Music Interventions for Stress Reduction

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Short Summary

The goal of this dissertation is to contribute to the body of scientific knowledge on music interventions for stress reduction. To cope with the negative impact of stress, millions of people around the world use tranquilizing medication, which in turn is associated with numerous contraindications and negative side effects. Therefore, there is an urgent need to develop and examine innovative and non-pharmacological interventions for stress reduction, especially for patient populations known to be more vulnerable to stress such as those with mild intellectual disabilities (MID). Moreover, the stress-reducing qualities of music have been associated with a broad range of positive outcomes in both medical and mental healthcare settings. This dissertation therefore aims to increase scientific knowledge on (a) the effects of different types of music interventions on stress-related outcomes, (b) how and why music interventions may specifically lead to stress-reducing effects, and (c) how stress can be accurately assessed, specifically in people with MID. Overall, this dissertation demonstrates that music interventions in the form of both music listening interventions and in the context of music therapy can greatly benefit patients in medical and mental health care settings. Furthermore, the added value of a qualified music therapist offering the music interventions is emphasized, which can be explained by the personalized, tailored approach of the music therapist. Despite the difficulty of examining these music therapy interventions, more robust research is needed, especially in patient populations proven to be more vulnerable to stress as those with cognitive impairments. The results of this dissertation also highlight the need for continued efforts to develop high-quality self-report stress measures for people with the MID to assess stress-related outcomes as valid and reliable as possible. Future research that focuses on both efficacy and hypothesized therapeutic factors is crucial to the further implementation of music therapy interventions in healthcare settings, particularly when it comes to stress reduction.

Contents

CHAPTER 1

General Introduction

1. Brief Introduction

To a greater or lesser extent, almost every human being has to deal with stress, and it therefore becomes part of many people's daily lives. Nevertheless, stress is commonly believed to be one of the major factors that negatively affects our health. More severe and prolonged stress experiences are considered a serious risk factor for the onset and progression of a wide range of physical and emotional problems (American Psychological Association [APA], 2017; Australian Psychological Society [APS], 2015; Cohen et al., 2007). According to the World Health Organization (WHO), stress has been classified as the "health epidemic of the 21st century".

High stress levels have shown to be strongly associated with many physical and emotional problems, such as cardiovascular disease, chronic pain, anxiety disorders, depression, burnout, and addictions (e.g., American Psychological Association [APA], 2017; Howe et al., 2013; McEwen & Gianaros, 2010). Even though people are aware of this negative impact of stress, it seems to be particularly difficult to understand the destructive impact of stress on our daily life experiences (Casey, 2017; de Witte et al., 2020), and to reduce or cope with stress without any professional support (WHO, 2010). To cope with stressors, millions of people around the world use tranquilizing medication, which in turn is associated with numerous contraindications and negative side effects (e.g., Bandelow et al., 2015; Olfson et al., 2015; Puetz et al., 2015). Therefore, there is an urgent need to develop and examine innovative and non-pharmacological interventions for the prevention and management of stress, especially for patient populations who are known to be more vulnerable to stress.

Music interventions can be considered such non-pharmacological intervention, as music has been used to provide calmness and relaxation for many decades, all around the world (Kamioka et al., 2014; Martin et al., 2018; Raglio et al., 2015). These stress-reducing qualities have been identified as the most widely studied effects of music (e.g., Chanda & Levitin, 2013; Koelsch, 2015; Mehr et al., 2019). In the past decade, music listening and music making have been associated with a broad range of positive health and well-being outcomes, resulting in music being increasingly used as a therapeutic intervention to reduce stress in a variety of healthcare settings (Juslin & Västfjäll, 2008; Koelsch, 2012, 2015; Zatorre, 2015). Moreover, the body of empirical studies on the effects of music interventions on stress has increased substantially in recent years, demonstrating the need for a better understanding of the effects of music in general and the specific circumstances in which music interventions lead to these effects.

In this dissertation, we therefore aim to increase scientific knowledge on (a) the effects of different types of music interventions on stress-related outcomes, (b) how and why music interventions may specifically lead to stress-reducing effects, and (c) how stress can be accurately assessed, specifically in people who are more vulnerable to stress, such as those with mild intellectual disabilities (MID).

2. The Construct of Stress

Almost everyone experiences stress from time to time, and stress is an inseparable part of our daily lives. Therefore, for many years, the stress system has been considered as one of the most important human systems. In physiology and medicine, a general definition of stress was introduced in the 50s by Selye (1956): "Stress is a general activation reaction to a stimulus that could mean both a challenge (in a positive way) and a threat (in a negative sense)" (p. 32). Aldwin (2007), on the other hand, emphasized the negative part and defined stress as the quality of an experience, produced through a person-environment transaction that, through either overarousal or underarousal, results in psychological or physiological distress. When we refer to "stress" in this dissertation, we are explicitly relating to the negative impact of stress.

