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Preparing Youth with Cancer for Amputation: A Systematic Review

Caitlyn A. Loucas, BA¹, Sarah R. Brand, PhD^{1,3}, Sima Zadeh Bedoya, PsyD², Anna C. Muriel, MD, MPH^{1,3}, and Lori Wiener, PhD²

¹Dana-Farber/Boston Children's Cancer and Blood Disorders Center; Boston, MA

²National Cancer Institute; Bethesda, MD

³Harvard Medical School; Boston, MA

Abstract

Children and adolescents who require limb amputation as part of cancer treatment face many physical and emotional challenges. Preparatory interventions may serve to facilitate positive coping and improve long-term adjustment during pediatric cancer treatment, including decreasing anxiety and post-operative distress. This review aimed to examine and identify the type and degree of psychosocial preparation provided to the child with cancer and family prior to amputation. Electronic databases including Embase, PubMed, and PsycINFO were searched for relevant research articles. Five studies were identified that satisfied inclusion criteria and revealed common themes for preparatory interventions, but results were limited by a lack of empirical approaches and revealed little consensus on pre-operative support prior to amputation. These findings demonstrate that there is a lack of studies to date that have adequately addressed psychosocial preparation prior to amputation for pediatric oncology patients. Future research on preparatory interventions is needed using prospective and quantitative research to establish evidence-based recommendations for interventions to support this vulnerable population.

Keywords

Childhood cancer; amputation; systematic review; pediatric psychology

Introduction

Limb amputation during childhood presents unique challenges in a child or adolescent's development. From a functional and physical perspective, the loss of a limb is a permanent change to the child's ability to ambulate, touch, play, and interact with their environment compared to their unaffected peers. The loss of a body part can also impact the child's emotional and social functioning. Childhood and adolescence are times of increased influence from peers as acceptance into peer groups gains heightened importance. Consequently, focus on body image is increased during this developmental stage. The visible changes following an amputation and the fear of stigmatization from social groups can be a

significant stressor in a young individual's life. Even children as young as first grade have been shown to indicate preferential reactions in favor of children without handicaps over physically handicapped children (Nowicki & Sandieson, 2002; Westervelt & Turnbull, 1980). Asserting independence can be challenged when an amputation results in the increased need for physical assistance and modifications to the child or adolescent's previous lifestyle (Kaiser, 2014; Lasoff, 1985).

Limb amputation as a surgical intervention can be categorized in two ways: Those that stem from traumatic injury and those that are due to biological factors (Atala & Carter, 1992; Smith & Campbell, 2009). In pediatric populations, traumatic amputations are approximately three times as prevalent as biological amputations, with the most common causes involving power tools or vehicular accidents (Bryant & Pandian, 2001). However, biological factors typically manifest as either congenital conditions or secondary to diseases, such as cancer. The most recent estimates from the American Cancer Society predict approximately 15,380 new cases of childhood cancer in children and adolescents under the age of 20 in the year 2016. Of these, approximately 461 (a combined 3%) are projected to be the result of cancerous tumors of the bone, such as Ewing sarcoma and Osteosarcoma (American Cancer Society, 2014). Although these are the most prevalent forms of cancers precipitating amputation, other less common soft tissue sarcomas may involve amputation as well (Atala & Carter, 1992; Clerici et al., 2004). As the efficacy of treatment options continue to improve, the prognoses for pediatric patients with these diagnosed malignancies have improved as well. Increased survival rates result in an increasing number of childhood survivors who now must learn to navigate both the physical and emotional consequences following the loss of a limb.

Preparatory interventions for children and adolescents undergoing medical procedures have shown a wide range of both physical and emotional benefits, including decreased anxiety, better coping and adjustment, use of less pain medication, and shorter hospital stays (Webb, 2010). Although not specific to pediatric populations, studies have shown preparation for amputation reduces long-term postoperative distress and complications and is therefore vital to the overall success of adjustment and rehabilitation following surgery (Andrews, Anderson, Fairbain, & Downing, 2012); Butler, Turkal, & Seidl, 1992; Gallagher & Maclachlan, 2000). An amputation as part of cancer treatment has significant distinctions from traumatic amputations, as it not an unanticipated event that occurs immediately following an accident to an otherwise healthy limb. This time frame is critical for the clinical team, as it can allow for supportive clinical intervention and preparation prior to the planned surgery date. Preparatory interventions can include structured therapeutic strategies, such as the use of medical play or peer-modeling from former similarly-amputated patients, and can be tailored to match each individual child's needs and developmental level.

