2007; Dunsmore & Quine, 1996; Knopf et al., 2008). Less than 20% of adolescents preferred patient-led decision making; the majority do not want to direct decision-making authority or make the final decision (Britto et al., 2004; Knopf et al., 2008). Knopf et al. (2008) suggested this may be due to their recognition of how complex and life-threatening the decisions really are. This finding is supported by (Kelly et al. 2017) who suggested that, during cancer treatment, older children and AYAs may prefer information exchange and communication to being involved in actually making treatment decisions.

Treatment decision making is not one-dimensional. The findings suggest there is an interaction between age, previous cancer experience, and decision making. Studies support the finding that TDM patterns change with time and as individuals become experienced with their disease. Many of these studies examine characteristics of AYAs or decision making in one dimension, rather than identifying the many factors that modify the situation. For instance, an 18-year-old, who is newly diagnosed with cancer may be naïve, but a 10-year-old may be very experienced, having lived with cancer for the past five years. Important dimensions that must be considered in studying or advocating for AYA involvement in TDM include age, the normal developmental progression of AYA decision making, experience with the illness, low-risk decisions versus high-risk decisions and decisions with a clearly identified best option versus no best option.

Any effort to involve AYAs in TDM must take into account the parents' perspectives and their role in the care and treatment of their children. Bluebond-Langner et al. (2010) reported that parents feel strongly that the responsibility for the decision rests with them. More research is needed to determine how AYAs with cancer interact with their family to make treatment decisions, (e.g., how each negotiate their roles, given findings suggesting that decision making by the AYA and their families is a dynamic, social process).

Limitations

The findings of this integrative review should be considered in light of several limitations. For example, most of the included studies included participants who were younger and older than the target AYA age range. Because most of these studies did not report findings for subsamples, it was not possible to determine how the reported results were similar or different with respect to age or condition. Another limitation was the lack of consistency in how TDM was defined, making it challenging to interpret and compare findings across studies. Many of the studies were retrospective, with participant recall of their TDM experiences, potentially leading to recall bias. Other methodological concerns included weaknesses across methods (i.e., whether quantitative studies had adequate sample sizes and whether qualitative studies were conducted with adequate rigor). Finally, most of the studies had inadequate representation of minority groups.

Implications for Nursing

The findings of this integrative review provide nurses with a comprehensive summary of the state-of-the-science with respect to AYAs' experience of TDM and influencing factors. In caring for AYAs, nurses must consider multiple factors, such as age, disease, treatment trajectory and relationships with parents and HCPs when encouraging AYAs to participate in decision making. Nurses must also be mindful that the AYAs' role in TDM may be situational and evolve with time to become more active. Recognizing the role of family in TDM is essential. This information could be incorporated into a teaching curriculum for HCPs of AYAs who have cancer or used to develop an assessment tool for AYA decision making.

Future research investigating our understanding of the AYA's voice and preferences for TDM, the actual involvement of AYAs in the decision-making process, and the interactions among factors that contribute to or impede this process is warranted. Studies including relevant outcomes of TDM participation are lacking, particularly related to congruence between desired and actual TDM roles. Important next questions include the following:

- How do AYAs want to be involved in TDM?
- In what types of decisions do AYAs want to be involved?
- How do AYAs negotiate their role and involvement in decision making?
- When does a transition of decision-making authority take place from the parents to the AYAs?

Conclusion

The findings of this review suggest AYAs' role in decision making evolves with time to become more active but is situational. The findings highlight the importance of the role of family and relationship with the HCP throughout their continuum of care. Barriers and facilitators to shared TDM include the AYAs' preferences, age, disease, and relationships with family and providers. Involvement in decision making can be particularly challenging for AYAs with cancer because of their diagnosis of a chronic, potentially life-threatening illness, family influence, developmental stage, and desire to transition to an independent young adult. Additional research would help to identify important areas to focus interventions, in turn moving forward the science directed to the care of the AYA with cancer.

