

Music therapy as procedural and treatment support in paediatric healthcare: a review of the literature from a Nordic perspective

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Abstract

Music therapy (MT) is here understood as a relational therapy with the potential to alleviate pain and anxiety in hospitalised children and support them in coping with medical procedures and treatment. Research indicates that children who receive MT as procedural support before, during and after painful or frightening medical procedures are better equipped for coping with future procedures and less likely to develop symptoms of paediatric medical traumatic stress. MT can improve the ability of a child to cooperate, thereby reducing the duration of the procedure. This article explores some of the methodological and epistemological issues in MT as procedural support research, and mixed methods research is suggested as a suitable approach for acquiring new knowledge. Few studies on MT as procedural and treatment support have so far been conducted in paediatric settings in Denmark, Sweden and Norway, and there is a need for more randomised controlled trials (RCTs). The Nordic countries have strong MT traditions, but only a small number of music therapists are currently working with procedural support for children. More studies can lead to inclusion of recommendations of MT as procedural and treatment support in national guidelines for evidence-based clinical practice in these countries as well as serve to further develop the theoretical framework. The available Nordic literature will be reviewed, and important aspects of research, clinical practice and theory development will be discussed from a Nordic perspective while drawing upon the rich international evidence base for MT as procedural and treatment support.

Keywords: *music therapy, procedural support, paediatric care, child traumatic stress*

Introduction

Children in paediatric hospitals are at substantial risk of developing long-term negative effects as a consequence of undergoing painful, frightening or unpleasant medical procedures (Young, 2005). There is a need for supportive interventions that can alleviate the pain and anxiety that young children experience in association with medical procedures (Young, 2005). Research suggests that music therapy (MT) can be helpful to patients and that music therapists can work in clinical paediatric settings employing various MT methods to address stress, anxiety, pain and emotional states in patients (Edwards & Kennelly, 2017; Ærø & Aasgaard, 2011). There is an increasing interest in MT as procedural support for children, and while the evidence base for paediatric MT has become quite substantial, there is a need for more research supporting MT for procedural support as an evidence-based practice (Beer & Lee, 2017; Edwards & Kennelly, 2017). There is considerable diversity across studies in terms of types of music interventions and methodology, and more good quality research is necessary for the development of systematic reviews. The main purpose of this review is to identify the available literature on MT as procedural and treatment support from Denmark, Sweden and Norway which might serve as an indication of recent developments within this area of MT in these countries. Firstly, a brief overview of the historical development of paediatric MT will be presented. Secondly, the rationale for using MT as procedural support will be discussed. Thirdly, studies performed in Denmark, Sweden and Norway on MT as procedural and treatment support identified through a literature search will be subject to analysis. Fourthly, issues regarding study methodology and design in MT as procedural support studies will be considered. Finally, the current use and implementation of MT in paediatric healthcare services in the Nordic countries will be explored, including recommendations for future research and clinical practice.

Literature search strategy

The following search engines and databases were used: Bergen Open Research Archive, Cochrane Library, the Danish National Research Database, the Norwegian Library System, PubMed, and WorldCat. Search terms included "Music", "Music Therapy", "Procedural", "Procedures", "Support", "Intervention", "Child", "Hospital", "Hospitalised", "Paediatric", and "Medical". *The Arts in Psychotherapy*, the *Australian Journal of Music Therapy*, the *British Journal of Music Therapy*, the *Canadian Journal of Music Therapy*, the *Journal of Music Therapy*, the *Journal of the Association for Music & Imagery*, *Music and Medicine*, *Music Therapy Perspectives*, the *Nordic Journal of Music Therapy*, the *New Zealand Journal of Music*

Therapy, Psychology of Music, and *Voices* were all hand-searched. Relevant publications were also identified through the websites of the Nordic MT research centres: the Center for Documentation and Research in Music Therapy (CEDOMUS) at Aalborg University, Denmark, the Royal College of Music in Stockholm, Sweden, the Grieg Academy Centre for Music Therapy Research (GAMUT) at the University of Bergen, Norway, and the Centre for Research in Music and Health (CREMAH) at the Norwegian Academy of Music, Oslo.

Inclusion and exclusion criteria

The following inclusion criteria were used for the literature review: the article was (a) original research (quantitative or qualitative), a review article or a case study, (b) written in English, Danish, Swedish, or Norwegian, (c) conducted in Denmark, Sweden or Norway, (d) published after January 2000, and (e) considering the topic of MT as procedural or treatment support. The following types of articles were excluded: (a) studies with participants in the neonatal period or adults, (b) studies without a music therapist responsible for the intervention, (c) studies not related to medical procedures or treatment, and (d) studies including participants with disorders of consciousness.

Terminology, definitions and distinctions

There is a need for clarification of the terminology, as paediatric healthcare personnel use music in a few ways, with varying incentives. The terms music medicine (MM) and music therapy (MT) are sometimes equivocated and used ambiguously in the literature despite the fact that MT and MM are two very different treatment forms based on different paradigms (Bonde, 2019b). In procedural support research it is therefore important to differentiate between MT and MM interventions (Gooding, Yinger & Iocono, 2016). MT as procedural support can be defined as "(...) the use of music and aspects of the therapeutic relationship to promote healthy coping and decrease distress in individuals undergoing medical procedures." (Ghetti, 2011, p. 6). MT can be defined as a relational and interaction-based therapy form, which can be viewed as a triad consisting of interaction between the child, the therapist and the music (Uggla, Bonde, Hammar, Wrangsjö & Gustafsson, 2018). The presence of the music therapist is seen to contribute as an individual factor to the pain-reducing effect of MT (Schou & Bonde, 2012), and so the therapeutic relationship formed through MT sessions is a key difference between MM and MT (Bonde, 2019b, p. 209). Music employed by other healthcare professionals is often used as means of fleeting distraction (e.g. from pain), whereas it is important in MT in paediatric care to obtain more sustained