Several studies have highlighted guidelines and intervention strategies to better help adults or a more general cohort adapt to the loss of a limb (Butler et al., 1992; Fitzgerald, 2000), but research to date that specifically addresses preparation for surgery or the psychosocial impact of amputation in children with cancer is lacking. The aim of this paper is to compile and review the available research reporting effective psychosocial preparation provided to

the child and family prior to amputation. This review aimed to provide recommendations to help prepare children with cancer for an amputation.

Methods

We conducted a systematic review with a comprehensive search of databases in the English language. The databases included Embase, PubMed, and PsycINFO. Eligibility criteria included: 1) Age: Subjects must be children or adolescents, 2) Etiology: Amputation must be due to disease or biologic factors (traumatic amputations were excluded). 3) Preparation: Papers must examine psychosocial support prior to amputation, 4) Language: All papers must be available in the English language; and 5) Publication in peer-reviewed journals or book chapters: Systematic reviews, observational studies, case studies, and expert opinion or consensus papers were included. The initial search revealed a limited availability of research specifically focusing in this area. The following keywords and search terms were selected and used in varying combinative forms for their specificity and relevance to the study topic, including “cancer” OR “neoplasm” AND “amputation” AND “pediatric” OR “child” OR “adolescent” AND “psychology” OR “quality of life” OR “coping” OR “psychosocial” OR “adjustment.” The last search was run on January 26, 2017. The software program EndNote X5 was utilized to collect and organize selected references and articles in accordance with the criteria for inclusion. Items of interest for review papers included review question, study group descriptions, inclusion and exclusion criteria, outcome measures, and findings. Studies were searched and reviewed in accordance with the Preferred Reporting Items for Systematic reviews and MetaAnalyses, PRISMA (Moher et al., 2010) (Figure 1).

For expert opinion or consensus papers, author recommendations were extracted. Two authors (CL, SB) reviewed all articles and performed independent assessments of eligibility with a result of 93% inter-rater agreement. Initially, sixty-eight articles were identified and were subject to review by the authors to assess their compatibility with the search eligibility criteria. Articles (N=63) that did not meet inclusion criteria, including those that were more broadly focused on adult populations or non-cancerous reasons for amputation, were excluded.

Results

A total of five articles met full inclusion criteria and are described in Table 1. While the importance of preparatory intervention and its impact on minimizing distress and facilitating positive adjustment was consistently noted, few standardized, evidence-based approaches were found. In fact, there was little consensus on pre-operative support prior to amputation in this population; however, specific themes regarding supportive interventions are summarized below.

Interdisciplinary and Family Communication

A pre-surgical approach, involving both the medical and psychology teams was described in two case reports (Clerici et al. (2004) and Ferrari et al. (2002). This approach included incorporating a timeline to discuss pertinent details of the amputation with the patient and family. Clerici and colleagues highlight the necessity of informing the parents (of children

up to the age of 21) of the option or necessity for amputation prior to informing the patient. This not only allows for an opportunity to ensure all initial questions are answered prior to gathering informed consent from the caregivers, but also allows the parents to experience their initial emotional reaction in a safe, separate space outside of the presence of their child. Discussions can then take place between caregivers and providers about how to best inform the child of the surgical plan and begin offering preparatory support. Authors of both articles highlighted the importance of multi-disciplinary involvement to enhance communication within the care team and reduce distress in the child and family. Standardized, pre-operative psychosocial support, as well as a basic foundation from which to create preparatory psychosocial interventions capable of being tailored to appropriately meet each individual child and family's needs and developmental level were emphasized, but not offered.

Psychosocial Screening and Assessment

Additional recommendations for preparing children for amputation included a thorough assessment of any pre-operational factors regarding both the child and the nature of the planned surgery (Atala & Carter, 1992; Clerici et al., 2004). For example, in a review of pediatric limb amputations, Atala and Carter (1992) explored factors that may play a role in mediating a child's reaction and adjustment to amputation. These included child-specific variables such as the patient's age, temperament, cognitive capacity, developmental stage, prior experience with medical procedures, and the location of the planned amputation. External variables regarding the family dynamic, such as parental reaction to illness and coping style were also highlighted as having a potentially significant impact on the child's ability to cope. The authors use this information to emphasize the importance of conducting a thorough pre-operative assessment to more effectively tailor intervention efforts to meet the child's and family's specific needs.