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Knowledge Translation:

- Nurses must consider illness factors, preferences of the adolescent or young adult (AYA), and the importance of the role of family in TDM.
- Information exchange and communication are important for AYA decision making.
- AYA decision making changes with time and experience, so nurses must continually assess the current state of their patients' decision making.

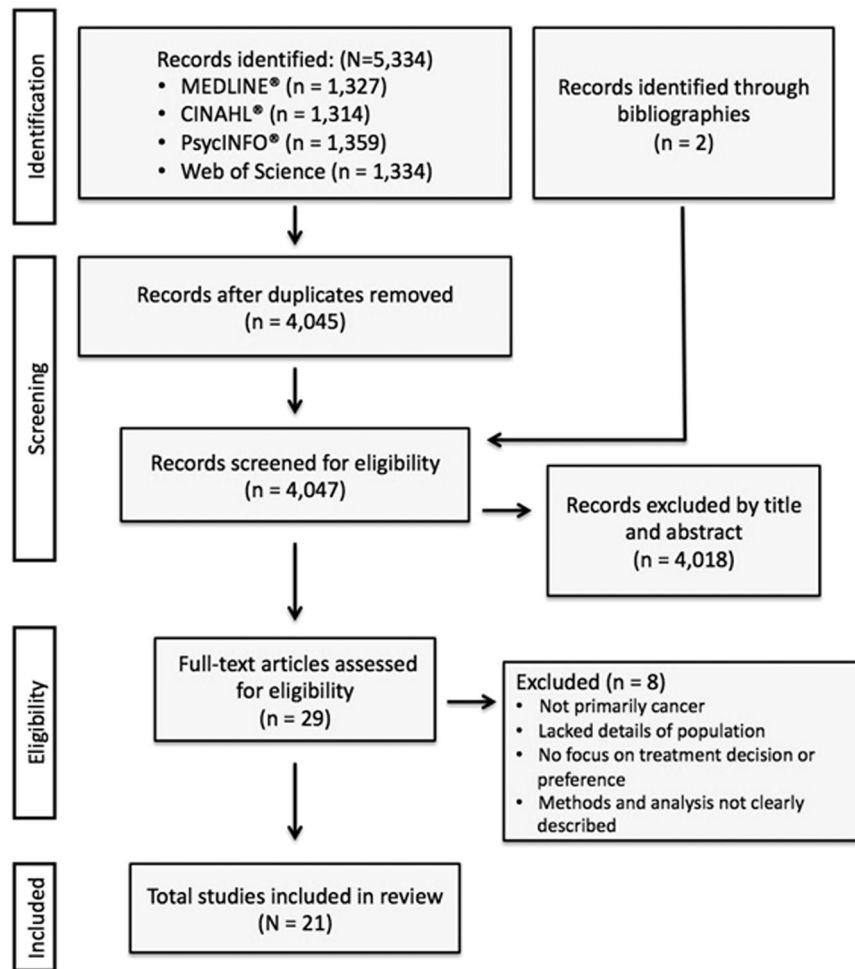


Figure 1. PRISMA flow diagram.
PRISMA—Preferred Reporting Items for Systematic Reviews and Meta-Analysis

TABLE 1.

