

Chapter 1

Introduction



INTRODUCTION

Case

We present Marian, 13-years-old, a survivor of retinoblastoma in infancy and recently diagnosed with rhabdomyosarcoma. Treatment (consisting of surgery, radiotherapy and chemotherapy) started shortly after her start at high school. Unfortunately, due to her high absence rate, Marian did not make adequate adjustment at her new school, or with her peers. Shortly after the end of her treatment, new friendships were yet to be made; this had a deleterious effect on both her mood and sense of well-being. In addition, because her physical condition was low compared to that of her peers, she was less able to participate in (extracurricular) school activities. She wonders if she will ever be able to lead the life of a normal adolescent.

General

In the Netherlands, every year about 650 children are diagnosed with childhood cancer. Overall, in the last decades an increasing number of children with cancer survive; nowadays, in the Netherlands, this is about 75%. Since the survival rates are increasing, more research is focusing on the (prevention of) late treatment-related complications such as second neoplasms and organ dysfunction, as well as on psychosocial and cognitive problems, i.e. the so-called late effects. These late effects can have substantial impact on the child's physical and psychosocial functioning. Research has shown that about 75% of childhood cancer survivors experience one or more late adverse health effects after a median follow-up of 17 years [1]. One of the late effects of chemotherapy and/or radiotherapy is impaired physical fitness; children often present muscle weakness and/or low cardiorespiratory fitness during and after treatment [2-6]. Reduced physical fitness on the long term may lead to fatigue, obesity and a diminished psychosocial functioning [7-13].

Childhood cancer patients and survivors are at risk of developing depressive symptoms and other psychosocial problems [14-18], and report lower levels of health-related quality of life (HrQoL). Childhood cancer patients are especially at risk during their treatment [19-20]. Psychosocial problems and lowered HrQoL of survivors can be a direct late effect of the disease and treatment, but can also indirectly be related to physical late effects.

Health-related Quality of Life

The World Health Organization (WHO) considers HrQoL to be a multidimensional concept including physical, social, cognitive and emotional health [21]. Childhood cancer impacts both the physical and psychosocial domains of HrQoL. During their treatment for cancer, children generally score their HrQoL as lower than that of the norm; for example, over 30% of the patients scored below -2 SD of the norm on most domains [20]. These scores tend to improve after the end of treatment and, as survivors, they generally reach the norm on most domains [20]. However, there are some established risk factors for diminished HrQoL during and after treatment, i.e. being newly diagnosed, diagnosed with acute lymphoblastic leukemia (ALL), female gender, high-risk treatment protocols, older age, more medical complications, and the development of posttraumatic stress disorder [19]. Poor physical HrQoL was more often reported by patients with low self-esteem, by less highly educated patients and by female patients [22]. Therefore, interventions should focus on the causes of HrQoL deficits; for this, a holistic and multidisciplinary approach on all domains (physical, social, cognitive and emotional) is advocated. This includes psychosocial interventions as well as other interventions, such as physical and occupational therapy [19].

In general, self-reported HrQoL of children tends to differ from parent-proxy reports. Among chronically-ill children the parents generally rate their child's HrQoL as being lower than that reported by the children themselves. In contrast, parents of healthy children rate their child's HrQoL as being higher than reported by the children themselves. More similarities are found between the scores of adolescents and their parents [19]. In general, parents and children base their evaluation of HrQoL on different types of information: parents are better in reporting observable behavior (e.g. physical functioning) whereas children are better in reporting their internal states or feeling, e.g. emotional functioning [23]. During childhood cancer treatment, parents report both poor physical and psychosocial HrQoL for their children, whereas children score only their physical HrQoL as being poor compared to the norm. After the end of treatment children rate their HrQoL as being similar to that of the norm. However, parents tend to rate their children's psychosocial HrQoL as poor compared to the norm after the end of treatment [20]. Because of the differences between self-report and parent-report, it is recommended to use both types of informants to evaluate HrQoL.

General psychosocial functioning

Despite the enormous impact of having (had) a life-threatening disease, most studies show that about 50-75% of childhood cancer patients and survivors report resilience and average levels of distress [24-27]. It appears that during their treatment the vast majority of patients is not more anxious or depressed than the healthy population [27-29]. However, the literature on survivorship shows contradictory results, with positive psychosocial outcomes for survivors of childhood cancer, e.g. showing resilience [30-31] or posttraumatic growth [32]. Also, more neutral outcomes for survivors of childhood cancer have been observed, e.g. they do not have significantly more anxiety, depressive symptoms and low self-esteem compared to the norm [33-34]. On the other hand, some studies show that survivors have more depressive, anxiety and posttraumatic stress symptoms than the general population [14, 17-18], and show more behavioral problems and problems in social functioning [24]. The physical consequences of the disease and its treatment could hamper social and personal functioning of childhood cancer survivors [34-35]. Among all the diagnoses, children with brain tumors and those with radiotherapy to the central nervous system are most at risk for later social and adaptation problems [24, 36]. Children with bone tumors are also more at risk for later adaptation problems [24, 37].