attention to the music in order to manage pain and anxiety (Edwards & Kennelly, 2017; Mondanaro, 2013). Live music has the potential for altering musical parameters, thus engaging the child in the musical activity for longer (Edwards & Kennelly, 2017). MM typically refers to the use of specially selected pre-recorded music for music listening in a hospital setting, usually administered by other healthcare personnel than music therapists, with the aim of improving well-being of patients (Edwards & Kennelly, 2017). As reviews often include both MM and MT, it can be challenging to determine the exact level of evidence for MT as procedural support. Several meta-analyses show that MT has a clinically more meaningful effect across somatic treatment forms than MM (Bonde, 2014a, 2019b; Lee, 2016), but more good quality research is needed in order to compare these interventions (Ghetti, 2016). Lastly, medical diagnostic and treatment procedures can be classified into (a) non-invasive and non-painful procedures, (b) invasive and painful procedures, and (c) surgery (Ghetti, 2011; Sanfi, 2012).

Paediatric medical traumatic stress

Children with serious medical conditions have experienced unsafety and are therefore in particular need of safety and control. Being in hospital can create a feeling of helplessness, a sense of foreboding and a feeling of actual or perceived threat or of life being at stake (Nielsen, 2013). Many factors determine what preprocedural expectations, beliefs and coping skills a person has, and their pain response is further influenced by factors related to the present situation in which the procedure is being carried out (Young, 2005). It is important to also keep in mind that ahead of any medical procedure, the child is likely to have experienced an accident or illness, which adds to the pain and anxiety associated with the procedure itself (Nielsen, 2013). Anxiety increases the pain response (Young, 2005) and, conversely, fear of pain increases anxiety (Schou & Bonde, 2012). Medical procedures and treatments are seldom recognised as one of the most frequent causes of post-traumatic stress reactions in young children (Gjems & Diseth, 2011; Nielsen, 2013), and pain is often undertreated in paediatric patients (Diseth & Reinfjell, 2018). Several studies have shown, however, how repeated painful procedures and medical treatments can lead to symptoms in children similar to post-traumatic stress symptoms associated with PTSD¹ (Diseth & Reinfjell, 2018; Kazak et al., 2005; Nielsen, 2013; Stuber, Shemesh & Saxe, 2003; Stuber,

¹ Post-traumatic stress disorder (PTSD) in children is a severe, often chronic, and impairing mental disorder seen in some children after exposure to traumatic experiences involving actual or threatened injury to themselves or others (Brent, Cohen & Strawn, 2019). PTSD is characterised by intrusive thoughts and reminders of the traumatic experience(s), avoidance of trauma reminders, negative mood and physiological hyperarousal (Brent et al., 2019).

1996; Young, 2005) and have resulted in a change in medical procedures in children. The youngest children, who are not able to understand the reason behind medical procedures, are at the greatest risk of being traumatised, while older children are generally less affected by medical procedures (Diseth & Reinfjell, 2018; Nielsen, 2013; Sanfi, 2012). Even minor procedures have been shown to cause significant pain and distress in children, and inadequate use of interventions to ameliorate this can predispose children to long-term negative effects (Young, 2005). Long-term effects are partly determined by memories of the painful procedure, which can be accurate in children as young as three years old (Young, 2005). Such memories can become exaggerated, triggering increased distress in future procedures (Noel, McMurtry, Chambers & McGrath, 2010; Young, 2005). Pain experiences in early life directly influence how a person responds to future medical procedures (Young, 2005), and patients with chronic or complex diseases may be reluctant to undergo treatment because of fear that there will be pain involved. Due to the potentially severe consequences, there is a need for early identification and intervention (Diseth, 2009) as initial pain and anxiety management is essential to the prevention of post-traumatic stress symptoms² (Diseth & Reinfjell, 2018). The pain response is to some extent modifiable through pain- and anxiety-reducing interventions (Young, 2005) such as MT, which can help the child regain a feeling of safety, comfort, strength and physical and mental self-esteem (Nielsen, 2013). MT can lower anxiety and modify distorted memories of procedural pain, thereby preventing a continuous negative cycle and lowering the risk of developing paediatric medical traumatic stress (Nielsen, 2013; Young, 2005).

International and historical perspectives

Internationally, there is a long-standing tradition of using music with hospitalised children, particularly in the United States and Australia, which began with MT in neonatology in the United States in the late 1980s (Shoemark & Dearn, 2017). MT support for newborns subsequently evolved in many parts of the world from the 1990s onwards to encompass a wide array of medical contexts where also older children were supported during hospitalisation (Shoemark & Dearn, 2017). MT for newborns as well as the general MT profession are now well established in countries such as Australia, Canada, Germany, Israel, Sweden, Switzerland, United Kingdom and the United States, where much of the clinical work and research has taken place (Shoemark & Dearn, 2017). MT in paediatric settings has evolved

² Pre-procedural psychological assessment and screening tools for the prevention of adverse psychological effects in children receiving medical treatment and their families are discussed in more detail by Diseth & Reinfjell (2018).