TDM Involvement in AYAs With Cancer

Study	Purpose and Design	Sample	Methods	Findings	Limitations
Barakat et al., 2014, (United States)	To describe how AYAs and parents make decisions about phase 3 CTs; qualitative; interview guides using semi-structured questions; review of the PRPQ	13 AYAs (aged 15–23 years) who were diagnosed with cancer and offered a phase 3 CT within the past 3–21 months; 16 primary caregivers; 11 primary HCPs	Semi-structured interviews were audio-recorded and transcribed; 2 coders were used to ensure reliability. Constant comparative methods were used.	38% of AYAs abdicated TDM to caregivers. Caregivers (44%) supported AYAs in TDM. Providers (55%) supported a caregiver-based AYA-approved approach.	Only 13 AYAs; large age range; single center; 77% of participants were Caucasian.
Broome et al., 2001, (United States)	To describe AYAs' experience and understanding of clinical research; qualitative, semistructured interviews using narrative analytic techniques	105 family members, including the child with the illness, mothers, fathers, and siblings; 34 AYAs aged 8–22 years who were diagnosed with diabetes or hematologic malignancy and enrolled in a CT	30-minute to 1-hour audio-recorded interviews; 26 open-ended questions; 2 authors read the transcripts, noting themes, patterns, comparisons, and contrasts; 2 investigators read, reread, and coded the data. Data were collected as part of a larger study.	Understanding of research was related to age, previous experience, and diagnosis. Younger children were less likely to know risks of research. Age and diagnosis were influential in predicting what a child could describe about the research and consent process.	Single-site study; 74% of the participants were Caucasian.
Broome, et al., 2003, (United States)	To describe how AYAs' relationship with adults, parents, and researchers influenced their involvement in clinical research; qualitative; semistructured interviews and narrative analytic techniques	34 AYAs aged 8–22 years with diabetes or hematologic cancer; enrolled in a CT within the past 2 months; 39 mothers, 14 fathers, and 19 siblings	Interview guide with questions about recollection of the research experience; individual interviews; 3 interviews were conducted, then the template was modified. Data were collected as part of a larger study.	Participants believed their parents would make the correct decision while involving them in the process. Parents respected the child's decision if it was different from their own or if they declined participation. Parents were the primary influence in TDM.	The larger study the data came from was not specified or referenced. Participants were English-speaking and primarily Caucasian (74%), limiting ethnic diversity. The study was conducted in a single site.
Coyne, et al., 2014, (Ireland)	To explore children's cancer experience and SDM from the perspective of the child, parents, and HCPs; qualitative	20 children (aged 7–16 years) with various types of cancer who were receiving therapy and within 6 weeks to 1 year of diagnosis; 22 parents (5 fathers, 17 mothers); 40 HCPs	Audio-recorded and transcribed interviews; analysis using a grounded theory approach; rigor achieved through a second person coding the data; list of categories compiled, and data reviewed again by 2 members of the research team.	Most major decisions were made by the HCP but discussed with parents. Participation by the child in minor decisions was acceptable. Children seemed satisfied with their level of involvement. Adolescents were frustrated with their lack of control.	Mostly mothers (77%) participated in the study. Most were Caucasian. Data were solely collected in Ireland. It is unknown if the protocols were research protocols or standard of care, or if there was a choice between options.
Dunsmore & Quine, 1996 (Australia)	To highlight the information and TDM needs and preferences of adolescents with cancer, whether those needs were being met, and implications; cross-sectional, exploratory, and descriptive	51 participants aged 12–24 years with multiple cancer types; participants were on (n = 10) or off therapy (n = 41) and were recruited from a teenage cancer camp.	Responses analyzed using content analysis; coding categories developed; chi-squared test applied as appropriate.	About half of participants wanted themselves, their parents, and physicians to be involved in TDM, but only 6% said that happened; 20% wanted only themselves and physicians to be involved.	This study is retrospective. Participants were, on average, about 5 years from diagnosis and mostly off therapy, so there may be recall bias.
Ellis & Leventhal, 1993 (United States)	To determine the information needs and TDM preferences of children with cancer and their parents; cross-sectional survey	50 children and adolescents with cancer (aged 8–17 years); 60 parents	Pilot interviews were used to develop the 132-item survey. Responses were compared between children and their own parents. Parents' survey items were compared between mothers and fathers.	63% of patients knew their diagnosis; 86% were involved in research; 38% knew they were involved. 89% wanted HCPs to make decisions, as did 69% of parents. 72% of patients and 80% of parents.	Only 6% of participants were African American, despite the large percentage of African Americans in the drawing population. Participants were English-speaking and primarily Caucasian males.

