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**MUSIC THERAPY - AN INTERVENTION  
EFFECTING QUALITY OF LIFE AND  
HEALTH IN CHILDREN GOING THROUGH  
HEMATOPOIETIC STEM CELL  
TRANSPLANTATION**

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# Music Therapy – an Intervention Effecting Quality of Life and Health in Children going through Hematopoietic Stem Cell Transplantation

## THESIS FOR DOCTORAL DEGREE (Ph.D.)

This thesis will be defended on Friday, June 14, 2019, at 10.00 am in Hall B 64, Karolinska University Hospital, Huddinge.

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“Happiness is neither virtue nor pleasure  
nor this thing nor that but simply growth.  
We are happy when we are growing.”

William Butler Yeats



## ABSTRACT

Hematopoietic stem cell transplantation (HSCT) is an established treatment for several types of leukemic, hematopoietic, and metabolic diseases. The child is isolated during 4-6 weeks, followed by extensive follow-up for 3-6 months. The huge psychological and physiological burden of HSCT requires careful supportive intervention. Music therapy can be applied in paediatric healthcare to help the child through difficult experiences and increase overall feelings of wellbeing. The aim of this thesis is to evaluate the experiences of music therapy in children undergoing HSCT. Music therapy intervention included both expressive and receptive methods. Choice was in focus, where the child could choose to play different musical instruments, sing and/or listen to music along with the music therapist with an option of parent and/or sibling participation.

**Study 1 and 2**, included a randomized clinical trial (RCT) of 38 children (age range 2 months to 17 years) randomized in two groups. The music therapy group received music therapy twice a week during inpatient treatment, whereas the control group received music therapy post-discharge.

**Study 1** included 24 patients, whose physiological parameters including blood pressure, heart rates and saturation were recorded morning and evening at intervention, twice a week for both music and control groups. The evening heart rate decreased significantly in the music therapy group compared to the control group ( $p < 0.001$ ), potentially indicating prevention of post-traumatic stress disorder (PTSD).

**Study 2** analysed 29 patients, where we compared health related quality of life (HRQoL) using the validated questionnaires PedsQL 4.0 generic core scales and PedsQL 3.0 cancer module. Questionnaires were issued at admission, discharge and 6 months follow up. In the music therapy group, an improvement of physical function was observed at time of discharge (adjusted  $p = 0.04$ ). The control group showed improved results in all domains of PedsQL 4.0 generic core scales after music therapy was initiated at 6 months follow up ( $p = 0.015$ ).

**Study 3** included six children and their parents, who had previously participated in the RCT. The aim was to explore the experiences of the interactive processes of children and parent during music therapy intervention. The data sampling method was a collaborative research method. An independent psychologist conducted the interviews with the children, parents and music therapist and performed the analysis. Three themes emerged; experiences of competency and recognition of self, interactive affect regulation as change potential, and importance of the therapeutic relationship.

**Study 4** was a qualitative focus group study, which included 7 members of the medical team. An independent psychologist facilitated the interviews, with analysis performed by the thesis author and an independent researcher. Emerging themes included the importance of music therapy, expressed both physically and mentally by the children and a sense of satisfaction in both child and family. Parents could choose either actively or non-actively participation, thus, providing the possibility of a rest period. The staff were an integral part of the treatment and music therapy addressed the children in an unsecure and isolated situation.

**Conclusion:** The lowered heart rate values 4-8 hours after music therapy in the intervention group as well as higher HRQoL estimations described by both groups suggests that music therapy can be a complementary, effective intervention during and after HSCT. Along with the support and dedication of the medical team, music therapy was found to be an important factor in managing the treatment period at the hospital. Our results suggest that music therapy should become an integrated part of the supportive care children undergoing HSCT.

## LIST OF SCIENTIFIC PAPERS

- I. Music therapy can lower the heart rates of severely sick children. **Uggla L\***, Bonde LO, Svahn BM, Remberger M, Wrangsjö B, Gustafsson B. *ACTA Paediatrica* 2016 Oct; 1225-30.
- II. Music therapy supported the health-related quality of life for children undergoing haematopoietic stem cell transplants. **Uggla L\***, Bonde LO, Hammar U, Wrangsjö B, Gustafsson B. *ACTA Paediatrica* 2018 Nov; 1986-1994.
- III. An Explorative Study of Qualities in Interactive Processes with Children and Their Parents in Music Therapy during and after Pediatric Hematopoietic Stem Cell Transplantation. **Uggla L\***, Mårtensson Blom K, Bonde LO, Gustafsson B, Wrangsjö B. *Medicines (Basel)* 2019 Feb; 6(1):28
- IV. Staff experiences from music therapy intervention for children undergoing hematopoietic stem cell transplantation. **Uggla L\***, Wrangsjö B, Bonde LO, Gustafsson B, Adolfsson A. *Manuscript*.



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## LIST OF ABBREVIATIONS

|                                   |  |
|-----------------------------------|--|
| ALPS II                           | Astrid Lindgren Children's Hospital Pain Scale               |
| AYAs                              | Young Adults   |
| BCE                               | Before Common Era  |
| CAST                              | Cell Therapy and Allogeneic Stem cell Transplantation        |
| CB                                | Cord Blood   |
| CE                                | Common Era   |
| CMV                               | Cytomegalovirus Infection                                    |
| FMS                               | Förbundet för Musikterapi i Sverige                          |
| GVHD                              | Graft-Versus-Host Disease                                    |
| HLA                               | Human Leukocyte Antigen                                      |
| HSCT                              | Hematopoietic Stem Cell Transplantation                      |
| HRQoL                             | Health-Related Quality of Life                               |
| MUD                               | Matched Unrelated Donor                                      |
| NICU                              | Neonatal Intensive Care Unit                                 |
| PedsQL 3.0<br>cancer module       | Paediatric Quality of Life Inventory 3.0 cancer module       |
| PedsQL 4.0<br>generic core scales | Paediatric Quality of Life Inventory 4.0 generic core scales |
| PRO                               | Patient Reported Outcomes                                    |
| PROM                              | Patient Reported Outcomes Measures                           |
| PTSD                              | Post-Traumatic Stress Disorder                               |
| PTS                               | Post-Traumatic Stress  |
| PTSS                              | Post-Traumatic Stress Symptoms                               |
| RCT                               | Randomised Controlled Study                                  |
| STS                               | Secondary Traumatic Stress                                   |
| TSS                               | Traumatic Stress Symptoms                                    |
| TMV                               | Therapeutic Music Video                                      |
| VAS                               | Visual Analogue Scale  |
| WFMT                              | World Federation of Music Therapy                            |



# 1 PRELUDIUM

*When standing in between the hallway and the hospital room I plan to knock on the door. "Should we meet today?" The sick child is in the room with the parent, sometimes a sibling is present. Since the child has no immune protection, I bring disinfected music instruments. Just before knocking, I compose myself, prepare myself in my mind, as I open my inner room.*

*Once in the room, we locate a place the child thinks is best and prepare the space together; it may be by the bed, on the floor or around a table. The most important is to create a safe place and a safe space. If the child is young, we start with a song, then I ask the question, "what do you think we should do today?" Now the moment is here, the moment when the child makes a break, sometimes short, to have an inspiration, an intuition of what attracts among the instruments and other possibilities.*

*The child doesn't need knowledge, but needs to feel confident with me, to dare to take the step into "the unknown" as the initiative brings. Sometimes you are aware of some hesitation, maybe resistance, but also desire and curiosity. The child needs to take an initiative that entitles me to be in the room. The child has the possibility to show her/himself, to take the leap into the unknown, leaving her/himself a moment to venture into the opportunities that music, interaction, playing, singing, improvisation, instruments, moving, creating or just listening to music together, offers. In that moment the child also needs to be aware of that we are there together, that I am there as a traveling companion, a guide and witness.*

*Sometimes the feeling to venture into something unknown will return several times during the session and the space of security and stability needs to be maintained. Sometimes it seems easier to go all in and in a way "give up ourselves" in reason to meet, and after the interplay, come back to ourselves, changed.*

*For how long will our meeting last? Even the ending point is something that may occur between us in the room. Often it is obvious when it is time to end the session; the child might show some small changes in the appearance. It is important to end the session before the child feels tired. Mostly something important has happened and it is time to finish the session. I ask the child if there is anything more we have to take care of, perhaps another song to sing before it is time for me to leave. To the young child a closing song is a way to end and can be experienced as a gateway back to the known.*

## 2 INTRODUCTION

This thesis is an interdisciplinary investigation concerning music therapy for children undergoing hematopoietic stem cell transplantation (HSCT). The disciplines of music therapy and HSCT appeared almost simultaneously in history in close connection with World War II. The practice of music as a healing medium is far older and has been described multiple times throughout history and in numerous cultures (1). Music therapy became established as an academic discipline where it was applied in the USA for the treatment of war veterans (2). Simultaneously, the field of stem cell transplantation began to develop as medical researchers tried to find novel ways to reinstate bone marrow function in radiation associated aplasia as a result of the atomic bomb (3). Decades of research finally culminated in the first successful paediatric HSCT that took place in 1968 (4, 5). Since then, the rate of allogeneic transplantations, has shown a steady global increase with the latest reports documenting 3725 children between the ages of 0-18 years of having undergone HSCT in Europe and associated countries (6). In Sweden, approximately 50 children undergo HSCT per annum as a treatment for serious haematological disorders, however the invasive nature of HSCT treatment requires the application of support measures that can alleviate stress and instil wellbeing (data collected from the Centuri register).

The aim of this doctoral study was to investigate the effects of music therapy on children who underwent HSCT. In our studies, we aim to give a broader understanding of the effects of music therapy and answer the following questions:

- How can we measure the effects of music therapy?
- What kind of measuring tools can we use to identify what might happen inside the child in connection with music therapy?