In general, responses to stress can be related to both increased *physiological arousal* and specific *emotional states*, while the underlying systems of both of these responses regulate and affect each other during stress experiences (Linnemann et al., 2017; McEwen & Gianaros, 2010). This can be explained by neuroscientific models of stress, which reveal the neurological pathways that mediate between the neuroendocrine and immune systems during stress experiences and their interaction with brain areas known to be involved with emotional processes (Aldwin, 2007). Emotional responses to stress can be described as emotional states of subjective worry, such as state anxiety, restlessness, or nervousness (Akin & Iskender, 2011; Cohen et al., 1983; Pittman & Kridli, 2011). Physiological responses to stress imply the activation of the hypothalamic-pituitary adrenal (HPA) axis which causes a release of adrenalin and noradrenalin. This increased activity of the sympathetic nervous system in turn results in increased physiological arousal such as heart rate, blood pressure, and cardiac output (e.g., Bally et al., 2003; Pfaff et al., 2007). In a parallel process involving the hypothalamus and the adrenal glands, cortisol is released. To properly evaluate the manifestation of stress, it is important to observe both physiological and psychological outcomes.

The general construct of stress provides a common ground that integrates many scientific fields and provides a comprehensive framework in which both environmental, psychological, and physical factors are all interrelated (Aldwin, 2007; Cohen et al., 2007). We highlight the part of this knowledge that is closely related to the further content of this dissertation. Many theoretical stress models acknowledge the importance of environmental effects on mental health, but they also recognize individual differences in the vulnerability to stress and/or ability to cope with stressors (Aldwin, 2007; Turner, 2003). This latter aspect is particularly relevant and means that specific individual characteristics of people such as their cognitive functioning may lead to greater vulnerability to stress. One of society's most vulnerable groups to whom this applies are people with mild intellectual disabilities (MID[1]). The literature shows that especially in this group, stress can lead to an increase of

1 By common Dutch definition adults with an IQ 50-70 as well as with an IQ 70-85 in combination with a disability in adaptive skills are considered to have a MID. In this article the abbreviation MID is therefore used to include people with what is often referred to as a borderline intellectual disability.

maladaptive coping strategies and serious health issues such as impaired cognitive functions, physical health problems, and substance abuse (e.g. Didden et al., 2009; Heyman & Hauser-Cram, 2015). According to recent studies, patients with MID account for more than 40% of the psychiatric in-patients in the Netherlands (Nieuwenhuis et al., 2021; Seelen-de Lang et al., 2019). Nevertheless, there is still a scarcity of suitable interventions for people with intellectual disabilities (Irvine & Beail, 2017), which strengthen the argument for developing therapeutic stress interventions particularly tailored to the needs and possibilities of people with cognitive impairments, such as those with MID.

3. Theoretical Assumptions on the Relationship between Music and Stress

Through the years, several theoretical models have been developed to gain a better understanding of the influence of music on stress-related outcomes. One of the most widely accepted models is rooted in biological and neurological theories. Previous research shows that music listening is strongly associated with relaxation through its quality of lowering the activity of the autonomic nervous system. This is demonstrated by the decrease of *physiological arousal*, indicated by reduced cortisol levels, lowered heart rate and mean arterial pressure when listening to soothing music (e.g., Burrai et al., 2016; Koelsch et al., 2016; Kreutz et al., 2012; Linnemann et al., 2015). In addition, many neuroimaging studies show that music can modulate those brain areas known to be involved in emotional and motivational processes, such as the limbic system, in particular the amygdala, hippocampus, and the pleasure and reward system. This in turn can be explained by the release of dopamine and endorphins when listening to pleasant music (Blood & Zatorre, 2001; Menon & Levitin, 2005; Koelsch et al., 2015, 2016), both of which are positively associated with a reduction in stress levels (e.g., Amir et al., 1980; Dief et al., 2018).

In addition to the neuroscientific view, it is also relevant to mention theories from social and cognitive psychology. When music is experienced in the presence of others, there is an especially strong correlation with stress relief. This may be explained by the fact that people synchronize with each other during music activities, possibly evoked by rhythmic entrainment which increases feelings of togetherness and social cohesion during the music experience (Boer & Abubakar, 2014; Linnemann et al., 2016; Tarr et al., 2014). From a cognitive perspective, listening to music can serve as a distractor by diverting people's stressful feelings or thoughts to something more pleasant (Bernatzky et al., 2011; Sendelbach et al., 2006).