Psychosocial interventions

In clinical practice, psychosocial interventions are available for children with cancer, provided by a psychologist or psychotherapist. Different types of interventions are offered, such as counseling or support (individual or group), as well as psycho-educational interventions by providing verbal and/or written information or information on the Internet [38]. Other interventions focus more on skills training, e.g. social skills training [39] or cognitive-behavioral therapy [40]. However, little is known about the effect such interventions have on HrQoL and on the psychosocial functioning of the child with cancer. Until now, few studies are available on the effects of psychosocial interventions for childhood cancer patients. The available interventions seem promising in decreasing distress and promoting adjustments by the parents of children with cancer, but show only limited effects on the children themselves [41-42]. Attempts are now being made to make psychological interventions for childhood cancer patients and survivors available through the Internet and, so far, this seems feasible [43-44]. However, randomized controlled trials are necessary to examine the effectiveness of such interventions.

Physical interventions combined with psychosocial interventions

Since it is established that childhood cancer patients are at risk for developing physical fitness and psychosocial problems, there is currently more focus on the development of interventions to prevent such problems, or to diminish existing problems. In adult cancer patients, rehabilitation programs encompassing both physical exercise and psychosocial support have shown positive effects on physical fitness and HrQoL [45-47]. However, in childhood cancer patients only a few studies have examined the effect of physical exercise programs; moreover, all these studies have methodological limitations (e.g. a small study group) and/or did not include a psychosocial intervention [48-52]. To our knowledge no studies are available that have investigated a combined psychosocial and physical intervention.

Back to the case description

After her treatment Marian showed reduced physical fitness resulting in less energy to complete her school day and, after school, she had to lie down on the couch to recuperate. Consequently, although she returned to school on a part-time basis her physical problems still prevented her from leading the life of a normal teenager; this continued to have a detrimental effect on her mood.

The QLIM study and its rationale

Because there is a lack of evidence-based physical, psychosocial and combined interventions for childhood cancer patients, despite an increased risk for impaired physical fitness and psychosocial problems in childhood cancer survivors, we conducted the QLIM study. This study aimed to assess the effectiveness and feasibility of a combined physical and psychosocial intervention for childhood cancer patients (aged 8-18 years), during or within the first year after their treatment. It was hypothesized that improving the level of physical fitness will enhance the patient's psychosocial functioning; vice versa, improved psychosocial functioning is hypothesized to increase a young person's motivation and willingness to engage in sport activities. By offering a multidisciplinary intervention on two domains, i.e. the physical and psychosocial, we aimed to enhance HrQoL.

Structure of this thesis

The aim of this thesis is to investigate the psychosocial effects of a combined physical and psychosocial intervention for childhood cancer patients aged 8-18 years, and to examine the acceptability and adherence of the intervention.

This chapter, **Chapter 1**, presents a brief introduction to the theoretical background of this thesis.

Chapter 2 describes the study design and the intervention itself. The intervention is a 12-week physical (24 sessions) and psychosocial (6 sessions for children, 2 sessions for parents) training for childhood cancer patients (aged 8-18 years) during or within the first year after the end of their treatment.

Chapter 3 examines the factors that influence childhood cancer patients to participate (or not) in the combined QLIM intervention. The non-participants were asked to fill in a one-time questionnaire survey concerning their reasons for non-participation, and details about their HrQoL and any behavioral problems.

Because the psychosocial part of the intervention was not pilot tested before the start of the randomized controlled trial, **Chapter 4** describes the explorative study that was conducted to evaluate the applicability of the psychosocial intervention among childhood cancer patients.

Chapter 5 investigates the effects of the actual QLIM intervention on psychosocial functioning and HrQoL. Assessment of HrQoL, depressive symptoms, behavioral problems and self-esteem (by means of questionnaires) took place at baseline, after 4 months (shortly after the end of the intervention period), and at 12 months post- baseline.

Chapter 6 presents an assessment of the effects of the QLIM intervention on physical fitness. In addition, we conducted a process evaluation by examining the effective intervention components, as well as the applicability of and adherence to the QLIM intervention.

Finally, **Chapter 7** discusses the conclusions of this thesis and presents some implications for the care of childhood cancer patients and for future research.

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