To evaluate the phenomenon, four different perspectives were chosen in order to document changes in conjunction to the music therapy intervention.

- Objective endpoints: heart rate, blood pressure and saturation.
- Subjective endpoints: evaluation of pain, mood and health related quality of life (HRQoL).
- Qualitative interview with children and parents.
- Focus interviews with members of the staff.

## 3 BACKGROUND

### 3.1 BACKGROUND 1: MUSIC THERAPY

#### 3.1.1 Historical perspectives

Music as a therapy and a healing medium for the mind and the body has a long and diverse history, with records spanning thousands of years and several ancient cultures (7, 8). In frescos from 4000 BCE the contextual use of music for healing is shown (9) and the use of a harp by David to soothe the troubled mind of King Saul is a well-known narrative from the Bible (1 Samuel 16:16-23).

The concept of music as therapy is grounded in the teachings of Aristotle 384-322 BCE and Plato 428-347 BCE where they both refer to the healing effects of music (10). The “Father of medicine” Hippocrates 460-370 BCE had a holistic health care philosophy and used music as a treatment for mental illnesses. The hypotheses in ancient Greece was that music healed the soul of the human being, which also affected the body and that specific diseases could be treated and cured with selected musical presentations (11). The theory of Hippocrates was that music balanced all four humours (blood, black bile, yellow bile and phlegm) of the body and could alter emotions, mood, temperament and behaviour (10, 12).

A thousand years later Boethius, approx. 480-524 CE, redrafted elements from Pythagoras 570-495 BCE, (who had previously discovered that the sound of a tone is in relation to the length of the string) in *De Institutione Musica*. Boethius divided the music in three levels, *mundana*, *humana* and *instrumentalis*. Later on, during the medieval period, the use of different scales to produce different psychological effects was widespread (13). In the early 19<sup>th</sup> century two medical dissertations concerning the therapeutic importance of music were written; E. Atlee, 1804 : “An inaugural essay on the influence of music in the cure of diseases” (14) and S. Mathews, 1806: “Effects of music in curing and palliating diseases”(15).

Music therapy, as we know it today, started in Britain and USA after World War I as a means to treat traumatized war veterans (13). The first course in Music Therapy was offered in 1919 at Colombia University; however, it would take until the 1940s before music therapy, including education and training, emerged as a discipline in its own right in the United States (2, 16). In the following years, several important music therapy associations were founded. In Sweden the concept music therapy has been in use since the 1950s, with the association “Förbundet för musikterapi Sverige” (FMS) grounded 1974 (17) and the availability of education programs available at Royal College of Music in Stockholm since 1981 (18). The last 15 years in particular have been fruitful with several

doctoral theses concerning music therapy in different contexts presented by Swedish music therapists (18-24).

### **3.1.2 Music therapy – cross disciplinary**

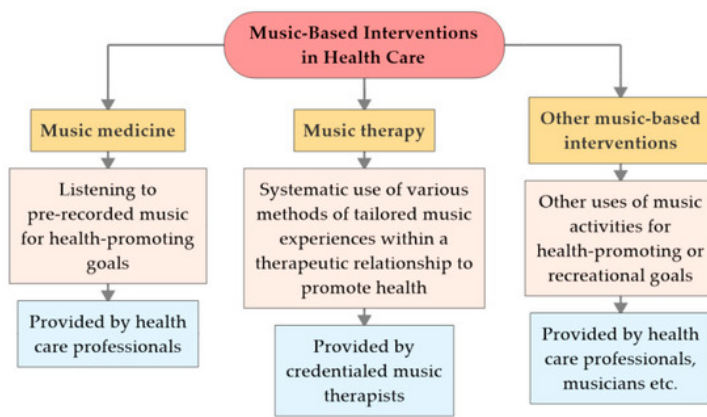
Experiences of music can be strong and life transforming (25) and the research field of music and health is rapidly expanding (26-28). Similar to other therapeutic methods, music therapy derives its conceptualization from various theoretical standpoints. Music therapy includes aspects derived from psychology, social science, cultural, biological and the neuroscience of how music affects us and how this can be utilized. Music therapy today is a global and a well-recognized form of therapy both in clinical and academic settings that has shown continuous progression and development (29). The foundation of music therapy is to facilitate interpersonal meetings via music, to play, sing, improvise, create, move, and listen with the music therapist in a shared experience. Music therapy can take the form of different models and intervention style (27). The aim of music therapy is to facilitate the interplay, both with and without words, enabling support and strengthening internal resources that contribute to wellbeing and change (17).

The Swedish music journalist, Eric Schüldt once asked the Swedish composer, Jan Sandström, if music can help in difficult times to which he responded: "Yes, but it is pretty simple, it is like if music places a warm hand on my shoulder and says: It's ok, it's going to be okay." (30) The power and relational aspects of music is manifested and can be heard in Schüldt's radio music program that reaches a high numbers of listeners each week.

### **3.1.3 Musical interventions**

In the context of health care different music-based interventions may be offered (Figure 1). **Music medicine** uses music to improve the patient's physical, mental or emotional status in connection with medical care (31, 32). This treatment involves passive listening to pre-recorded music. The music is often selected in advance and other professionals than music therapists, e.g. nurses handling the treatment; thus there is no direct interaction with a music therapist (33).





**Figure 1. Different Music-Based Interventions.** Stegemann *et al* (34).

**Music therapy** is practiced in various medical treatments where the object is to help the patient through challenging experiences and to improve well-being. **Other music-based interventions** include music activities, concerts and musicians visiting health care institutions.

Both music medicine and music therapy have shown proven outcomes; although in a meta-analysis concerning different interventions, Dileo reported that music therapy showed significantly greater effect sizes (35). In a study on adult cancer patients, Bradt reported that both music medicine and music therapy have benefits for the patient during cancer treatment. Listening to familiar and pre-recorded music in music medicine, improves the managing of symptoms whereas music therapy enables psychosocial support and support inner resources (36). A notable observation in this study is Bradt conclusion on the importance of patient accessibility to a music therapist during both kind of interventions.

### 3.1.4 Definition of Music Therapy

The definition of **Music therapy** according to the World Federation of Music Therapy (WFMT) is: “Music therapy is the professional use of music and its elements as an intervention in medical, educational, and everyday environments with individuals, groups, families, or communities who seek to optimize their quality of life and improve their physical, social, communicative, emotional, intellectual, and spiritual health and wellbeing. Research, practice, education, and clinical training in music therapy are based on professional standards according to cultural, social, and political contexts”.

This definition can be defined by four key characteristics

- professional use of music and musical elements.
- works with individuals, families and groups.
- improving physical and psychological wellbeing.
- research, practice and education based on professional standards.

## **3.2 BACKGROUND 2: HEMATOPOIETIC STEM CELL TRANSPLANTATION**

### **3.2.1 HSCT procedure**

HSCT is a highly specialized and advanced medical procedure where the goal is to replace the patient's own stem cells with new stem cells from a donor. HSCT is applied in the treatment of severe haematological malignancies, certain severe benign diseases or metabolic diseases. Due to the severe nature of the side effects and difficulties with the treatment, HSCT is generally applied if there are no other treatment options available (37). In HSCT the stem cells of the patient are replaced by healthy cells from a donor who may be related or unrelated. The child is initially conditioned with chemotherapy that is sometimes given in combination with radiation therapy. After completing conditioning therapy the stem cells, either derived from stimulated peripheral stem cells, bone marrow or cord blood are infused.

For a period of 1-3 weeks, until the donors cells are established in the patient's bone marrow the patient is aplastic and has an enhanced risk of life-threatening infections. Once the stem cells are established there is also the threat of severe graft-versus-host disease (GVHD), i.e., the donor T cells react against the foreign body. GVHD can cause treatment failure and become life threatening. Acute GVHD grade III-IV mainly effects the gastrointestinal tract, liver, lung and the immune system and the mortality risk within two years post-HSCT is 25-55 % (38, 39). Chronic GVHD causes problems in a broader range of organs and resembles symptoms of an autoimmune disease. Chronic GVHD may however have a reducing effect on relapse and improve survival in childhood leukaemia (40). In addition to the pronounced physical stress of HSCT, the child is also exposed to mental strain. The isolation imposed due to the child's susceptibility to infections leads to reduced contact with friends and other family members. Even after HSCT, they are often required to stay in hospital for extended periods, as a result of severe GVHD, infections or treatment for suspected relapse (41-44).

### **3.2.2 Physiological complications of HSCT for the children**

Paediatric care has developed dramatically over the past 40 years. In the Swedish paediatric health care system, parents are viewed as an integral part of the treatment and play a key role in supporting the child during the process. The HSCT treatment is very intense for a period of approximately 3-6 months, where there is a high risk for severe infections, reactivations of viral infections, GVHD, and relapse. The medical treatment includes various interventions, such as antiviral therapy, anti-GVHD treatment (45), and treatment for relapse. Long term there is also an increased risk of chronic GVHD-effects on tissues and organs such as the eyes, gastrointestinal tract, skeletal system and immune system.

There is also a high risk of future infertility, and the child is regularly checked during adolescence and until adulthood (44). In a recent study, 88.9% of the children receiving allogeneic HSCT were reported to have early or late complications, such as viral reactivation, severe bacterial or fungal infections. Pulmonary insufficiency and impaired respiratory function were also described in children in both acute GVHD and chronic GVHD (46).