4. Music Interventions

Worldwide, there is a growing recognition of the valuable role that music can play in improving health and wellbeing (Bainbridge et al., 2021; Fancourt & Finn, 2019), whether in everyday life in music programs designed for health and wellness, or in specific therapeutic interventions offered by qualified (music) therapists in various kinds of healthcare settings (Fancourt, 2017; Juslin & Västfjäll, 2008). The term "music interventions" is generally used as an umbrella term for all therapeutic interventions in which *music* is the key component in achieving the desired treatment effect. Music interventions can be regarded as purposeful musical exercises or methods in which music listening, music making, or singing is central. Music interventions can be offered by a music therapist, in the context of music therapy, but can also be self-administered or offered by other (healthcare) professionals, or can concern music making or singing outside a therapeutic context (American Music Therapy Association [AMTA], 2018). In the scientific literature, however, a clear distinction is made between evidence regarding music listening interventions administered by medical or healthcare professionals, also referred to as "music medicine", and music interventions offered by a qualified music therapist within a therapeutic context, referred to as music therapy (Agres et al., 2021; Bradt et al., 2013; Magee, 2019). In both the context of music medicine and music therapy, receptive music interventions are offered, meaning that the patient does not actively make music but only listens to the music.

4.1 Music Listening Interventions in the Context of Music Medicine

Thus, receptive music interventions can be categorized as "music medicine" when listening to pre-recorded music offered by medical professionals or if self-administered by the patient (Dileo, 1999). These music interventions are mostly offered in medical settings to enhance or facilitate medical treatment or to assist rehabilitation. The music is used to positively influence a patient's physical, mental or emotional states before, during, or after medical procedures (Dileo, 1999; Trondalen & Bonde, 2012). Patients usually select their preferred music since music disliked by the patient could increase stress levels (Bradt & Dileo, 2014; Heiderscheit et al., 2011).

4.2 Music Therapy

Music therapy can be defined as the clinical and evidence-informed use of music interventions to accomplish individualized goals in a therapeutic relationship in order to achieve physical, emotional, mental, social and cognitive needs (Aalbers et al., 2019; Agres, et al., 2021; AMTA, 2018). Although the term "music therapy" in the literature often refers to any kind of use of music as an intervention in health care settings, music therapy should be offered by a trained, and qualified music therapist, with the required knowledge and expertise in psychology, medicine, and music (Agres et al., 2021; AMTA, 2018; Bradt et al., 2015; Magee, 2019).

Receptive music therapy interventions refer to listening to *live* or *prerecorded* music offered by the music therapist. Afterwards, the patient may verbally process their own emotions and/or experiences. Active interventions, on the other hand, involve the patient actively doing something during the music therapy sessions, such as musical improvisation, composing music or songs, or singing (Bruscia, 1998; Wheeler, 2015).

During both active and receptive music interventions, music therapists use the unique qualities of music (e.g., melody, rhythm, tempo, dynamics, pitch) in a therapeutic relationship to access a patient's emotions and memories, to address social experiences, or to influence behavior (Bruscia, 1987; Wheeler, 2015). This specific kind of responsiveness to the patient's needs can be regarded as the key competence of the music therapist (Agres et al., 2021). More specifically, during music therapy the music therapist tunes in to the patient by adjusting the way of music-making as an immediate response to the patient's needs (e.g., Aalbers et al., 2019; Magee, 2019). It is precisely this experience-based approach to music therapy that is particularly well suited to the needs of people with MID, for whom more cognitive approaches to intervention are not often the best fit (Hooper et al., 2011; Moonen & Didden, 2014).

5. The Outline and Aims of this Dissertation

5.1 Identifying Overall Effects and Effect-Moderating Factors (Chapter 2 and 3)

The scientific literature shows that the number of studies examining the effects of music interventions on stress-related outcomes is growing rapidly. The first aim of this dissertation was to examine whether and the degree to which music interventions, including both music listening interventions and music therapy interventions, are effective in reducing stress in various patient populations. Despite the large number of studies and the theoretical explanatory models addressing the relation between music and stress, it has proved to be difficult to deduce from the large number of available studies how strong the effect of music interventions on stress reduction is, and what exactly contributes to the positive effects of these interventions. A related aim was therefore to also examine possible moderator effects of study, sample, and intervention characteristics, which could influence the strength of the effects of music interventions on stress-related outcomes.

Chapter 2 presents the results of a systematic review and two multilevel meta-analyses of 104 randomized controlled trials (RCT) on the effects of music interventions on both physiological and psychological stress-related outcomes. Chapter 3 presents the results of a systematic review and meta-analysis, which included 47 trials (RCT and CCT) on particularly the effects of music therapy interventions on stress reduction.