Study	Purpose and Design	Sample	Methods	Findings	Limitations
Hinds et al., 2005 (United States and Australia)	To identify preferred EOL choices in children and adolescents with advanced cancer and factors considered by parents and physicians; descriptive study	20 children and adolescents aged 10–20 years with a refractory solid tumor, a brain tumor, or leukemia; 17 were Caucasian. In addition, 19 parents and 14 HCPs took part.	6-item interview guide; individual interviews; ethnograph software to organize data and code every phrase; 2 study team members reviewed the first 3 interviews and developed first-level codes and a code dictionary. The rest of the team used the dictionary to complete a semantic content analysis.	90% accurately recalled the 2 or 3 options presented to them. They identified death as an outcome, considered the outcome of decisions, and could consider more than 1 element at a time. Patients, parents, and HCPs frequently reported consideration of and sensitivity to others.	Small number of participants per group; some had central nervous system tumors and had cognitive deficits so may not recall events accurately.
Jacobs et al., 2015 (United States)	To compare AYAs' EOL preferences with parents who participated in the advance care planning intervention arm (session 1 only) of a 2-arm RCT; survey	17 AYA/family dyads; 34 participants; AYAs were aged 14–21 years with varying cancer diagnoses; 70% were on therapy, and the remainder were survivors.	Individual surveys; data recoded into dichotomous variables; very important or important, or otherwise; congruence between AYAs' and parents' responses were examined using Kappa statistics.	70% of AYAs found discussing EOL decisions appropriate; 65% preferred dying at home; 71% had never heard of an advance directive; 53% had never talked about EOL preferences; 88% felt their parents would respect their wishes.	Heterogeneous with regard to disease and age; 70% were in active treatment, with the rest in follow-up. Single-site study; preponderance of African Americans (58%), so may not reflect Caucasian, Asian, or Hispanic findings
Kelly et al., 2017 (United States)	To determine how children and adolescents viewed involvement in TDM; qualitative study using interactive interview techniques; purposive sampling	29 children and adolescents aged 9–17 years; participants had various cancer diagnoses and ranged in ethnicity, treatment experience, and time since diagnosis. Almost half were female and younger than 13 years old.	20 participants were interviewed using interactive techniques. Cards describing different degrees of TDM involvement were used. 10 more interviews were conducted to confirm results. Constant comparative analysis was used.	Participants reported that parents and HCPs acted in their best interest. Excessive information at introductory visits was confusing and frightening. Most did not want to make decisions they felt were big or consequential.	Retrospective study including those more than 24 months since diagnosis; participants' therapy status not provided; the order of presentation of the cards was not specified and could have influenced responses.
Miller et al., 2013 (United States)	To examine adolescent patients' perspectives of their understanding and decision making about a pediatric phase I cancer study; qualitative; descriptive; structured open- and closed-ended questions	20 participants aged 14–21 years; multiple cancer types; multicenter (6 U.S. children's hospitals); sample of parents considering enrolling a child in a phase I study	Part of a larger study that included parents; interviews using a structured interview with open- and closed-ended questions; questions related to phase I treatment participation.	85% of participants reported making the final enrollment decision; 10% reported being a partial decision maker; 75% identified a reason for participation as potential lengthening of life or cure; 45% participated because they had no other option.	Small sample size; mostly Caucasian males
Pearce et al., 2016 (United Kingdom)	To explore perceptions and experiences of AYAs participating in CTs and explore those of the participants' HCPs; interpretive, qualitative, semistructured narrative interviews	21 AYAs aged 15–24 years with primary bone cancer; all participants in 2 bone or soft tissue CTs; 18 HCPs caring for those participants	Transcripts analyzed using an interpretive approach, including constant comparison methods; a theoretical framework emerged; 2 authors coded the data, and 2 others validated the coding.	Some AYAs thought CTs were the same as chemotherapy or alternative treatment. AYAs and HCPs valued face-to-face conversations. Support from family, peers, and HCPs was critical.	Retrospective study; participant results were not differentiated between on or off therapy; single center; racial and socioeconomic data not provided
Read et al., 2009 (United States and Canada)	To determine personal factors (attitudes) of AYAs about their decision to participate in and experience research and compare AYA to parental	21 AYAs aged 12–22 years and 31 parents	Questionnaires developed using the Dillman method and validated using focus groups, expert review, and a pilot study; Pearson's chi-squared test to assess the association between	26% of AYAs thought participating in research would help them; 67% thought it would help others. The main deterrent to participating was that research would take up too much time (45%). Having too	No specified length of time between diagnosis, offering the CT, and completing the questionnaire. Questionnaires were anonymous, so there was no way to verify if the AYA had actually participated in a