Pre-Operative Education

In a sample of 20 adolescent oncology patients undergoing amputation, Tebbi et al. (1989) reported overall good adjustment within twelve months of the amputation surgery, but did identify academic, occupational, and social settings as specific areas for preparatory clinical improvement. The majority (88%) of patients considered themselves independent, yet only 58% of participants indicated a good pre-operative understanding of the functional limitations they would face following surgery. In response, the authors suggested future directions for improving care, including providing pre-operative education for the patient, as well as caregivers, medical staff, and patient's classmates about the psychosocial effects of the procedure as an avenue for clinical intervention. Although these suggestions were not yet offered as part of the clinical standard of care, the authors theorized that this type of education and pre-surgical information could act as a prophylactic exercise to enhance adjustment and pre-empt elements of distress experienced in this population.

Child Interventions

Commonly utilized intervention strategies, such as filmed modeling, play therapy, emotive imagery, hypnosis, and behavioral techniques, were also suggested to help improve the child's coping skills both before and after the amputation surgery. A published case study by Atala and Carter (1992) illustrated the utility of clinical interventions on a 10-year-old

patient undergoing amputation for sarcoma of the femur. In this clinical example, therapeutic interventions, including role reversal, puppetry, and progressive muscle relaxation were effective as preparatory measures in reducing the patient's inpatient procedural anxiety, as well as facilitating positive coping during the rehabilitation process. Peer modeling was also discussed, and included an example where pre-surgical conversations between a patient scheduled for surgery and a former patient who underwent the same amputation improved pre-and post-surgical outcomes by making the patient more amenable to the operation and facilitating easier post-surgical functional and emotional adjustment (Clerici et al., 2004).

Family Interventions

In addition to interventions designed to directly support the pediatric patient, family-specific therapies were also proposed as an effective component of pre-surgical psychosocial involvement. One such clinical suggestion included interventions designed to identify and address discrepancies in coping style and stages of grief, as parents and children may likely vary in their reactions to the planned amputation (Atala & Carter, 1992).

Jaraway et al. (2013) evaluated the retrospective feedback of caregivers of children diagnosed with bone cancer following an intervention where the parents were allowed to view their child in the operating room following amputation, but prior to reversing anesthesia. The study found that all respondents who viewed their child and the results of the amputation in the operating room prior to reversal of the anesthesia reported it as valuable and a positive preparatory experience. The researchers hypothesize that this interventional strategy allowed parents to express their grief, shock, and emotional reaction to their child's post-surgical condition in a safe, controlled space, so that their presence once the anesthesia had subsided could be more targeted toward supporting their child's coping. The authors stressed the need for prospective research, but recommended this opportunity be extended to all pediatric oncology centers for parents of patients undergoing amputations to help promote positive coping and adjustment.

Discussion

We reviewed the available research reporting effective psychosocial preparation provided to the child and family prior to amputation searching for common themes and recommendations and found that conclusions were primarily based on clinical, theory-driven opinion, rather than empirical data. While the limited number of available studies do not provide data or specific empirical recommendations based on child age or emotional or cognitive development, suggestions for pre-operative intervention are available and fall into five broad categories: Interdisciplinary and family communication, psychosocial screening and assessment, preoperative education, child interventions, and family interventions.

More specific research related to how preparatory interventions can be adapted to different developmental stages of childhood and adolescence is needed. Future directions for this developmental framework should also be expanded to include empirical research on the entire family unit, as well as the individual patient. Family variables were often noted as factors with assumed influences on the child's coping and adjustment processes and parents were commonly included in clinical recommendations about how and when to inform the

child once amputation becomes necessary - However, these factors were often described anecdotally or through clinical suggestions without empirical support. Given the intensity of the physical and emotional adjustment that follows an amputation in the context of the existing dependency of childhood and adolescence, future research that specifically focuses on or includes interventions designed for family members of children undergoing amputations due to cancer will be a crucial component in addressing the needs of this population. Due to the low numbers of new cancer related amputations, there is a role for both prospective as well as retrospective qualitative and quantitative research, and single case studies, which may be more feasible than randomized controlled studies. The findings demonstrate the need for research that will lead to evidence based recommendations of disease-specific psychosocial interventions in this vulnerable pediatric population.