along with changes in medical practice and is still growing as a profession in healthcare worldwide (Ghetti, 2016), moving steadily towards becoming an integral part of paediatric care in many Western countries (Mondanaro, 2013). Numerous music therapists are at present employed in paediatric oncology, including in Australia, Germany, Norway, Sweden and the United States (O'Callaghan & Aasgaard, 2012). However, the establishment of hospital-funded music therapist positions continues to pose a challenge for clinical practice and research in several countries including Australia, Norway and the United States (Ghetti, 2016). In line with the growth of the profession, the amount of internationally published literature on paediatric MT has increased significantly since the 1990s (Ghetti, 2016). Over the years a selection of systematic reviews (Klassen, Liang, Tjosvold, Klassen & Hartling, 2008; Yinger & Gooding, 2015), meta-analyses (Bradt, Dileo, Magill & Teague, 2016; Lee, 2016) and RCTs (Robb et al., 2008, 2014; Yinger, 2016) have been published on paediatric MT including procedural support. Both the systematic reviews and meta-analyses point out the need for differentiating between MT and MM interventions, and although findings generally indicate that MT may be effective as procedural support in hospitalised children, there is considerable heterogeneity and a high risk of bias among the studies included. The RCTs investigated the efficacy of MT interventions only and recommend the use of MT for procedural and treatment support as it potentially improves outcomes. Comprehensive introductions to paediatric MT can be found in *The Oxford Handbook of Music Therapy* (Edwards & Kennelly, 2017) and in *Guidelines for Music Therapy Practice in Paediatric Care* (Bradt, 2013). Even though increasing emphasis is placed on providing MT as procedural support as part of a family-centred care model (Edwards & Kennelly, 2017), research on MT with hospitalised children *and their families* has until recently not been very prevalent in the literature and is still being developed in clinical practice (Jacobsen, 2019).

Historical development in the Nordic countries

Norwegian music therapists began working clinically with hospitalised children in the mid-1990s and have since been gradually integrated in some paediatric departments, making them pioneers in the area within the Nordic context (Ærø & Aasgaard, 2011). MT in Norway is historically closely linked to special education work, which provides a developmental perspective on working with procedural support for children, also in being resource-oriented (Ruud, 2008). Early MT work in Norwegian paediatric wards often took place in communal areas where more children and their families could take part³ (Ruud, 2008). This perspective is still relevant, as making institutional changes in the hospital environment can have a positive impact on patients, which in turn may help them cope

3 MT in this setting is known as 'Environmental music therapy' (Sanfi & Bonde, 2019).

better with medical procedures (Ruud, 2008). Paediatric MT is within the Nordic context relatively well established in Norway (Sanfi & Bonde, 2019). In Denmark MT remains a comparatively new and not yet fully integrated treatment form in paediatric medicine and is still being developed within healthcare services (Sanfi & Bonde, 2014). Research in Denmark has so far focused particularly on guided imagery and music (GIM)⁴ for treatment of side effects of chemotherapy and on MT as procedural support (Sanfi & Bonde, 2019). MT in Sweden has had close ties with MT associations in Denmark and Norway for a long time (Hammarlund, Körlin, Johansson, Wallius & Paulander, 2008). Its beginnings can similarly be traced back to special education work and has since expanded to include a number of work areas for music therapists, including paediatric settings (Hammarlund, 1993). Swedish MT also has a tradition of GIM training, practice and research (Hammarlund, 1993). MT in Sweden has been characterised by multidisciplinary work, where music therapists have collaborated with various health professionals as well as art therapists (Hammarlund et al., 2008). The increasing amount of outcome-focused research emphasising specific procedures is testimony to how MT as procedural support is shaped by the field of medicine (Gold, 2012) but can be regarded as an addition to the rich humanistic European MT tradition (Ridder & Bonde, 2019a). The current expansion of MT practice within paediatric healthcare is very much in line with the developments seen in somatic healthcare in Northern Europe in the past few decades (Bonde, 2014b). Due to the development of an holistic and patient-centred medical healthcare model and, by extension, the integration of MT services with healthcare, music therapists in the Nordic countries are opportunely placed for further advancing the use of MT as procedural support (Bonde, 2014b).

Methods in music therapy

Music is used in a systematic way in MT to promote health and positive changes in a child (Bonde, 2019a). MT as procedural support is described in the literature as an augmentative practice that facilitates medical procedures (Bonde & Trondalen, 2019). Music experiences and activities are applied before, during and after completion of the procedure (Sanfi, 2016). For instance, the music therapist can engage the child in MT prior to a procedure, which then continues throughout the procedure itself (Ghetti, 2011). Therapeutic goals of MT as procedural support are (a) to reduce stress, anxiety, pain and the duration of the procedure, (b) to prevent trauma and negative long-term effects, (c) to promote relaxation

4 Guided Imagery and Music (GIM) is a receptive MT method developed in the 1970s in the United States and is often referred to as the Bonny Method of GIM (Trondalen & Oveland, 2008). GIM has grown in popularity in particularly Asia and Europe and is today practised worldwide (Bonde, 2019c).

and well-being, and (d) to support and promote the ability of the child to cope during the procedure and help with mental processing of the experience (Sanfi, 2012, 2016). Methods that are commonly applied across different MT models and orientations are based on improvisation, song-writing, therapeutic use of voice and receptive methods such as music listening and music imagery (Jacobsen & Bonde, 2019). The use of improvisation allows the therapist to motivate and engage the child, offer support and acceptance, and give them the joy of being creative (Sanfi, 2012). In addition, improvisation can be used as means of expressing feelings, to create an understanding of and ascribe a purpose to a situation (Sanfi, 2012). A well-documented approach that is used in improvisation and has been shown to alleviate pain in children is based on *entrainment*, a synchronisation of physiological rhythms and external musical stimuli (Ærø & Aasgaard, 2011). Musical parameters (e.g. frequency, dynamics, melody, tempo and rhythm) can be used in specific ways to affect the autonomic nervous system, the processing of nociceptive stimuli, and a number of physiological processes (e.g. heart rate, respiratory rate and arousal) (Bonde, 2019b). More often than not, the music therapist will consecutively apply a combination of methods (Jacobsen & Bonde, 2019) in order to accommodate the needs of a child throughout the course of a procedure (Reid, 2017; Sanfi, 2016). In summary, MT as procedural support is individualised according to the child's needs and the nature of the procedure (Sanfi & Bonde, 2019).