Study	Purpose and Design	Sample	Methods	Findings	Limitations
Ruhe, Badarau, et al., 2016 (Switzerland)	To explore patients' perspectives in pediatric oncology on participation in discussions and decision making	17 AYAs aged 9–17 years receiving treatment at Swiss centers; 6 females, 11 males; time from diagnosis: 3 weeks to 2 years	Data were a subset of a larger mixed-methods project. This qualitative part was composed of face-to-face interviews using a semistructured guide adapted by the interviewer. Transcripts were coded and analyzed using MAXQDA software.	When children and AYAs participated in more than a minor decision, the decision was made with parents and HCPs. Participants were suspicious if only parents talked to HCPs. AYAs valued being able to choose their involvement level.	CT. Data were based on participants' memory, with potential for recall bias. 4 of 21 AYAs refused the interview. Racial makeup was not reported but likely limited. Treating oncologists recruited the children (possible selection bias). Small sample with wide range of ages and times since diagnosis
Ruhe, Wangmo, et al., 2016 (Switzerland)	To learn how patient participation is accomplished in pediatric oncology; subgroup analysis of a larger mixed-methods project conducted in 8 Swiss centers; semistructured, one-on-one interviews about communication, decision making, patient involvement, and inclusion in TDM	17 participants aged 9–17 years who were currently on treatment; 19 parents and 16 oncologists interviewed	Semistructured interview performed individually; the child's primary language was used; thematic analysis by at least 2 researchers; findings presented to a third researcher; disagreements resolved by consensus; MAXQDA software used.	Patients' preferences mostly were taken into account for symptom management; they rarely were taken into account for more essential decisions. A common form of participation was observing or overhearing discussions between parents and HCPs.	Interviewing in 4 different languages may have led to miscommunication. Some parents could not participate because a common language could not be found. Participants were selected by their oncologist, so substantial selection bias is possible.
Snehen et al., 2006 (United States)	To describe patterns of decision making when a child is a participant in a CT; secondary qualitative analysis of a larger study of 34 children and their parents about their experience with research	14 family units; 15 children and AYAs (5 aged 8–11 years, 6 aged 12–15 years, and 4 aged 16–20 years); 28 parents; chronic illnesses included cancer (n = 9), bone marrow transplantation (n = 3), and diabetes (n = 3)	Parents and children/AYAs were interviewed following a semistructured interview guide, and thematic analysis was conducted. Matrices were developed, allowing comparisons within and across family units.	Patterns of TDM were based on parents' goals, the child's level of involvement, and parents' perceptions of their roles. Patterns of TDM were exclusionary, informative, collaborative, and delegated. No specific pattern of TDM related to age.	The participants' education and cultural diversity were not discussed. The sample was about 66% Caucasian. The sample was small, with few participants in each chronic illness group.
Stegenga & Ward-Smith, 2008 (United States)	To determine if the research question was appropriate and if adolescents can provide rigorous data; qualitative, pilot, exploratory study using phenomenological methods and semistructured interviews	3 adolescents aged 13–15 years diagnosed with cancer within the past 4–6 months.	The interviews underwent content analysis and ranged from 15–40 minutes. Both investigators reviewed the transcripts. Inter-rater agreement was initially 90%.	The adolescents believed decisions should be made by the healthcare team, who was most knowledgeable. They wanted information—to know what was going on and what to expect. Family and friends provided support.	Single-center pilot study with only 3 participants; single interviews; no demographics reported other than age and gender; the focus of the study was on the time close to diagnosis only.
Tenniglo et al., 2017 (Netherlands)	To determine supportive care topics important to patients and parents and their preferred role in decision making; qualitative; in-person and online focus groups	18 parents (9 mothers, 9 fathers); 11 patients aged 12–18 years at diagnosis; 2 months after treatment initiation to 6 months after termination of treatment; 54% had acute lymphoblastic leukemia.	2 in-person (parent) focus groups and 1 anonymous online (patient) focus group; data analyzed using thematic analysis; traditional focus group sessions were audio recorded; online focus group data were entered into Atlas.ti.	Patients and parents were involved in medication management, and both wanted to be involved in medication management. Although there was great demand for information at diagnosis, they had minimal time or energy to look for it.	Online focus group questions were set in advance and came from parental in-person focus groups; no pilot testing of questions; recruitment through a patient cancer association; no racial or socioeconomic data
Unguru et al., 2010 (United States)	To determine the understanding of children with cancer about the research in which they participated, research-related treatment, and preferences for	37 participants aged 7–19 years at diagnosis; those who signed a research assent form from 2005–2007 were	69-item quality-of-assess instrument with open- and closed-ended questions; content validity, clinical relevance, clarity, comprehension,	51% were unaware their treatment was research, 86% did not understand the physician's explanation of the trial. Most enrolled to help future children	Unclear length of time since diagnosis; some children interviewed more than 2 years after enrollment; single center; responses may be what children