The recent developments in HSCT and the development of strategies in supportive care have made improvements in the outcomes for both adult and children (47, 48). The main causes of death are in particular the recurrence of primary diseases, infections, organ failure or GVHD (49). The mortality rate differs between different diseases and diagnoses, with the origin of donor stem cells a key indicator in predicting survival rates. The most preferable donor source is a human leukocyte antigen (HLA)-matched sibling donor, but this source is only possible for approximately 30% of HSCT patients. For the remaining 70% potential donors are either matched unrelated donors (MUD) or haploidentical donors. The sources of stem cells typically used are either bone marrow, stimulated peripheral stem cells or cord blood (CB) (50). Recently there has been a drop in the rates of in-hospital mortality in the non-malignant group of stem cell recipients; however there has also been a notable increase in cytomegalovirus infections (CMV) and adenovirus associated infections (51). Long-term side-effects include an increased risk of secondary cancer and overall general worry and stress of what may happen in the future (37). Other long-term physical effects that may affect quality of life include chronic GVHD, neurological dysfunction (52), neuropsychological effects (53), and various endocrine disorders, including infertility (54, 55).

### **3.2.3 Psychological complications of HSCT for the children, parents and siblings**

Although the entire family is affected in the acute phase, the negative consequences of HSCT can persist in the long term. A Danish study reported that in the acute phase parents have described numerous interaction problems, either with each other, with the affected child, towards other children in the family or with the nurses supervising the HSCT. These feelings of isolation resulted in reduced contact with other adults, inappropriate focusing on the wellbeing of the child e.g. obsessing over lab results and the incapability of leaving the hospital room (56). Paediatric survivors, of more than five years post HSCT also reported lower levels of physical health, disturbed partner relations and reduced sexual function compared to the general population (57). A study with adolescents and young adults (AYAs) at least 3 years after paediatric HSCT reported themes such as the persistence of

physical consequences including affected self-images, social withdrawal, sense of lack of choice and the need for special attention (58).

### 3.2.3.1 *Health related quality of life (HRQoL)*

In recent years, health care research and the patient's perspective on the treatment has gained particular attention. Patient Reported Outcomes Measures (PROM) are tools for measuring Patient Reported Outcomes (PRO) and include dimensions such as functional status and health related quality of life (HRQoL) (59). HRQoL refers to a multidimensional evaluation of the individual's estimations of how the disease and treatment affect the sense of overall functioning and wellbeing (60).

A study from 2016 comparing adolescent cancer survivors with healthy controls reveals significant differences in HRQoL. Cancer survivors have fewer years in school, less social support, but conversely also have better quality of life, more positive outlook and increased awareness of health problems. Risk factors for lower HRQoL in the group of cancer survivors are e.g. female gender, haematological disorders and HSCT (61, 62).

For children and adolescents going through HSCT, the intensive medical treatment affects the HRQoL of both child, parents and siblings (63). For the child, the HRQoL is reduced pre-transplant and even more compromised during conditioning therapy with the lowest ratings typically observed between 1 month and 3 months post HSCT but is improved 4 to 12 months post HSCT (64-68). In one study the survivors of HSCT had the same or better HRQoL 6 months to 8 years after HSCT compared to normal population (64). There are quite few studies investigating long term effects for HSCT survivors (69). Although, Reinfjell *et al*, concluded in a review, including studies reporting 5 years follow up or longer, that HRQoL is impacted in the longer term. Risk factors for lower HRQoL are severe chronic medical condition, GVHD or chronic pain (70). However, it generally takes approximately between 1-3 years to return the same HRQoL level that existed before the onset of HSCT (64, 71, 72).

### 3.2.3.2 *Traumatic experience*

Children with cancer are a well-studied cohort at increased risk of post-traumatic stress disorder (PTSD) (73, 74) and require supportive intervention (75). Graf *et al* studied young children with cancer (ages 8-48 month) 15 months after diagnosis and reported rates of 18,8 % for full PTSD and 41,7% for partial PTSD (76). Children on active treatment (77) or those who just received a cancer diagnosis have higher occasions of post-traumatic stress symptoms (PTSS) (78). In addition children who had previously faced recurrent and severe stressful life events had increased PTSS in relation to the cancer experience (79).

One study, focused on children with haematological disorders described difficulties in expressing emotions after medical treatment indicating PTSD symptoms such as avoidance and emotional numbing (80). The importance of interventions directed to reduce the levels of distress during the acute phase of the transplant process was also reported (81). Among the numerous psychological reactions in HSCT survivors, traumatic stress symptoms (TSS) and PTSD have been described (82). The diagnosis of PTSD includes both psychological and physiological symptoms such as intrusions, avoidance including, emotional numbness, mood alteration and hyperarousal. Symptoms may occur and persist one month or more after exposure to a traumatic event. For diagnosis the child or adult needs to show one or more symptoms from four main clusters: intrusions, avoidance, negative alterations in cognitions and mood and alterations in arousal and reactivity (83) and the symptoms create distress or functional impairment.

It is not only the severely ill child that shows PTSD symptoms, parents may also display symptoms of PTSD, with multiple research showing a direct correlation between parental PTSD and the incidence and severity of PTSD in the child (79). Parents of paediatric cancer patients report PTSS several years after the completion of treatment (84). In a study comparing symptoms of stress through disease groups, TSS may be more significant for parents of paediatric transplantations (85). PTSD is a greater risk for caregivers of children going through HSCT than either anxiety or depression (86). A group of parents show ongoing levels of high distress several years after HSCT (87), with one third of the mothers of children who survived HSCT in a state of persistent distress (88). The siblings of the child going through HSCT are also affected with approx. 30 % of siblings experiencing moderate to severe post-traumatic stress (PTS) (89). Non-donor siblings presented with significantly higher rates of school problems with donor siblings describing higher levels of anxiety, lower self-esteem and psychological distress compared to the non-donors (89, 90). Donor siblings of successful transplants with no complications typically expressed positive experiences, whereas donor siblings of successful transplants with complications felt responsible for the outcome and finally sibling donors of transplants that resulted in death, typically expressed feelings of blame, guilt and anger (91).

Phipps *et al* discusses the relevance of using the PTS model in the group of children with cancer since levels of PTSS are associated with adaptive style (92), and claims that the PTS model is more appropriately applied to the parents of paediatric cancer patients. In a study comparing traumatic events between children with cancer and healthy peers, 52.6% of the paediatric cancer patients identified cancer as a traumatic event, declining to 50% 5 years after diagnosis. The study concludes that there were no

differences between the cancer survivors and healthy peers regarding PTSS, also worth noting was the reported psychological growth in the group of children with cancer (93).

#### *3.2.3.3 Traumatic growth*

Paediatric cancer and the experience of HSCT may be considered as a family disease where the long-term effects involve the entire family (94). Traumatic experiences may not only trigger negative experiences. Struggles with psychological difficulties could result in posttraumatic growth (PTG). The theory behind PTG is that a trauma shatters the world assumptions of an individual, thus the trauma survivor has the experience of the need to rebuild his or her life world. This need could bring growth and experiences of functioning on a higher level (95). A person who experience PTSD as a result of trauma may at the same time experience PTG (96). One year after cancer treatment a majority of the adolescents survivors and their parents reported PTG (97). Life threat and an increased treatment severity perception was related to PTG. Increased PTG following a trauma was also associated with decreasing levels of PTSD over the long term.

#### **3.2.4 Elevating heart rate and PTSD**

Elevating heart rate coupled to stress is a well documented phenomenon, where the heart rate can serve as a vital indication of anxiety and arousal and appears to robustly predict posttraumatic stress or PTSD (98-101). A systematic review, including 5186 individuals, concludes that higher heart rate predicts PTSD symptoms, with a small effect size, for the younger population (102).

#### **3.2.5 Staff working in the health care**

Working with severely ill patients, in pediatric and HCST contexts can be challenging for health care workers. The work may bring meaning in life and personal growth, but can also be coupled to secondary traumatic stress (STS) or compassion fatigue (103). STS is defined as emotions and behaviors that originate from other individuals' experiences of a traumatic event (104-108). Previous research has reported that 50% of pediatric nurses were affected or displayed symptoms of secondary stress (109) with an a general recognition in the rate of underestimation of PTSD in physicians (108).

### **3.3 MUSIC THERAPY RESEARCH, A BRIEF OVERVIEW**

The research of music therapy has expanded since 1945 and today 55,125 articles can be accessed on the web site "KI research". The first article including paediatric music therapy

was published 1946 by Chenoweth, R. “Music as therapy for convalescent children” (110). Music therapy is currently playing an increased role within the health care system in many countries and has shown effect in different clinical, therapeutic and medical settings.

### **3.3.1 Children**

From studies in music therapy used in the neonatal intensive care unit (NICU), a meta study found that music therapy has effect on infant respiratory rate and decreased maternal anxiety (111). Improved cardiac and respiratory function, increased feeding manners, extended periods of quiet-alert states, supported bonding and decreased stress symptoms in parents have also been reported (112). The musical experiences in music therapy may support brain development in preterm infants (113) and parental infant-directed singing may alter the experience of pain for both the infant and the parent (114). For children with autism spectrum disorder, music therapy can help children in improving their skills in e.g. social interaction, verbal communication, initiating behaviour and social emotional mutuality (115). The relationship that the autistic child and the music therapist develop is an important predictor for the improvement of social skills, communication and language (116). Benefits of music therapy have also been described in children with cystic fibrosis (117), congenital heart disease (118), and in regions of conflict (119). Music therapy is valued for the family in paediatric palliative care (120), and as a well-functioning tool to meet different needs for holistic well-being (121).

### **3.3.2 Paediatric cancer**

Previous research has shown the positive effects of music therapy in the area of paediatric cancer (122), where music activities can help the child to become more social and active (123). Music medicine has been reported to decrease pain scores, lower heart and respiratory rates and reduce anxiety in children with cancer undergoing lumbar puncture (124) and in children with leukaemia in an outpatient setting (125). Music-based activities have been shown to provide a degree of comfort and encouragement in paediatric cancer patients (126) and the process of music therapy CD creation for children undergoing radiation therapy was reported as an engaging and developmentally applicable intervention that offered effective coping strategies (127). The provision of music therapy in an outpatient setting provide the child and family with resources of communication, self-expression and creativity (128). A study comparing different hospital activities showed that music therapy stimulated more engaging behaviour in the children (129), with active music engagement intervention supporting coping-related behaviours in children with cancer (130). Several studies have reported on the music therapy experiences from the parental perspective, where family bonding was seen as the most important factor (131), followed

by improved communication and expression (132) and an increase perception of positive experiences despite the severe challenges (133).