Literature review: research on music therapy as procedural support in Denmark, Sweden and Norway

Three RCTs have been published on the topic of MT as procedural and treatment support in paediatric settings in Denmark (n=1) (Sanfi, 2012) and Sweden (n=2) (Uggla et al., 2018; Uggla et al., 2016; Ullsten, 2017). Ullsten (2017) focused on MT as procedural support for preterm and term neonates undergoing venepuncture in the form of lullaby singing performed by a music therapist. While beyond the scope of this review, this conforms to a tendency internationally where there appears to be a predilection for clinical trials on MT for neonates or hospitalised infants. The remaining two RCTs will be subject to analysis, the first using MT in relation to peripheral intravenous access (PIVA) in young children (Sanfi, 2012) and the other investigating the efficacy of MT for children undergoing haematopoietic stem cell transplantation (HSCT) (Uggla et al., 2018; Uggla et al., 2016). An interview study (Uggla, Blom, Bonde, Gustafsson & Wrangsjö, 2019) published in relation to the latter and, finally, two selected case studies conducted in Norwegian (Aasgaard, 2002) and Danish (Nielsen, 2013) paediatric departments respectively, will be discussed.

Needle-related procedures

Procedures that require the insertion of needles into a vein can be defined as invasive and can cause pain, elevated anxiety and distress in children to the extent that it is often necessary to hold children still by physical restraint (Sanfi, 2012). Sanfi (2012) assessed the efficacy of MT during needle procedures in a study at a Danish nephro-urological paediatric outpatient clinic. Children were randomly divided into an MT group who received MT directly before, during and after the procedure and a control group on whom the procedure was carried out according to standard PIVA procedure (Sanfi, 2012). The MT intervention comprised receptive, re-creative, improvisational and compositional music experiences (Sanfi, 2012). MT support led to a significant reduction in the duration of the procedure⁵ by 33% compared to the control group, which can most likely be explained in terms of the child cooperating better with the medical staff (Sanfi, 2012). This is a considerable advantage not only for the child and everyone involved, but equally from an economic perspective (Sanfi, 2012). Similarly, child anxiety⁶ was 18% lower in the MT group and thereby significantly reduced by MT support (Sanfi & Bonde, 2014). While the MT intervention did not have a significant effect on child distress, the author found that the parents of the children who had been allocated to the MT group were content with the MT intervention and had found it to be helpful with regard to the medical procedure⁷ (Sanfi, 2012). In summary, the study provides preliminary evidence of the efficacy of MT as procedural support for PIVA by showing that MT reduced anxiety in children, who thereby became more compliant, which in turn reduced the duration of the procedure (Sanfi, 2012). Self-reported pain may be subject to social desirability bias (Sanfi, 2012), but duration of the procedure is an objective, quantitative outcome measure that allows for comparison of results between clinical trials. Because the MT intervention and subsequent assessment of outcome measures were done within a relatively short time frame, it is beyond the realms of the study to predict any possible long-term effects of MT as procedural support intervention (Sanfi, 2012). MT sessions were provided by the same music therapist, and involvement of more therapists in future trials may account for any possible therapist-specific effects (Sanfi, 2012). The results from this trial are likely to be applicable to paediatric settings in Sweden and Norway, where PIVA is performed in a similar way. Arguably, the results may be generalised to children with other diseases undergoing PIVA, but a separate trial would be needed to evaluate whether

5 The duration was measured by a research assistant by use of stopwatches (Sanfi, 2012).

6 The tools used to measure child anxiety levels were Visual Analogue Scales (VAS) and a modified version of the Faces Pain Scale - Revised (FSP-R) (Sanfi, 2012).

7 The author used a Likert-type scale for rating parents' overall satisfaction with the procedure and also included optional comments from parents but underlines that the latter were non-validated data, and that this was not a mixed methods study (Sanfi, 2012).

MT has the same effect on children with more severe or chronic diseases who repeatedly undergo PIVA and other medical procedures (Sanfi, 2012).

Haematopoietic stem cell transplantation

Haematopoietic stem cell transplantation (HSCT) is a hazardous procedure used to treat severe haematological conditions. The treatment involves isolation, poses a substantial risk of complications, and causes high levels of anxiety (Uggla et al., 2018). HSCT leads to a reduction in health-related quality of life (HRQoL)⁸ in children that lasts for up to one to three years after end of treatment (Uggla et al., 2018) and is known to cause post-traumatic stress disorder symptoms or PTSD (Uggla et al., 2016). Hence, specific interventions are called for to mitigate the risk of negative mental health effects (Packman, Weber & Bugescu, 2010).