Limitations of our findings include drawing from a literature base that is sparse and outdated. Available studies include small sample sizes and lack quantitative, empirical methodology. Additional limitations include the intentional, but specific focus on amputations due to cancer. While an amputation due to cancer presents with inherently specific psychosocial considerations that must be taken into account when engaging in clinical work, there may be other similar populations that merit consideration for future research that may benefit oncology patients. For example, there are a number of preparatory intervention techniques, including peer modeling and medical play, that have been highlighted in more general subsets of pediatric and adolescent patients. Additionally, although it is important to recognize the significant differences between the two groups, children with traumatic limb amputations also suffer similar post-surgical physical limitations as children with cancerous amputations and report psychological issues as well, including depression and anxiety (Khan et al., 2016). It could be beneficial to examine the literature focused on this larger population to supplement the relatively scarce research on cancer patients. While the timeframe for traumatic amputations cannot offer insight into pre-operative intervention, there may be useful existing research on ways to improve postoperative interventions and support for the entire rehabilitation process.

Conclusions

To the best of our knowledge, this is the first review of the literature examining preparation for amputation in children and adolescents living with cancer. Amputations during pediatric cancer treatment present with unique challenges and must be viewed as a distinct subset of cases that necessitate targeted psychosocial care and intervention. This review demonstrates that despite preparatory psychosocial involvement being commonly acknowledged as crucial in enhancing adjustment in pediatric and adolescent oncology patients undergoing amputation, the current literature base is inadequate in describing and informing specific preparatory interventions aimed to support these pediatric patients and facilitate positive coping and adjustment. Recommendations to include preparatory interventions including pre-operative education, film and/or peer modeling, play therapy, hypnosis, relaxation training, and coping skills development would benefit from further study and may significantly enhance the coping capacities and long term adjustment of youth undergoing amputation.

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Figure 1.
PRISMA Diagram

Table 1

Characteristics of studies included for review (N = 5)

Study	Purpose	Method	Type	Sample	Results	Psychosocial Recommendations
Atala & Carter (1992)	To describe the factors that may impact coping and adjustment in pediatric limb amputation, as well as a variety of intervention strategies. A brief case study is included.	Review paper and case study	Review/Case	N=1 pediatric patient (age=10)	Adjustment to an amputation is influenced by many factors, including social, cognitive, developmental variables, as well as the child's medical history and planned procedure. Pre-operative therapeutic interventions were effective in reducing anxiety and improving adjustment for a cancer-related amputation.	Psychosocial approaches, including play modalities and CBT, should be offered pre- and post-operatively and tailored to meet the patient's developmental and medical circumstances. Medical and surgical teams should be closely involved.
Clerici et al. (2004)	To describe the reactions of 5 case studies of pediatric patients undergoing amputation for malignancy and the support provided by the multidisciplinary team.	Case studies	Review/Case	N = 22 candidates for amputations (N = 5 recorded case studies); ages 12–17	Patients exhibited varying, but strong emotional reactions to amputation, but the surgery was more easily accepted if the affected limb was causing pain.	Preparatory psychosocial support should be available to patients and families to help facilitate optimal recovery. This support should include multidisciplinary teams that include members with specific psychological training.
Ferrari et al. (2002)	To use anecdotal case studies of pediatric patients requiring amputations due to cancer to illustrate the emotional impact of the procedure and the clinical support used throughout treatment	Case Studies (Transcribed board conference)	Case	N=3 adolescent patients (ages 13–17)	Psychosocial support and intervention can address defense mechanisms to improve quality of life and functional adjustment. The psychological impact of pediatric amputation due to cancer is challenging to assess quantitatively.	Staff with psychological training should be utilized to support patient adjustment. Oncologists should ensure good communication between the medical team and family.
Jaraway et al. (2013)	To retrospectively evaluate the effectiveness of a preparatory intervention designed to allow parents to view their child in the operating room following amputation, but prior to reversing anesthesia.	Structured interviews	Quantitative	N=22 adult parents of pediatric patients (patient ages 6–16 at time of surgery)	Parents (N=16; 72%) who chose to see their child in the operating room prior to reversing anesthesia reported this as a positive experience and valuable in helping them cope with their child's amputation.	All caregivers of patients undergoing amputations should be given the opportunity to view their child while in the operating room prior to reversing anesthesia.
Tebbi, Perrilli, & Richar ds (1989)	To examine the psychosocial and functional adjustment of adolescents who lost a limb to cancer	Structured interviews	Quantitative	N=33; ages 10–20	Participants report good overall adjustment after their surgery, but identified multiple areas of concern. The most commonly reported categories of dysfunction and impaired adjustment following amputation surgery were in academic, workplace, and social environments.	Pre-surgical psycho-education about the physical and emotional effects of amputation should be provided to all patients, family, and peers, as well as to medical staff to improve social and physical adjustment. Tutoring should also be offered during hospitalization to ease the patient's transition back to school.