### **3.3.3 The adult population**

A recent Cochrane review of music interventions, encompassing 52 trials with 3731 participants concluded that music intervention may affect anxiety, fatigue, pain and Quality of life (QoL) (134). Music therapy for patients undergoing HSCT was shown to significantly improve mood, reduce anxiety and relieve pain (135). Adult patients undergoing autologous stem cell transplantation reported reduced mood disturbances following music therapy (136). The use of music therapy has shown positive outcomes in the treatment of diverse diseases and conditions including, schizophrenia (137), Parkinson disease (138), cardiovascular parameters (139), dementia (140), mental health (141), depression (142, 143), PTSD in female military veterans (144) and palliative care (145-147).

## **3.4 MUSIC THERAPY RESEARCH IN PAEDIATRIC HSCT**

### **3.4.1 Children and young adult perspectives**

Previous research has shown the effect of music therapy on children and young adults (AYAs) undergoing HSCT. Robb *et al*, Burns *et al* and Sahler and colleagues (148-152) have performed research in this area. The most common strategy involves patients receiving 2 sessions a week for a total of six sessions, where all sessions are coordinated by a licensed music therapist. The interventions include song writing and digital video production, music therapy with relaxation imagery and therapeutic music video interventions.



**Table 1****Performed scientific studies concerning paediatric hematopoietic stem cell transplantation and music therapy studies 2003-2014.**

| Author              | Year | Age at HSCT                    | N   | Title  | Design/Method   | Results  | Intervention   |
|---------------------|------|--------------------------------|---|--|---|--|--|
| Robb <i>et al</i>   | 2003 | 9-17                           | 6   | Song writing and digital video production interventions for paediatric patients undergoing bone marrow transplantation, part I: an analysis of depression and anxiety levels according to phase treatment                        | Exploratory study, quantitative measures examining anxiety and depression levels agreeing to phase treatment. 3 patients in the music group, 3 in the no-music group. 2 sessions/week in 3 weeks. | Four participants (3 in the music group and 1 in the non-music group) experienced decreased anxiety after the majority of sessions.  | Song writing and digital video production.   |
| Robb <i>et al</i>   | 2003 | 9-17                           | 6   | Song writing and digital video production interventions for paediatric patients undergoing bone marrow transplantation, part II: an analysis of patient-generated songs and patient perceptions regarding intervention efficacy. | Exploratory study, qualitative outcomes of music therapy intervention. 3 patients in the music group, 3 in the no-music group. 2 sessions/week in 3 weeks.  | Content analysis of songs by the patient: hope, positive coping, appreciation, mental status, control, time, bewilderment, treatment, and diagnosis. Insight into each patient's experience was provided.  | Song writing and digital video production.   |
| Sahler <i>et al</i> | 2003 | 4 years or older, even adults. | 23 received treatment, 19 case control patients | The effect of using music therapy with relaxation imagery in the management of patients undergoing bone marrow transplantation: a pilot feasibility study.   | Case control study Pre/post music/relaxation pain and nausea using a VAS; determination of time-to-engraftment.   | Self-reported pain and nausea significantly reduced compared to ratings before the session. Time to engraftment was reduced in the treatment group (p<0.01).   | 45-minute music-assisted relaxation and relaxation imagery sessions, twice a week from enrolment to discharge. |
| Burns <i>et al</i>  | 2009 | AYAs 11-24                     | 12  | Exploring the Feasibility of a Therapeutic Music Video Intervention in Adolescents and Young Adults During Stem-Cell Transplantation   | Randomized, explorative study. T1 (baseline), time 2 (T2, postintervention), and time 3 (T3, 100 days post transplantation). 6 sessions in total, twice a week.                                   | At T2 positive trends for hope, spirituality, confidence/mastery, and self-transcendence. At T3, positive results: symptoms distress, defensive coping, spirituality, and self-transcendence and improvements in quality of life.  | Therapeutic music video TMV or Audiobook   |
| Robb <i>et al</i>   | 2014 | AYAs 11-24                     | 113   | Randomized clinical trial of therapeutic music video (TMV) intervention for resilience outcomes in adolescents/young adults undergoing hematopoietic stem cell transplant: a report from the Children's Oncology Group.          | Randomized clinical trial, 6 sessions over 3 weeks. Test points at baseline (T1), post treatment (T2) and after 100 days (T3).  | The TMV intervention improves courageous coping, social integration, and family environment. Significant better courageous coping, at T3, significant better social integration and family environment, no significant for spiritual perspective and self-transcendence. | TMV or low-dose control (audiobooks) group.  |

### **3.4.2 Parental perspective**

Music therapy for hospitalized children going through HSCT was perceived by the caregivers as an overall positive experience (153). Parental benefits from music therapy intervention include sharing positive emotional experiences, witnessing improved symptom distress, wellbeing (154) and as a shield for the challenges relating to HSCT (155).

## **3.5 MUSIC THERAPY IN PAEDIATRIC HSCT**

### **3.5.1 Music**

Humans are inherently musical beings. Music is introduced to humans early in life by different ways. Through pulse, rhythm, pitch and sound – it begins *in utero* as an interaction between the mother and the foetus and continues through life. These personal experiences supply us with our own relation to music that is as unique as a fingerprint. This musicality is strongly embedded in our brain and remains even after stressful conditions such as neurological trauma or harm (156). Music affects our emotions and has the possibility to engage and distract us from our surrounding (157-159). Music is social and communicative, affecting our behaviour as well as our identity (160-162). Music is also multi-layered and when we listen to music we interpret it in an unlimited number of ways (163). Music has been part of early human evolution where music and associated behaviours were important in supporting emotional interaction, social bonding and the development of cooperation and group dynamics (164).

### **3.5.2 Music and brain**

Music affects the whole brain. While the right hemisphere is usually described as the dominant side for music ability, the left-brain is more prominent in those with a high level of music ability (1). When we listen to music, the music travels from the cochlear nuclei to the brain stem and the cerebellum, moving up to auditory cortices in both sides of the brain (165). The cerebellum is involved in rhythm recognition and is activated when we listen to music. As part of its connection with the amygdala, it is wired into the limbic system, which regulates emotions and affects our autonomous nerve system by controlling vital functions such as heart rate and breathing (165). The effect of music on the brain is complex and involves numerous regions of the central nervous system. Listening to music activates memory centres in the hippocampus and the lower frontal lobe, reading music involves the visual cortex and recalling or listening to lyrics activates language centres in the temporal and frontal lobe. The action of playing music activates various centres depending on context, the frontal lobe (planning), motor cortex (co-ordination) and sensory

cortex (tactile feedback). The right hemisphere of the brain and its contacts with limbic and brain stem functions, is connected with controlling the emotional aspects of functioning (166), whereas the left hemisphere is more involved in cortical and cognitive function. Research focusing on music and neuroscience has rapidly expanded and shown that music has the possibility to improve reward, motivation, pleasure, immune function, social attachment as well as reducing stress and anxiety (167). The effect of music on stress levels has been shown to involve reduction in the levels of the stress hormone cortisol, which also regulates immune response (168).

### **3.5.3 Music and bonding**

We have an inherent human capacity to share attentions, experiences and actions through music. This capability makes it possible to share the lives of others on an existential level (169). The child discovers themselves in relationships with others (170), where their development and ability to interact has been investigated using video microanalyses of the interactions between infant and the mother (171, 172). In this first interaction involving wordless communication, a mutual creation of a melodic, rhythmic and dynamic dialogue between the infant and the caretaker is established. This opens the mind for music during the time of life when the baby lays the foundation for relating, creating and learning (172, 173).

### **3.5.4 Music and body**

Music therapy takes a holistic approach to humans involving body, mind and spirit. During the 20<sup>th</sup> century, several philosophers and physicians developed the term “the lived body”. “The lived body” is an intending entity, a living in relation to others, to things and to the surrounding environment. The severely sick child’s experience of control is greatly reduced while the perception of the body is altered. Being sick may also be understood as an “unhomelike being-in-the-world”, that can produce a feeling that the body is an alien, although it is still my body (174). This may entail a double sense of homelessness for the isolated child. According to Merleau-Ponty, we perceive the world with our bodies while we are our body and we perceive the world with our senses as a whole, not divided based on the different sensory organs (175). The body is both me and something else but also something in between me and the others/environment. The living body is able to simultaneously perceive and grip, an intertwining as Merleau-Ponty expressed it (176). An example is the case of a little baby boy, when he discovers that it is his moving hand, which makes the maracas create the sound that makes his whole body experience a nice shiver. This makes him want play the maracas all the time. Meeting his mother’s affirmative eyes,

is a moment of revelation when he understands that his mother also knows what he has just discovered.

### **3.5.5 Music and emotional regulation**

Emotional regulation is an important component of mental health, thus it is no surprise that emotional dysregulation is a major factor underlying many psychopathological clinical syndromes (177, 178). The parent's interactions with the infant is a powerful facilitator of emotional regulation, where the child builds internal working models in relation to the caregiver (179), and an implicit, unconscious knowing, regarding self, what you can expect from others and how interaction works (180). The concept "affect regulation" refers to strategies to decrease, maintain or increase affects and these strategies are learned at an early age (180). Examples of successful emotion regulation strategies are when you alter the way a certain situation is attended, you interpret the meaning of the situation or you actively change the situation (181). In the brain, emotional regulation is characterized by heightened activation in the anterior cingulate cortex, the orbitofrontal cortex and the lateral cortex resulting in a concomitant decrease in amygdala associated activity (182).