Heart rate outcomes

Uggla et al. (2016) investigated the effect of MT as a psychosocial intervention for children undergoing HSCT by measuring objective physiological parameters including heart rate (HR). A control group received standard care (i.e. no MT during hospitalisation), and an intervention group received MT sessions (approximately 45 minutes) twice a week during hospitalisation for four to six weeks until donor engraftment was established (Uggla et al., 2016). In the MT sessions the child was invited to sing, play instruments and listen to music with the therapist, and sessions were guided by the child's initiative and involvement (Uggla et al., 2016). Oxygen saturation, blood pressure and HR were measured regularly according to hospital protocol (Uggla et al., 2016). Infection parameters and haematological blood tests showed no difference between the two groups, supporting the fact that they were eligible for comparison (Uggla et al., 2016). HR was the primary outcome of the study, as previous studies have shown a correlation between HR and PTSD, and increased HR has been proposed as an early marker of PTSD risk (Bryant, Salmon, Sinclair & Davidson, 2007; De Young, Kenardy & Spence, 2007). By relying on standard vital sign recordings on the paediatric ward, Uggla et al. (2016) found that children in the MT group had a significantly lower HR compared to the control group that persisted between four and eight hours after the MT intervention, which points towards a potent effect of MT in this context. Although HR can be precisely measured, it is subject to great variability, and changes are inconsistent (Young, 2005). Young (2005) therefore recommends that physiological changes should

⁸ Health-related quality of life (HRQoL) is defined as 'an individual's or a group's perceived physical and mental health over time' (National Center for Chronic Disease Prevention and Health Promotion, 2018). The PedsQL 4.0 generic score scales and PedsQL 3.0 cancer module scores are created for assessment of HRQoL in children (The Pediatric Quality of Life Inventory) (Varni, 2019).

be interpreted with care and treated as corroborative evidence only. However, repeated measurements over a period of time increase the reliability of the data in this trial. While it is likely that the MT effect is also reflected in immunological and endocrinological biomarkers, studying these biomarkers would only be of value for understanding the impact of music if they were all studied in relation to each other and not in isolation (Fancourt, Ockelford & Belai, 2014). As this would require numerous additional blood samples, it would probably defeat the object of reducing stress associated with medical treatment and procedures, and choosing a 'non-invasive outcome measure' such as HR is therefore both practical and sensible (Uggla et al., 2016). The authors propose that significantly lower HR in the MT group potentially reduced the risk of PTSD in these children (Uggla et al., 2016). One might, however, question whether prevention of tachycardia is in fact sufficient for lowering the risk of PTSD. As such, the conclusion that the decreased HR observed in the MT group indicates a reduction in stress levels and possibly a lower risk of developing PTSD may be too strong. A weaker claim might be more appropriate, such as 'an increased HR can to some unknown extent predict PTSD, and as MT interventions led to a reduction in HR, MT interventions could constitute part of preventive measures or early interventions for children that may potentially be at risk of developing PTSD due to HSCT'.

Health-related quality of life outcomes

In a second article the primary outcome measure was health-related quality of life (HRQoL), which was measured using the PedsQL 4.0 generic score scales⁹ and PedsQL 3.0 cancer module scores¹⁰ (Uggla et al., 2018). The MT group received MT sessions during hospitalisation, while the control group received MT in an outpatient setting between discharge and follow-up at six-months (Uggla et al., 2018). Measurements of HRQoL were done in both groups at admission, at discharge and after six months through a set of questionnaires¹¹ (Uggla et al., 2018). At discharge the MT group showed significantly better estimated physical functioning than the control group and showed fewer signs of treatment anxiety and worry (Uggla et al., 2018). At six months, the control group showed improved results in all items measured on the PedsQL 4.0 generic score scales (Uggla et al., 2018). Hence, both groups experienced an increase in scores, indicating a better HRQoL during the time period in which they received MT. In particular, the outcome measures that had been significantly better in the MT group at discharge had remained stable at

9 Measuring (1) physical functioning, (2) emotional functioning, (3) social functioning, and (4) school functioning (Varni, 2019).

10 Measuring pain and hurt, nausea, procedural anxiety, treatment anxiety, worry, cognitive problems, perceived physical appearance and communication (Varni, 2019).

11 Where appropriate (i.e. in the youngest children), parents evaluated their children's status and filled in the questionnaires (Uggla et al., 2018).

follow-up at six months (Uggla et al., 2018). The study design allowed for assessment of both short and long-term effects of MT on the well-being of children undergoing HSCT and made it possible to evaluate the impact of timing of MT interventions with regard to the medical procedures (Uggla et al., 2018). The combined findings of reduced HR four to eight hours after the MT intervention and improved HRQoL that persisted after the end of treatment further strengthen the evidence that MT has positive physiological and mental health effects on children undergoing HSCT and should be offered as a complementary intervention (Uggla et al., 2018).

Discussion of limitations

It is difficult with a small sample that reduces statistical power to produce significant results (Uggla et al., 2018), and low-power experiments are subject to false negatives and false positives, the latter being the most likely in this case. Consequently, the results from this pilot study undoubtedly warrant future studies with larger sample sizes. However, because of the severity of the treatment and vulnerability of these children, all evidence indicating even a subtle improvement in physical functioning and reduction of anxiety is extremely valuable (Uggla et al., 2018). It is questionable to what extent results from this study are generalisable to other procedures and treatments of a less complex and hazardous character. The trial has high reproducibility, and results are likely to be applicable to children undergoing HSCT in Denmark and Norway. Blinding is not possible in the case of MT interventions, and individualisation is intrinsic to MT as procedural support. The quality of RCTs on MT as procedural support may also be reduced by factors such as confounders (e.g. prognosis and medication) being unevenly distributed in children with severe conditions (O'Callaghan & Aasgaard, 2012). Due to the highly subjective nature of outcomes that are measured, such as pain, MT trials in this context may be at risk of positive bias and estimation of larger effects (Nunan & Heneghan, 2018). Arguably, standardised measurement scales (such as questionnaires) used in RCTs often include information that is less relevant to the patient than to the researcher (O'Callaghan & Aasgaard, 2012). Results were not analysed in terms of age or gender of participants, presumably due to small sample sizes.