Study	Purpose and Design	Sample	Methods	Findings	Limitations
Weaver et al., 2015 (United States)	Investigated TDM preferences of adolescent patients with cancer and their perception of parental and clinician behaviors that support their preferred level of TDM; qualitative; 4-question interview guide	40 English-speaking participants aged 12–18 years; diagnosed or relapsed within 6 months of the study; participants were asked about their preferred decisional role on a recent decision.	Interviews transcribed and analyzed using Atlas.ti; 2 blinded team members coded transcripts; intra- and inter-rater reliability calculated; 3-variable ordinal scale and 4-variable nonordinal decisional preference category score.	13% picked their involvement based on the situation. 23% preferred being passive. 23% had mid-level involvement. 43% preferred to be fully engaged in the decision.	Only English-speaking participants; only the patient perspective studied; recall bias; refusal rate was 17%–33% (29% overall); 17 of 40 interviews were not performed alone; numbers in each category were small.
Zwaanswijk et al., 2007 (Netherlands)	To investigate the preferences of children with cancer, survivors, and parents for communication, information, and decision making; qualitative; iterative, asynchronous web-based focus group	25 child and adolescent participants on (n = 7) and off therapy (n = 18); 11 parents; recruitment was by personal request and email solicitation from 2 Dutch oncology units.	Online focus groups were conducted separately with the children and adolescents who were on therapy, by age. Questions were posed daily for 5 days. On days 6 and 7, participants could pose questions. The researchers functioned as moderators.	All wanted open communication with HCPs. Some wanted to be present for physician consultations; others preferred communicating with parents. Survivors and adolescents believed they should make the final decision, with parents providing assistance.	Participants could see other participants' responses; passwords could be shared; small number of patients still on therapy; attrition rate during the course of the 7-day focus group not reported
Zwaanswijk et al., 2011 (Netherlands)	To investigate communication preferences of children receiving treatment, parents, and childhood cancer survivors (treated within the past 5 years), HCP empathy, and information exchange; questionnaire using random selection of 10 of 200 vignettes.	34 children with cancer (aged 8–16 years); 59 parents; 51 survivors (aged 8–16 years at the time of diagnosis, currently aged 10–30 years). Patients and parents participated in the study immediately after a decisive consultation.	Part of a larger multicenter study; questionnaires distributed in person or by mail; vignettes randomly screened for consistency; deconvolitional analysis using multivariate techniques.	Information should be given to parent and child simultaneously if the child is older. Younger, frightened patients should only receive information when they ask. Parents want to receive bad prognosis information first and separate from children.	Hypothetical situations may not represent what is actually done. The survivor group was 2–14 years from diagnosis (potential for recall bias). Treatment and parent groups were asked to participate in person, but survivors were invited by mail.

AYA—adolescent and young adult; CT—clinical trial; EOL—end-of-life; HCP—healthcare provider; PRPQ—Pediatric Research Participation Questionnaire; RCT—randomized, controlled trial; SDM—shared decision making; TDM—treatment decision making