Music evokes and affects emotions (157), and music involvement can activate autonomous regulating systems via these emotions (183). People of different ages use music to balance and regulate emotions on a daily basis (184). The use of music as an emotion regulator is supported by behavioural and neural evidence involving the role of music in early infant-parent bonding and developmental fitness (185). Trehub *et al* show in a study of musical affect regulation that infants show more positive facial and vocal expressiveness and greater visual fixation during singing episodes compared to speech episodes (186). A systematic review (187) comprising 811 participants, aged 12-60 years, indicates that music experiences such as listening, singing and improvisation can impact emotion regulation by stimulating defined regions of the brain. This effect is music dependent and can vary depending on the nature of the music, personal preferences or previous exposure. For example, listening to minor, dissonant, sad or unpleasant music results in activation of the amygdala; familiar music or music making and singing stimulates the anterior cingulate cortex; preferred or detailed music stimulates the orbitofrontal cortex; and lastly familiar, preferred or active music making stimulates the lateral prefrontal cortex.

### **3.5.6 Music therapy and PTSD**

The music therapy profession is deep-seated and interweaved with the treatment of trauma, where historically it was used as a treatment for returning war veterans from the first and second world wars (16). Previous research has shown that people suffering from PTSD are particularly receptive to music therapy, with theoretical and empirical evidence supporting its

benefits (188). In the post-processing phase, a non-threatening medium such as music can stimulate traumatic memories. In the acute phase, traumatic stress leads to a reduced ability for self-expression and verbal expression (189). Music therapy is being used in the treatment of children with PTSD. Music can be viewed as a familiar or safe language for children, since they are exposed to the non-verbal features of music even while developing *in utero* (190). Additionally, music as a non-commanding medium is of importance for the children as they are less hindered in their expressions in music (191). In a study using music therapy for the treatment of PTSD in pre-school children, music had an active role in reducing feelings of vulnerability and increasing the resilience of children to previously experienced traumatic events (191).

### **3.5.7 Music therapy, a relational therapy**

Relational psychotherapies have interplay both in focus and as a starting point for achievements of understanding gained in therapeutic work (192). Music therapy is a relational (193) and art-based form of therapy, enabling intersubjective experiences through involvements and relating to music. Affective attunement through musical experiences allows the participants to take musical initiatives and affect interaction and interplay (193). The new-born child has an innate competence to communicate with others (172, 194). The ability to meet in intersubjective meetings develops at the end of the child's first year and continues through life (194). Intersubjectivity, grounded in theories of developmental psychology and phenomenology, is an interpersonal created and shared world of meaning (195); to share attention, intention and affective states. In music therapy, musical features such as rhythm, melody, movement and dynamic shifts gain and achieve experiences of intersubjective regulation (2). The shared intentions and affective states in music therapy, do not need to be translated into words, they are cross-modal and involve different sensory modalities (193).

To meet in a dialogue is to become involved, facing the other's whole person (196). In a life perspective, Ricoeur refers to the human being as *homo capax*, a capable human being, who has the ability to speak, act, narrate and be responsible, including the ability to motivate and prioritize his/her preferences (197). *Homo capax* also encompasses, both that the acting human being always is a suffering person, and simultaneously the suffering human being is still an acting person. To suffer, both psychological and physiological pain, is a reduction or destruction of the capacity to act, and this is experienced as an attack on the integrity of the self. However, from this perspective, suffering should not be viewed only as a shortcoming; instead, vulnerability may be recognized as a fundamental condition of life that makes us human, and opens up the person to others and the world (198). In accordance with Gadamer, Ricoeur uses the

concept *play (Spiel)* as a model for understanding the hermeneutic experience of transformation (*Verwandlung*), because whoever plays is also being “played.” This playful figure can be incorporated into music therapy as a model for the experience associated with musical interplay where the music itself, becomes the focus and not the individual participants. The music in this relational context is twofold, both an agent and a dialoguing tool (193) and reflects something true or real because of the nature of the play area, which also provides an opportunity for change (197). The participants are bonded through the music, sharing an experience, although not identical (193). Still, to be affected through musical interaction can be perceived as threatening to the self and it is therefore essential that music therapy in the HSCT context takes place within the child's window of tolerance (199) in order to avoid further traumatization. Window of tolerance is a concept expressing the personal space wherein you are awake, calm and safe enough to be curious, creative, social and learning (199).

### **3.5.8 Music therapy intervention during HSCT**

How can the contribution of music therapy to the healing process be understood when compared to music medicine? Undergoing HSCT is a strenuous and traumatic event. Since the child is endangered, the confidence in the parent is threatened (179, 200), and the child's need of support from an “evoked inner companion” (201) built on internalised experiences of being cared for by a self-regulating-other, (mainly attachment figures) may not be strong enough to regulate the trauma. In a session of music medicine there are two parties involved namely the listener and the music. However, in music therapy there are three constituents existing as a triad; the patient, the therapist and the music. The music therapy interaction can be visualized as a triangle where all three are mutually interconnected and a dynamic relation is possible. Additionally, each side of the music therapy triangle facilitates the relation between the other two (202). In music therapy with severely sick children, this mutual relation is essential, since it is the child's approach that allows the therapy to take place. In the music therapy interaction, security and intersubjectivity are re-created and in that atmosphere an attachment experience of trust may become stabilized thus allowing the music and the music therapist act like a substitute attachment support (203). The music therapeutic interaction has similar qualities to a situation where the child has experienced a functioning attachment to a parent (172, 179). This experience gives both an external support and strengthens the supportive and caretaking capacities of the child's “evoked inner companions”, which facilitates self-soothing and regulation of feelings of fear and abandonment. To benefit both child and the parent in maintaining emotional regulation, the music therapy setting needs to be open and stable in order to keep the affect level within the child's window of tolerance (204).

The universal need of recognition is followed by a paradox between our own independent will and the need that someone confirms and recognizes it (205). The possibility for the child to make its own choices during the music therapy interplay and for this to be confirmed is essential in this meaning. The familiarity in the musical experience is valued in context and vital to increase the therapeutic relationship and feelings of safety (187). The familiarity and prediction in musical appreciation is also connected to feelings of musical pleasure (206).

### **3.6 EPISTEMOLOGY**

In order to clarify the epistemology standpoints (207) in this thesis, the study belongs to two research traditions, namely natural science and human science. The natural sciences use hypothetical, casual methods (208), where hypotheses are tested against observations. In study 3 and 4 the overall methodology was hermeneutical phenomenology, interpreting texts statements as well as music and non-verbal communication (209).

#### **3.6.1 An inter-disciplinary work**

This thesis is an interdisciplinary study. Internationally, it is common to support interdisciplinary research since scientists and politicians are convinced of the merits of merging different disciplines. However, interdisciplinary projects are risk projects and it is difficult to evaluate the consequences and draw conclusions from failures. It is important to have supervisors with expert skills, internationally oriented quality assurance and that that the research is published in international journals (210).

## **4 AIMS**

### **4.1 GENERAL AIM**

The overall aim of this doctoral thesis was to investigate music therapy intervention from different perspectives in children 0-17 years of age undergoing HSCT.

### **4.2 SPECIFIC AIMS**

- To measure physical variables, analyze and compare between the music therapy group, receiving music therapy twice a week during HSCT and the control group, receiving supportive conventional treatment.
- To measure the subjective experiences concerning health and quality of life of the children after music therapy. We chose to compare self-documented and parent proxy questionnaires of the childrens HRQoL from admission, after discharge from the inpatient ward and at 6 months follow up in the music therapy group versus the control group. The music therapy group received music therapy between admission and discharge. Due to ethical concerns, the control group was offered the intervention after discharge. This made it possible to evaluate the effect of music therapy in both the short-term and long-term on the HRQoL for children undergoing HSCT.
- To explore the holding context and important parameters in the music therapy system, the subjective experiences and memories of the interactive processes during the music therapy session between the child, the parent and the music therapist.
- To explore the holding context in the psychosocial system, within the hospital team. The holding context is of importance in a traumatic situation. To investigate the attitudes and experiences from the staff working in the inpatient ward and in the day care unit towards patient centered music therapy.



### **4.3 HYPOTHESES**

#### Study 1 and 2

We formulated our hypothesis that music therapy could reduce anxiety, improve mood, support the mental health recovery and influence physical recovery after allogeneic HSCT.

#### Study 3

In study 3 our hypothesis was that important components and potential common threads could be identified in the interactions between child, parent and music therapist during the music therapy interventions.

#### Study 4

The hypothesis of study 4 was that music therapy could increase staff members understanding of the children's experience of their situation and the need to process their feelings.



## **5 METHODS AND PARTICIPANTS**

### **5.1 STUDY 1**

#### **5.1.1 Participants and setting**

Twenty-four children aged between two months and sixteen years of aged were included in the study. The children were randomized into two groups. The music therapy group involved thirteen children (including one dropout) and the control group comprised eleven children (including two dropouts). The study was carried out at Cell Therapy and Allogeneic Stem cell Transplantation (CAST) at Karolinska University Hospital in Huddinge, Sweden. The controls as well as the music therapy group were followed up as outpatients at the paediatric haematological ward at Astrid Lindgren Children's Hospital at Karolinska University Hospital in Huddinge.

#### **5.1.2 Intervention in study 1 and 2**

The music therapy intervention included both receptive and expressive parts. The first goal of the music therapy intervention was to create a safe and therapeutic alliance between the child and the therapist. The child was invited to take initiatives and to play on several musical instruments that were brought to the room. It was possible to sing, improvise and create music together with the music therapist, make songs or paint to music as well as moving or dancing to the music. If the child wished, parents or siblings could also participate. The aim of the music therapy was to be flexible, varied, person centred and to have the physiological and psychological well-being of the child in focus. The music therapy setting had the goal to support both the child and parent to stay emotional regulated and therefore the intervention was intended to provide a holding structure.