Music therapy as an interpersonal process

In a third article qualitative aspects of the MT intervention were explored through collaborative research interviews with families both from the MT group and the control group 7–13 months after completion of HSCT treatment (Uggla et al., 2019). The music therapist responsible for the MT intervention also participated in interviews with the intention of recreating the dynamic that had been present during MT sessions and thereby helping

families evoke memories of these interactions (Uggla et al., 2019). Questions were focused on the interpersonal relationships created through MT sessions, and the aim was to identify key elements in the interaction with the music therapist. Three common themes were derived from the data: (1) the experience of MT bringing forward memories of mental states from times when the children had felt at their best self, (2) being emotionally moved by the musicking¹², causing feelings of happiness and a sense of being recognised, thereby allowing for intersubjective affect regulation, and (3) the experience of how essential the therapeutic relationship was for regulating both bodily sensations (e.g. diversion from pain) and emotions (positive and negative) (Uggla et al., 2019). Negative feelings were reinforced in the absence of MT, highlighting not only the importance of the developed relationship with the music therapist but also the importance of creating an optimised framework for MT interventions for children undergoing HSCT (Uggla et al., 2019). The authors conclude that interactions with the music therapist played a significant role in helping children cope during hospitalisation, and they stress the importance of developing MT interventions that include the family of a child undergoing HSCT (Uggla et al., 2019). Uggla et al. (2019) were the first to use this study design in a paediatric setting, and the study is highly relevant for promoting family-centred paediatric MT interventions (O'Callaghan & Aasgaard, 2012). The fact that the data was collected and analysed by the same person is addressed by the authors, as it puts the study at risk of interviewer bias. Results are also susceptible to recall bias in participants, e.g. due to the opportunity to play music during interviews. The study did not illuminate further the topic of the timing of the MT intervention (during or after HSCT), as families were mostly content with and even preferred taking part in MT in the period they had done (Uggla et al., 2019). Although ethical concerns relating to withholding MT support from one group during hospitalisation were perhaps not unfounded (O'Callaghan & Aasgaard, 2012), this study shows that the design was probably justified. While the results are based on personal experiences of MT in a vulnerable setting, the findings are relevant for music therapists working with families during complex medical procedures and treatment.

Case studies

Song creation

Aasgaard (2002) performed five case studies in children undergoing treatment for haematological disease in Norwegian hospitals, using song creation with children and their families. The different elements of the song creation process, which encompassed various

12 'Musicking' is a proposed understanding of music as an activity and form of interaction that has the ability to promote health and thus lies at the heart of MT for supporting hospitalised children (Aasgaard, 2008).

techniques, are explored and analysed in order to gain new insight from their narratives about the potential of song activities in this context to add health and meaning to life, including beyond the hospital setting (Aasgaard, 2002). While the primary focus of the songs was on health and normality, this practice served to alleviate some of the discomfort related to the procedures the children underwent through the expression of emotions in song activities (Aasgaard, 2002). In Denmark a project conducted by Bonde and Sørerensen showed similar results (Sanfi & Bonde, 2019). Children who were currently undergoing treatment at an oncological paediatric ward or as outpatients took part in a number of MT sessions in conjunction with their medical treatment, which helped shifting the focus of the children and their parents from pathology to normality through creativity (Sanfi & Bonde, 2019). The writing and composing of own songs together with the music therapist is a key method in MT as treatment support, but little research was done on this topic before 2000. The case studies by Aasgaard were some of the first published research of this kind (Aasgaard, 2008).

Treatment of traumatic stress symptoms

Sometimes, regardless of what other psychosocial support the child has received, a medical procedure leads to traumatisation of the child (Nielsen, 2013). Nielsen (2013) details a case study from a Danish hospital of a four-year-old boy with two chronic conditions that required numerous medical procedures. After an induction of anaesthesia had not gone according to plan, the boy had panicked and refused all subsequent medical treatment, which was understood as paediatric medical traumatic stress (Nielsen, 2013). Due to the nature of his diagnoses, however, it had been necessary that he undergo the same procedures again, and this had led to further traumatisation manifesting in stress symptoms that negatively affected his everyday life, as well as making future hospitalisation very unpleasant for both the boy and his parents (Nielsen, 2013). He was referred to MT together with his parents and six-year-old brother, who had also shown changes in behaviour as a consequence of the frequent hospital admissions of his younger brother (Nielsen, 2013). The family participated in MT over the course of ten sessions while the boy was not currently being treated in hospital (Nielsen, 2013). By using music and play therapy, song writing and GIM, the author helped the boy change behaviour and learn coping strategies for managing procedures in the future (Nielsen, 2013). Uncomfortable and anxiety-provoking procedures are not only a burden for the child but can be particularly difficult for the parents or guardians and challenging for the healthcare worker responsible for performing the procedure (Aasgaard, 2002; Ærø & Aasgaard, 2011). Parents or guardians and other family members such as siblings should therefore be included in MT sessions whenever possible, as they are the most important psychosocial support the child in hospital has and are at risk of developing post-traumatic medical stress symptoms themselves (Alderfer, Labay & Kazak, 2003; Diseth

& Reinfjell, 2018; Long & Marsland, 2011; Nielsen, 2013; Stuber, 1996). Furthermore, if the parents are not able to cope well with their child undergoing a procedure or experiencing distress and discomfort, this can negatively affect the child (Norberg, Lindblad & Boman, 2005; Sanfi, 2012). Case studies like this are a valuable contribution to research as they are close to clinical practice, and detailed descriptions allow readers to judge for themselves the relevance to their own work and research (Ridder & Bonde, 2019b).