#### **5.1.3 Method**

A randomized clinical trial (RCT) was performed where the children in the music therapy group received music therapy twice a week during the in-patient treatment for approximately 4-6 weeks. Both the intervention group and the control group were offered conventional treatment as play therapy, clown visits, school and psychosocial support if needed. Music, as communication and expression, was only performed at the ward in the music therapy setting during the study period. In connection to the music therapy session in the music therapy group, physiological parameters as heart rates, blood pressure and saturation were measured in the morning and in the evening twice a week. These

parameters were retrieved from the medical protocols of the ward. These parameters were also sampled from the control group twice a week, morning and evening.

The children's disease severity was valued by the doctor every day by using the Lansky Play Performance Scale, where a 100 % score shows a perfectly healthy child. These parameters were documented as well as the nutritional status, weight and blood values twice a week in both groups.

#### **5.1.4 Statistical analysis**

Differences between the groups concerning the morning and evening measurements of blood pressure, heart rate, saturation and blood values were analysed. Continuous variables were compared by using the Mann-Whitney test or Wilcoxon's matched pair test.

## **5.2 STUDY 2**

### **5.2.1 Participants and setting**

In total thirty-eight children between the age of two months and seventeen years of age were enrolled in the RCT at CAST, eighteen in the music therapy group and twenty in the control group. The twenty-four patients from study 1 were also included in study 2. Two patients declined participation and two were excluded due to medical status, equally distributed between the groups. Later in the process two children died, one from each group, and three dropped out, one from the music therapy group and two from the control group. In total fourteen patients in the music therapy group respectively fifteen in the control group were analysed.

### **5.2.2 Method**

A RCT evaluating the self-reported HRQoL of the child and the parent proxy reported HRQoL at three times, at baseline, discharged and after six months. The international and valid questionnaires Paediatric Quality of Life Inventory 4.0 generic core scales (PedsQL 4.0 generic core scales) and Paediatric Quality of life Inventory 3.0 cancer module (PedsQL 3.0 cancer module) were used. Both groups answered the questionnaires at baseline, discharge and after six months. The music therapy group received music therapy between baseline and discharge, and the control group was offered the intervention after discharge. This enabled evaluation of both early and late intervention. At the start and end of each music therapy session during the inpatient ward a research nurse made a subjective evaluation of the mood of the child in the music therapy group on a five-point scale. At the same time, before and after each session, the child valued his/her pain, using the Visual

Analogue Scale (VAS) . In children younger than four, the parents estimated the pain of the child using the Astrid Lindgren Children's Hospital Pain Scale (ALPS II). The children in the control group met the research nurse twice a week during the inpatient ward, when estimations including mood evaluation by the nurse and self-reported and parent reported level of pain were documented. The doctor documented the Lansky play performance scale during the HSCT in both groups. All evaluations made in study 2, including HRQoL measurements, was made by a research nurse.

### **5.2.3 Statistical analysis**

We used the non-parametric Mann-Whitney *U*-test to compare the differences between the groups concerning the PedsQL 4.0 generic core scales and PedsQL 3 cancer module. To calculate the effect sizes within groups paired *t*-test was used. We used Wilcoxon's signed-rank test to observe differences within the same group. Due to multiple comparisons within blocks we used Bonferroni's corrections and adjusted the *p* values. To evaluate the mode and the pain scales we used linear regression with cluster-robust standard errors (individuals as cluster).

## **5.3 STUDY 3**

### **5.3.1 Participants and settings**

Six children aged from 1 to 18 years and their parents were included. The patients were collected from the RCT and 8 families were asked in chronological order to participate in the study. Two families declined. In total six children, two girls and four boys, and their parents participated in the study, three children from each treatment group. The interview took place 7–13 months after HSCT and the duration was 45–60 min. Five out of six interviews took place in the music therapy room at the hospital. Due to medical treatment, we visited one family in the child's hospital room and brought some musical instruments to facilitate the interview. It was possible for the child to make drawings during the interview or play on the instruments.

### **5.3.2 Method**

This was a qualitative study using collaborative research interviews as a data collection method. An independent licensed psychologist, psychotherapist, supervisor and trainer in psychotherapy conducted the interviews and analysis. The data collection method, collaborative research interview, was developed by Tom Andersen within the method of the "reflecting team" (211). The method has been used in diverse psychiatric and social clinical

settings to evaluate therapeutic interventions. The aim is to learn from the child and parent, with a structured turn-taking dialogue, providing the clients, therapist and interviewer time to listen, reflect and respond, while all present in the room. This method has possibility to activate the mutual interacting between the participants.

### **5.3.3 Analysis**

Each interview was audiotaped and analysed using thematic analysis by the same psychologist who conducted the collaborative research interviews.

## **5.4 STUDY 4**

### **5.4.1 Participants**

Seven informants from the staff, working at the inpatient ward and from the day care unit for children going through HSCT at Karolinska University Hospital were included. The interviews took place in Nov 2017. The group of participants included one physician, two registered nurses and four assistant nurses.

### **5.4.2 Method**

A qualitative study using focus group interview (212) as data collection method. The seven informants were divided into two focus groups. The interviews were conducted by an independent, licensed psychologist.

### **5.4.3 Analysis**

The two focus group interviews were audio-recorded and transcribed verbatim and analysed according to principles of thematic analysis (213, 214). In order to avoid bias the whole analysis process involved a qualitative researcher with no earlier involvement in music therapy together with the music therapist.

### **5.4.4 Methodology considerations and reflections**

#### **Study 1**

Evaluation of physiological parameters in connection with music medicine has been researched previously in children (125), also within RCT studies (124). Most often, the physiological estimations have been taken in direct relation, before and after the music medicine session. In the research of one single music therapy session with the intervention of singing familiar songs, a study including 9 children from the paediatrics burn ward conducted physiological measurements during the session (215). Physiological measurements in connection with active, expressive music therapy processes in the

paediatric setting has not been presented earlier. Since the children going through HSCT undergo multiple sampling due to the medical treatment, ethical considerations were made not to expose the children in the music therapy study for any further physical measurements of any kind in addition to the samples that were already made. Therefore, morning and evening measurements from blood pressure, heart rates and saturation were collected from the protocol of the ward.

#### Study 2

Previous research show that evaluating pre and post HRQoL for children undergoing HSCT is well used. Evaluating music therapy for children undergoing HSCT or children with cancer using HRQoL questionnaires has not previously been performed as to the best of our knowledge. Since previous studies of HRQoL in conjunction with paediatric HSCT show a well-defined relationship, this method was chosen to evaluate the music therapy intervention.

#### Study 3

The experience to be involved in music therapy during severe medical treatments can only be told by the child and the parents who have experienced the therapy. The chosen methodology refers to facilitating that memories and experiences of the participants, present moments (216) close to the lived life, can be knowledgeable and told. The task of the interviewer, who had no proximity to the therapeutic processes, was to find what stood out and what was meaningful from the interviews. This enlightenment of what had been important to the participants in the interplay was difficult to achieve using any other method.

#### Study 4

The staff see the children from another perspective, and they have broad experience of many different patients. The commitment of the staff looks different, they have different modes of expression and therefore the focus group's methodology was a way to get hold of different experiences. This methodology also included an interviewer with no prior preunderstanding of the music therapy at the ward. In a dialogue between professionals, deeper or inactivated affects may be stimulated and motivate a wider and deeper sharing. The disadvantage of the method may be the difficulties to express a contradicting opinion. The sample in this study were not randomly chosen and participants can only represent themselves although the method had qualitative components and dimensions in focus, not to measure the outcome quantitatively.

## Reflection

According to Hasson and Thiele Schwarz (217) the use of evidence is a balancing act between following methodology and study groups on one hand, and reality on the other; a group that deviates compared to the evidence are not identical in all respects e.g. diseases, age, gender on the other hand.

Modifications and adaptations need to be made and may result in different approaches to get a manualized controlled framework to fit the methodology of the treatment.

There is a parallel to the methodology of research, the methods in our study needed to be flexible within the frames because of the fragile context. Considerations to the scientific research methodology includes some basic assumptions:

RCT reduces the risk of coincidental data, though we couldn't perform a double-blind study. Physiological measurements are objective and qualitative research has to take into account the risk of bias, although results that show effects in the same direction strengthens the total outcome of different separate studies. Earlier music therapy research in the paediatrics, oncology and HSCT populations described music therapy intervention manuals. Why is there not a treatment manual in our study? In the close relational music-therapeutic work with severely ill children, a manual with pre-determined detailed content wasn't seen to fit the core of the treatment, furthermore, it could even have been destructive. Therefore, we chose to use the concept of music therapeutic guidelines in the articles, to express what is essential to consider in relation to the child and what is needed concerning music features. Core components in the clinical method in our study has had the focus on the therapeutic relation with the child, thus the music therapy intervention needed to be flexible, varied, person centred and within the window of tolerance of the child.

In this thesis, we have used both quantitative and qualitative methodology and tried to evaluate the intervention in this context from different standpoints. According to Bornemark (218) there is a gap between the overall structures, measuring instrument, documentation - as a parallel measurement world- and the health ideology; the immediate experienced world that you share with the patient and the necessity to relate to the unknown and the uniqueness of each situation. In this thesis we have tried to bridge between that gap and bring data and knowledge from both sides.

## 5.5 ETHICAL APPROVALS

The Regional Ethical Review Board in Stockholm, Sweden, approved all the studies (2012/1059-31, 2015/100-31/2, 2017/1845-32 and 2017/1111-32). Written informed consent was obtained from the parents, from children over the age of 7 and from the staff participants. Information was given that involvement in the study was voluntary and that it



was possible to withdraw participation at any time without presenting any reason. The control group received the intervention at the day care unit due to ethical considerations and this enabled evaluation of early versus late intervention.