Reflections on research methodology and design

It is a key condition that researchers reflect upon their choice of epistemological position when deciding what method to apply in order to acquire new knowledge on a topic. Ridder and Bonde (2019b) suggest dividing research into three areas of science: (a) exploratory, (b) descriptive, and (c) explanatory (causal) rather than the qualitative versus quantitative paradigm. According to this view, studies with a qualitative design belong to the exploratory area and RCTs to the explanatory, or causal, area. In explanatory research the aim is to identify causal relationships between variables identified in advance (Ridder & Bonde, 2019b). A researcher using a descriptive approach will define the main variables of the study but not make assumptions about their causal relationships (Ridder & Bonde, 2019b). In the case of exploratory research, one does not know what one does not know, and the data, which is often rich and complex and based on few participants, must be approached open-mindedly (Ridder & Bonde, 2019b). Exploratory research might ask how certain phenomena are *experienced* by participants during MT as procedural support and is dependent on words, music and non-verbal communication (Wheeler, 2017).

Mixed methods research

Because MT interventions for procedural support are complex, it is intrinsically difficult to standardise the independent variable (i.e. the MT intervention) in explanatory research, and it is perhaps misleading to understand MT as procedural support interventions as a single variable (O'Callaghan & Aasgaard, 2012; Tråsdahl, 2011). As a result, it is challenging to identify the exact component or combination of components in MT as procedural support intervention (e.g. musical factors, relational factors and ways in which parents are affected by the musicking that the child notices) that is responsible for change in a child and whether they simply disconnect the child from the unpleasant stimuli or rather refocus their attention (O'Callaghan & Aasgaard, 2012). A better approach to addressing such questions may be to collect different types of data through a combination of study

designs, i.e. a mixed methods approach. Although much MT research is gradually becoming more oriented towards the sciences and medicine (Ridder & Bonde, 2019b), mixed methods research is relatively new in MT research (Wheeler, 2017). Most often RCTs do not contain sufficient information as to *why* a particular MT intervention for procedural support is effective, because the experiences of children benefiting from individualised MT sessions cannot be accessed in this type of study (Bradt, Burns & Creswell, 2013). Mixed methods research can incorporate elements such as systematic enquiry, clinical experience and individual patient perspectives, thereby compensating for the problems of only using either a quantitative or a qualitative methodology (Bradt et al., 2013). Despite pragmatic and philosophical concerns over combining methods (Wheeler, 2017), the application and combination of methods from different areas of science is important, as they generate different kinds of knowledge about MT in paediatric settings (Reid, 2017). Mixed methods studies can provide a more comprehensive understanding of mechanisms at play in the topic being investigated, which in turn will render them more applicable to clinical settings (Bradt et al., 2013). Systematically combining different types of data (known as 'merging') is an essential but challenging part of mixed methods research (Bradt et al., 2013). As the authors point out, Ugglia et al. (2019) did not merge results and theirs can hence not be considered mixed methods research (Bradt et al., 2013). By reporting results separately, they do not take full advantage of integrating the objective data (i.e. HR and HRQoL) with the subjective interview data (i.e. participant perspectives) (Bradt et al., 2013). This combining may lead to new insight into data collection in one dataset based on analysis of another dataset. It may determine the degree to which the exploratory data is supported by the explanatory (causal) data, and it may result in increased depth of understanding of the numerical outcome measures based on interviews (Bradt et al., 2013). For instance, exploratory mixed methods studies that include child and parent perspectives may contribute to our understanding of why children respond differently to MT as procedural support (Ghetti, 2016). RCTs on MT as procedural support prove a meaningful addition to the knowledge base insofar as they are based on a sound theoretical framework and take into account interpersonal interactions, and the intervention is carefully documented (Bradt, 2016). The significance of the interpersonal relationships formed in paediatric MT underlines the importance of also applying research methods other than RCTs, which by extension would strengthen evidence-based practice in paediatric MT (Bradt, 2016).

Implementing music therapy in paediatric departments in Denmark, Sweden and Norway

In 2019 the numbers of professional music therapists working in somatic paediatric care were five in Denmark, five in Sweden and ten in Norway (Bonde, 2014a; Due & Ghetti, 2018; Sanfi & Bonde, 2014).

Denmark

Although several clinical MT trials have been conducted in Denmark, more research performed on Danish patient populations has been requested for implementation of MT in paediatric departments (Bonde, 2014a). The profession is now highly present within healthcare and is facing demands for quantitative research (RCTs), a requirement for the inclusion of MT in Danish national clinical guidelines for evidence-based practice (Bonde, 2014a). According to Bonde (2014a), music therapists are part of a good multidisciplinary collaboration and are often involved in pain management (p. 227). Bonde (2014a) suggests that all larger hospitals may in the future employ at least one music therapist who can provide individualised MT interventions to patients as well as develop MM procedures and products that can be used by other health professionals in the hospital (p. 227). MT is recommended in the national guidelines for palliative treatment of children, young patients and their families for alleviation of pain, distress and anxiety as well as for emotional support (Sundhedsstyrelsen, 2018). According to a recent report, fewer than 12% of Danish hospitals use MT as an intervention (Jensen & Nielsen, 2019), even though Sanfi (2012) showed that MT as procedural support was feasible in the clinical setting, was compatible with the working routines of the paediatric department, and was cost-effective as it saved time for physicians and nurses.

Sweden

MT is offered in Swedish healthcare in accordance with national guidelines and is considered an integral part of palliative care (Socialstyrelsen, 2013). However, only a small number of Swedish music therapists currently work in paediatric departments and have the experience and expertise to work with procedural pain and perform research on this topic. Studies such as the ones by Uggla and colleagues are likely to have a positive influence on the implementation of MT services in more Swedish paediatric departments than currently offer MT. The national recommendations emphasise the benefits of MT for the whole family of a child in hospital but point out the lack of research evidence for the effectiveness of MT interventions (Socialstyrelsen, 2013). A main challenge in Sweden is promoting recognition

of MT within healthcare services and elsewhere, which is a common theme in the Nordic countries and to a great extent dependent on more Nordic research (Hammarlund et al., 2008). Increased demand for professional music therapists is predicted, which will require an increase in funding of MT in healthcare (Socialstyrelsen, 2013).