## **5.6 DIFFERENT ROLES OF THE AUTHOR**

Through this entire study period, the thesis author has had different roles. Including writing a research plan, giving therapies, analysing data and being the first author in all articles. The aims of the studies were to evaluate the music therapy through various methods. Since it was desirable to evaluate both quantitative and qualitative effects of music therapy in this context, both methods were used.

In order to circumvent interference from the thesis writer, a research nurse documented the ratings from the heart rates, blood pressure and saturation of the children in study 1. In study 2 the research nurse distributed and collected the questionnaires. The children and the parents answered the HRQoL questionnaires at the same time at the hospital and the research nurse assisted the children to read the questions if needed. The research population is small, and the experience of music therapy in this context was only found at Karolinska University Hospital Huddinge in Sweden. In total, there were only three music therapists in the paediatrics setting throughout Sweden at the time of the study. To avoid bias as much as possible the interviewer in both study 3 and 4 was an independent psychologist. The analysis was performed by her in study 3. To gain the data sample the method of collaborative research interview was chosen and the attendance of the music therapist was needed. In study 4, an independent qualitative researcher, with no music therapeutic experience to reduce pre-understanding and pre-expecting of results, was involved during the whole analysing process. This cooperation was presumed to strengthen the objectivity. The discussion and conclusion section in study 3 and study 4 involved the entire author group in order to strengthen the objectiveness and among the purposes was to give a relevant interpretation of the results.

## **5.7 LIMITATIONS**

Our studies have some limitations, first to be mentioned is the use of single music therapist that is also the thesis author. Secondly, the group was heterogeneous in terms of age and different diagnosis. For ethical reasons, the control group was given music therapy, and it could be speculated that results that are more obvious may have been obtained with a baseline. At the same time, the design enabled evaluation of both early and late intervention. The sample size is small, but previous scientific studies in this area also include less patients, except from the multi-site study with adolescents and AYAs (Table 1).



## 6 RESULTS

### 6.1 STUDY 1

The music therapy group displayed significantly reduced heart rates values in the evening four to eight hours after the music therapy session compared to the control group, where an increase in heart rate was observed ( $p < 0.001$ ). There were no significant differences between the groups concerning blood pressure or saturation, although the saturation increased in the music group in the evening measurements. There were no differences between the groups regarding medical parameters e.g. C-reactive protein and lymphocytes, which could be interpreted as the fact that the two groups were statistically comparable. However, at admission we observed a significant difference based on the Lansky scale, ( $p = 0.03$ ), the music therapy group had lower scores regarding disease severity compared to the control group. At discharge there were no significant differences between the groups.

### 6.2 STUDY 2

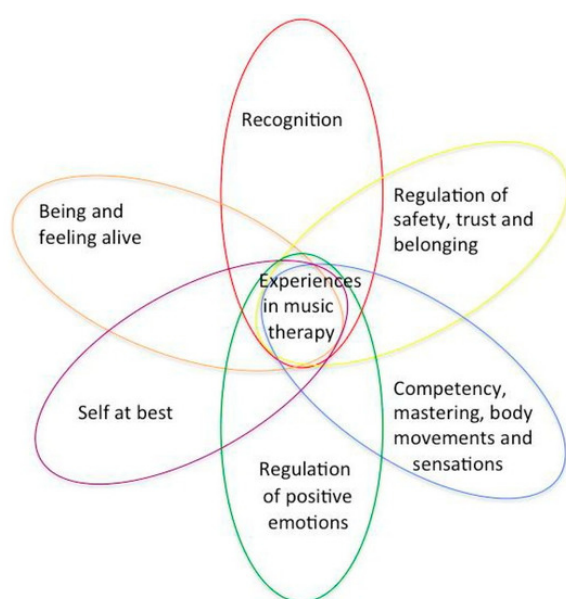
The children in the music therapy group estimated higher mean levels in 50% of the domains in both PedsQL 4 generic scales and PedsQL 3 cancer module at discharge. The domain of physical functioning increased significantly (adjusted  $p = 0.04$ ). Higher estimation showed increased levels of HRQoL. The control group showed reduced mean values in all domains at discharge. The control group was offered music therapy between discharge and six months follow up. At six months follow up, the children in the control group showed improved mean levels in all domains, with the PedsQL 4 generic scales showing a significantly improved HRQoL in all domains ( $p = 0.015$ ). At six months follow up the music therapy group displayed higher levels in all areas except for one.

The parents proxy report showed different and non-significant results. The parents in the music therapy group estimated lower levels of HRQoL of their child in all domains except from the worry domain at discharge. At six months follow up they reported increased HRQoL for their child in eleven out of twelve domains. The parent proxy report in the control group showed, in contrast, increased levels at discharge in eight out of twelve domains. At six months follow up they reported decreased HRQoL of their child in five domains out of twelve. Concerning the estimations in conjunction to the music therapy session, mood and pain, the results showed increased mood ( $p = 0.000$ ) after music therapy treatment in the music therapy group and diminished pain, although not statistically significantly.

### 6.3 STUDY 3

Three main themes developed through the analysis from the collaborative research interviews: (1) experience of competency and recognition of self; (2) experience of interactive affect regulation as potential for change; (3) experience of the importance of the therapeutic relationship.

The analysis ended up in six themes (Figure 2), as the essence and on a different abstraction level: Recognition in meeting the therapist, themselves with family members and the music; Regulation of safety, trust and belonging; Competency, mastering body movements and sensations; Regulation of positive emotions, affects; Self at best, experiencing self in relation, self-assertion; Being and feeling alive.



**Figure 2. Six main themes from the analysis of responses from children and parents in the study. Ugglå *et al* (219).**

### 6.4 STUDY 4

The analysis from focus group interviews with the staff resulted in five themes: (1) The importance of music therapy was expressed both physically and mentally by the children; (2) Music therapy provides the child and the family with satisfaction, affecting the body of the child in a positive way; (3) The parents chose to attend either actively or non-actively, thus, music therapy gives a possibility to rest for the parents; (4) The staff see themselves as an integral part of the music therapy treatment at the department; (5) Music therapy addresses the children in an insecure and isolated situation/milieu.

## 7 DISCUSSION

The results of our studies support our hypotheses and encourages us to proceed with further studies into the field.

In study 1, the results showed that evening heart rates were lowered in the music therapy group 4-8 hours after intervention. This physiological phenomenon can be interpreted as a stress reducing effect, affecting both the physical and psychological recovery of the child. We interpreted these results as potentially preventing PTSD. Symptoms of post-traumatic stress reactions and PTSD has previously been reported in paediatric HSCT patients (63, 82) and increased heart rate is an early risk marker of PTSD (102). In this study, we also observed elevated heart rates in the control group in the evening. We believe these results can argue for increased use of music therapy in the health care of severely sick children. Music therapy is hypothesised, through its stress releasing potentials, to have capacities to prevent PTSD for children going through HSCT. In study 2 the HRQoL increased in 6 out of 12 domains, reported from the children in the music therapy group compared to the children in the control group who reported worse valuations in all domains at discharge. The music therapy group had significantly improved results in physical functioning. It may seem strange to evaluate physical function so close to HSCT. One study even excluded that specific item due to very intensive treatment (220), however it is worth remembering that all music activities include and effect the body as music moves us both emotionally and bodily (221).

In study 2 we also reported that mood increased after the music therapy session. Treatment anxiety and worry showed medium effect size (Cohen's *d*) in the music therapy group compared to the control group at discharge. These results show that music therapy can help the child to receive increased mood directly after the music therapy session and declining anxiety and worry in a longer perspective. The differences between the parental groups at discharge were not significant, although the contrasting results between parent proxy and child self-evaluation in both music and control group show the importance of asking both the child and the parents to evaluate HRQoL. We also believe, that in order to make the evaluation after HSCT broader, the Lansky play performance scale performed by the doctor, should be completed by including self- estimations from the children and parent-proxy reports regarding the HRQoL of the child.

The analysis in study 3 reported that music therapy enabled the child to feel recognised and competent. Musical attunement gave the experience of being able to interact and influence and facilitated an intersubjective affect regulation meeting without words. Music therapy is often considered as a non-invasive treatment and an intervention without negative side effects. Although we have to consider that if something may have positive

effect, it may also have possible negative effect (222). It is of great importance to add that to achieve the potential of music therapy in the best way it is vital to provide academic and clinical training to the music therapists and careful selection of the music therapy methods to meet the need of the patient (34).

In study 4 our findings show that the staff is an integral part of music therapy and even that the parents may have some rest during the intervention, though they still are present in the room. The hospital team did also mention that they perceive a different atmosphere in the child's hospital room after the music therapy session. Previous research shows the importance of psychological care of the children in the paediatric oncology ward and that it is integrated with the medical care (223) and music therapy seems to be easily implemented in the clinic settings (34).

This thesis can be seen as an investigation of music therapy in a HSCT context in four different circular systems. The inner circle is the physiological system of the child, next circle his/her subjective evaluation. The third circle is the experiences in the child-parent system and the fourth circle is the psychosocial system of the staff working with the children and families.

These four systems are intertwined and affect each other in a circular way, how the family perceive the treatment depends on how they are treated by the staff and how the music therapy intervention is experienced by the child and vice versa; music therapy, facilitated by the other different systems, initiate an activation of good repetitive self-perpetuating interaction processes. The different systems depend on and affect each other.

## 8 CONCLUSION

- The combination of lowered heart rates of the children in the music therapy group 4-8 hours after music therapy and improved HRQoL in both groups after intervention suggest that music therapy is an effective and complementary therapy during and after the HSCT treatment.
- The creative and relational process in music therapy is important, giving the child a greater sense of control and enables emotional self-regulation.
- Music therapy developed into a meaningful and supportive experience for the participants, both the child and the parent, and was a significant factor in coping with and managing the treatment period at the hospital.
- The staff supported the child, family and the music therapy treatment at the wards during the whole HSCT process, essential for music therapy to become an integrated part of the care. The staff also benefit from music therapy, an important part of the treatment and rehabilitation during and after HSCT.

## 9 FURTHER PERSPECTIVES

- Music therapy for children undergoing HSCT is currently not part of the regular supportive care in Sweden. Our study has been conducted at one HSCT centre, with the same music therapist and in order to evaluate the generality, it would be desirable to perform a multi-site study inspired by the evaluation methods we used in our study.
- Stress symptoms and PTSD are reported earlier from this population. We did not evaluate stress symptoms or PTSD specifically in these studies. It would be required to evaluate stress symptoms after music therapy in the short term for children undergoing HSCT, long term for HSCT survivors, as well as the impact of music therapy in the stress symptoms in parents and siblings.
- HSCT affects the whole family and therefore there is a need to involve and develop interventions to include the whole family, with the aim to support the communication and family bonding during HSCT.
- The HRQoL in connection to HSCT in the family context is complex. In order to evaluate how music therapy, affects HRQoL, both self-evaluation of the child, proxy evaluation from both parents and parent HRQoL self-report could be meaningful to investigate.
- Our studies have children undergoing HSCT in focus, however, it would increase knowledge to further investigate music therapy interventions for other paediatric patient groups in Sweden.



## 10 SVENSK SAMMANFATTNING

En hematopoietisk stamcellstransplantation (HCST) är en etablerad behandling för barn med aggressiv leukemi men också för barn med avancerade metaboliska och hematologiska sjukdomar. Behandlingen är påfrestande och kan liknas vid ett kraftigt trauma mot kroppens alla vävnader. Den medicinska utvecklingen (av behandlingen) har dock bidragit till att fler HSCTs görs idag, och att fler barn överlever.

Målsättningen är att ersätta sjuka stamceller med stamceller från en donator. Efter konditioneringen, då de egna cellerna slås ut, infuseras friska stamceller. Barnen är isolerade 4-6 veckor eftersom det är risk för svåra infektioner innan de nya stamcellerna producerar nya celler i benmärgen. Vården är intensiv under 3-6 månader och barnet följs regelbundet med många täta kontroller. Riskerna efter HSCT är återfall och transplantationsrelaterade komplikationer som t ex infektioner och graft-versus-host disease (GVHD) då de transplanterade cellerna reagerar mot den nya kroppens vävnader.

Den intensiva behandlingen både före och efter HSCT påverkar hela patientens familj och deras inre relationer. Förutom en uttalad fysisk påfrestande medför HSCT även psykisk belastning. Barnets hälsorelaterad livskvalitet (HRQoL) påverkas, den är som lägst 1-3 månader efter HSCT och det kan ta 1-3 år innan barnet återfått sin HRQoL. Även föräldrarnas och syskonens livskvalitet försämras. Tidigare forskning har visat att patienter som överlevt HSCT kan drabbas av posttraumatisk stress symptom (PTSD) och neurokognitiv dysfunktion. Även höga nivåer av stress och depressiva symptom finns rapporterade i familjer till barn som genomgått HSCT.

Musik påverkar hela hjärnan och våra system för belöning, motivation och välbefinnande. Musik bidrar också till att sänka stressnivåerna och stärka våra sociala relationer. Musik väcker och påverkar känslor och musikens funktion i samspelet mellan spädbarn och förälder underlättar den emotionella regleringen. Amygdala är en del av limbiska systemet i hjärnan och har kontakt med vårt emotionella system men också med fysiologiska processer som hjärtfrekvens och andning. Amygdala reagerar på stressfyllda situationer. Musikaliska aktiviteter som att sjunga, lyssna på känd musik, skapa och improvisera påverkar emotionell reglering och reducerar aktiviteten i amygdala. Musikterapi används inom den pediatrika vården för att hjälpa barnen genom svåra upplevelser och stödja välbefinnandet. Musikterapi är också en relationell terapiform som syftar till att öka patientens intersubjektiva erfarenheter genom samspel och relaterande till musik.

Tidigare forskning visar på ökat välbefinnande och minskad smärta vid procedurer genom att använda musikaliska interventioner för barn med cancer. Musikterapi-forskning för barn som genomgår HCST rapporterar sänkta nivåer av ångest och för

unga vuxna rapporteras förbättrade strategier att klara behandlingen och social integrering efter musikterapi.

Stress är förknippat med högre puls. Forskning visar att förhöjd puls hos barn i samband med trauma, kan förutsäga risken att senare utveckla PTSD symptom.

Denna avhandling handlar om att utvärdera och undersöka om musikterapi påverkar barnen både ur fysiologiskt och psykologiskt perspektiv. Vi har även velat förstå hur det är att spela och sjunga tillsammans med en musikterapeut för både barn och föräldrar, samt slutligen hur personalen uppfattar musikterapin för barnen.

Från februari 2013 till november 2017 genomförde vi en randomiserad studie på Karolinska Universitetssjukhuset i Huddinge. Sammanlagt inkluderades 38 barn i åldrarna 2 mån – 17 år. Barnen lottades till två grupper, där musikterapigruppen fick musikterapi 2 ggr per vecka under den inneliggande perioden. Barnen i kontrollgruppen erbjöds musikterapi efter utskrivning då de kontrollerades på dagvården.

Musikterapiinterventionen erbjöd både expressiva och receptiva delar. Vi träffades i barnens sjukhusrum och barnet erbjöds att spela på olika instrument, sjunga och lyssna på musik tillsammans med musikterapeuten eller måla och röra sig till musik. Föräldrar och syskon kunde också delta. Första målet med terapin var att bygga en trygg terapeutisk allians mellan musikterapeuten och barnet, med barnets önskan och vilja i fokus. Sessionen syftade till att vara flexibel, varierad och erbjuda en tillräckligt hållande struktur för att hjälpa både barn och föräldrar att vara emotionellt reglerade.

**Studie 1** omfattade 24 patienter vars fysiologiska mätvärden utvärderades. Blodtryck, puls och saturation - syremättnad i blodet – dokumenterades morgon och kväll samma dag som barnet hade musikterapi och två gånger i veckan för kontrollgruppen. Kvällspulsen för barnen i musikterapigruppen, ca 4-8 timmar efter musikterapin, minskade signifikant jämfört med kontrollgruppens kvällspuls ( $p < 0.001$ ). Vilket skulle kunna tyda på minskade stressnivåer och förminskad risk för PTSD.

**Studie 2** utvärderade 29 patienter och vi ville jämföra den hälsorelaterade livskvaliteten (HRQoL) för barnen efter musikterapi. Vi använde internationella och validerade frågeformulär. Barnen skattade själva i åldrarna 5-17 år och föräldrarna skattade barnens HRQoL i åldrarna 2-17 år. Frågeformulären användes vid tre tillfällen, vid inskrivning, utskrivning och vid uppföljningen vid 6 månader. I musikterapigruppen, som fick musikterapi mellan inskrivning och utskrivning steg HRQoL i hälften av alla frågeområden vid utskrivningen. Fysiologisk funktion förbättrades signifikant efter musikterapi (justerat  $p = 0.04$ ). Kontrollgruppens medelvärde försämrades i alla frågeområden vid utskrivning.

Vid uppföljningen vid 6 månader, då kontrollgruppen haft musikterapi, förbättrades deras HRQoL inom alla domäner och PedsQL 4 visade signifikant resultat ( $p=0.015$ ). Musikterapiogruppernas HRQoL förbättrades vid 6 månaders uppföljningen i 11 av 12 frågedomäner.

**Studie 3** inkluderade sex barn och deras föräldrar. De deltog tillsammans med musikterapeuten i en kvalitativ studie som använde sig av samforskningsintervju som metod för datainsamlingen. Syftet med metoden är att musikterapeuten får lära sig av deltagarnas erfarenheter av musikterapi och hur musikterapin uppfattas. En oberoende psykolog genomförde intervjun och analysen av materialet. Tre teman växte fram genom analysen: erfarenheter av kompetens och erkänsla av jaget, erfarenheter av interaktiv affektreglering (att i samspel med andra hantera egna känslor) som potential för förändring och erfarenheter av den terapeutiska relationens betydelse.

**Studie 4**, var en kvalitativ fokusgruppintervjustudie som inkluderade 7 personer ur personalen. Syftet med studien var att undersöka personalens upplevelser och attityder till musikterapi för barnen. En oberoende psykolog gjorde intervjuerna och analysen gjordes av avhandlingsförfattaren tillsammans med en oberoende forskare, utan någon tidigare erfarenhet av musikterapi. Teman från analysen: Betydelsen av musikterapi uttrycktes både fysiskt och mentalt av barnen och gav barnet och familjen en känsla av nöjdhet. Föräldrarna deltog mer eller mindre aktivt och musikterapin gav föräldrarna en möjlighet att vila. Personalen var en integrerad del av musikterapin och musikterapin mötte barnen i en osäker och isolerad situation.

**Sammantaget** visar våra studier att musikterapi har en klinisk effekt under och efter HSCT. Kombinationen av minskad puls 4–8 timmar efter musikterapi i musikterapigruppen och förbättrad HRQoL i båda grupperna efter interventionen visar att musikterapi kan vara en effektiv och komplementär intervention under och efter HSCT. Musikterapi utvecklades till en betydelsefull och välgörande erfarenhet för deltagarna, en viktig ingrediens för att klara av behandlingsperioden på sjukhuset. Personalen var engagerad och stöttade barnet och familjen genom behandlingen, vilket är avgörande för att musikterapin ska bli en integrerad del av vården.

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