Norway

MT has roots within the humanities in both Denmark and Norway (Ridder & Bonde, 2019b) and there is a qualitative MT research tradition (Tråsdahl, 2011), but this is changing as there is increasing demand for evidence supporting MT within somatic healthcare. MT is recommended in national guidelines as an intervention in palliative care for children and young people (Due & Ghetti, 2018). Due and Ghetti (2018) performed a focused ethnographic case study in a Norwegian paediatric department identifying key challenges with regard to establishing more full-time music therapist positions and developing MT services in paediatric medical settings in Norwegian hospitals, such as the need for documentation, hospital funding and gaining acceptance within the paediatric team. In some cases implementation of MT may require a change in the hospital culture and beliefs among healthcare professionals (Due & Ghetti, 2018). Documentation of MT processes and outcomes in patient journals may increase referrals to MT by other medical personnel and allow for the best possible multidisciplinary care of the child (Due & Ghetti, 2018). The authors conclude that MT practice is still a long way from being systematically integrated in most paediatric departments in Norway as part of the standard treatment offered (Due & Ghetti, 2018).

Ongoing research

There are currently at least two ongoing projects on MT as procedural and treatment support. One is a collaboration between paediatric oncologic departments at Aarhus University Hospital (Denmark), Rigshospitalet Copenhagen (Denmark) and university hospitals in Oslo (Norway), Tromsø (Norway) and Lund (Sweden), investigating music imagery in conjunction with chemotherapy (Sanfi & Bonde, 2019). Another ongoing project is investigating MT for procedural support and pain relief at Haukeland University Hospital (Norway). There is likely to be an increase in national and multisite trials in the Nordic countries such as these, which can provide not only larger sample sizes and better powered studies but also allow for cross-cultural collaboration and examination, transfer of skills and knowledge and broader expertise (Ghetti, 2016).

Suggestions for future research and clinical practice

Even though MT has been shown to effectively reduce anxiety in children, some children already cope well with procedures, irrespective of the MT intervention (Sanfi, 2012). By identifying the children and families who experience the most distress and are the most vulnerable to suffering from harmful effects, interventions can be focused on those most likely to benefit from MT (Sanfi, 2012, 2016; Yinger & Gooding, 2015). Lack of sufficiently large sample sizes is a general problem in MT research, and larger sample sizes would increase reliability of results (Sanfi, 2012). Also, inclusion of more music therapists would enable assessment of potential therapist effects (Sanfi, 2012). When only one MT intervention is performed (which is often the case with MT as procedural support), it is possible to measure and compare the efficacy with other non-pharmacological and pharmacological interventions (Ærø & Aasgaard, 2011; Sanfi, 2012), which might shed light on important properties of MT. A further topic for future research is the *timing* of MT interventions, which in turn could contribute to the development of a framework for MT support regarding specific procedures and treatments (Uggla et al., 2019). There is little research evaluating long-term outcomes of MT as procedural support interventions (Ghetti, 2016), as there are often limited possibilities for follow-up (Sanfi & Bonde, 2019). Long-term interventional studies would allow for investigations into the potential of MT as procedural support to act as a buffer against traumatisation during and after hospitalisation and whether positive effects are sustained over time (Ghetti, 2016; Sanfi, 2012). Paediatric pain is typically assessed either through evaluation of the child's behaviour, physiological parameters or self-report of pain intensity by the child (Young, 2005). However, pain and anxiety reactions in distressed children can be indistinguishable (Edwards & Kennelly, 2017), and there may be discrepancies between outcomes reported by children and by their parents or staff (Sanfi, 2012; Uggla et al., 2018). It is also important to be aware that when using physiological parameters as outcome measures, these may change in response to musical stimuli without being direct indicators of changes in pain and anxiety levels (Yinger & Gooding, 2015). As a consequence, consistent use across studies of instruments for evaluation of pain and anxiety levels, multidimensional assessment of intervention outcomes, and careful reporting of study design and methodology are important (Young, 2005). Detailed reporting of MT interventions contributes to increased validity and reproducibility of results (Bonde, 2019a; Yinger & Gooding, 2015). Ghetti (2011) and Yinger and Gooding (2015) recommend that future studies aim to develop further the theory of MT as procedural support, as knowledge in a field is acquired through a combination of advances in theory and new empirical evidence. A better understanding of for whom, under which procedures, and in what ways different MT approaches are the most beneficial can help clinical decision-making regarding MT (Sanfi, 2012; Yinger & Gooding, 2015). MT researchers in Denmark,

Sweden and Norway are in a privileged position to make further valuable contributions to the development of the theoretical framework of MT as procedural support. Lastly, efforts to establish standardised referral of hospitalised children to MT services are called for in all three countries.

Final remarks

MT as procedural and treatment support is a specialised practice that normally aims to take into account the individual child and their family, the single procedure or longer treatment, and the multidisciplinary organisation of the paediatric department (Sanfi & Bonde, 2019). The literature indicates that MT as procedural support can lower pain and anxiety levels and improve coping abilities in children undergoing medical procedures and treatment, thereby preventing paediatric medical traumatic stress (Nielsen, 2013; Sanfi, 2012; Uggla et al., 2016). MT is currently being integrated in paediatric care in Denmark, Sweden and Norway (Bonde, 2014a), but further good quality research is needed for increased awareness and implementation of MT as part of evidence-based practice in paediatric care.

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Disclosure statement

The author have no conflicts of interest to declare.