5.2 Collecting Practice-Based Knowledge on Music Therapy Interventions (Chapter 4)

It is well-known that music therapists often perform their interventions based on rich, experiential, tacit knowledge. It is important for both clinical practice and future research to not only integrate evidence from outcome studies, but also systematically illuminate the knowledge already present in clinical practice, such as the tacit knowledge of music therapists. The second aim was therefore to systematically collect practice-based knowledge on *how* music therapists lower the stress levels of their patients with MID during music therapy sessions. Chapter 4 presents the results of a qualitative focus group study, including music therapists from different countries and clinical institutions in Europe working with adults with MID (N = 13).

5.3 Providing more Insights into How to Assess Subjective Stress Experiences (Chapter 5)

As persistent stress can lead to the development of psychopathology and severe physical conditions, it is increasingly important to timely recognize stress-related symptoms, especially in populations known to be more vulnerable to stress, such as people with MID. Although more and more research is being conducted on the use of physiological measures to assess people's stress levels, it is clear that in addition to these measures, self-report measures are still necessary to gain the correct understanding of an individual's perception of stress. The third aim of this dissertation was therefore to provide (a) more insights into the validity and reliability of existing self-report stress measures and (b) more information on their suitability in adults with MID. Chapter 5 presents a scoping review in order to provide an overview and evaluation of the existing self-report stress measures, complemented with an expert consultation to provide more information about their suitability for adults with MID.

5.4 Developing a Music Therapy Micro-Intervention for Stress Reduction (Chapter 6)

The development of music therapy interventions for stress reduction is becoming increasingly important, particularly in populations where needs may be greater such as people with MID or other cognitive impairments. Since the previous studies of this dissertation showed precisely the direct effects and applicability of music interventions for stress reduction, the concept of micro-interventions fits the best for this purpose. To integrate the perspectives from both empirical literature and clinical practice, the fourth aim was to provide a detailed description of a music therapy micro-intervention for stress reduction which can be used by music therapists as well as provide a foundation for future research. Chapter 6 presents the development process of a music therapy micro-intervention through an iterative process aimed at integrating theory-, evidence-, and practice-based knowledge, complemented by expert evaluation.

5.5 Overall Conclusions

Chapter 7 provides the overall conclusions of this dissertation. The findings of the studies are discussed in the light of previous research on music interventions for stress reduction and the influence of music on the stress response in general. Additionally, the strengths and limitations of the dissertation are noted. The chapter concludes with directions for future research.

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CHAPTER 2

2

Effects of Music Interventions on Stress-Related Outcomes: A Systematic Review and Two Meta-Analyses

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Abstract

Music interventions are used for stress reduction in a variety of settings because of the positive effects of music listening on both physiological arousal (e.g., heart rate, blood pressure, and hormonal levels) and psychological stress experiences (e.g., restlessness, anxiety, and nervousness). To summarize the growing body of empirical research, two multilevel meta-analyses of 104 RCTs, containing 327 effect sizes and 9,617 participants, were performed to assess the strength of the effects of music interventions on both physiological and psychological stress-related outcomes, and to test the potential moderators of the intervention effects. Results showed that music interventions had an overall significant effect on stress reduction in both physiological (d = .380) and psychological (d = .545) outcomes. Further, moderator analyses showed that the type of outcome assessment moderated the effects of music interventions on stress-related outcomes. Larger effects were found on heart rate (d = .456), compared to blood pressure (d = .343) and hormone levels (d =.349). Implications for stress-reducing music interventions are discussed.

Keywords: music interventions, music therapy, arousal, stress, state anxiety, multilevel meta-analysis.

1. Introduction

Stress is believed to be one of the major factors negatively affecting our health. High stress levels have shown to be strongly associated with many physical and emotional problems, such as cardiovascular disease, chronic pain, anxiety disorders, depression, burnout, and addictions (American Psychological Association [APA], 2017; Australian Psychological Society [APS], 2015; Casey, 2017; Howe et al., 2013; McEwen & Gianaros, 2010). Furthermore, there is a strong relationship between these stress-related health problems and higher absenteeism at work (UK Health and Safety Executive, 2016). To cope with stressors, millions of people around the world use tranquilizing medication, which is associated with numerous contraindications and negative side effects (e.g., Bandelow et al., 2015; Olfson et al., 2015; Puetz et al., 2015). Because of the difficulty of reducing or preventing stress without any professional support and the great demand for nonpharmacological stress reduction interventions, the relevance of the development of cost-effective interventions for stress reduction is high (Casey, 2017; Holahan et al., 2005; Howe et al., 2013; McEwen & Gianaros, 2010; World Health Organization [WHO], 2010).

Music listening and music making have been associated with a broad range of positive outcomes in the domains of health and well-being (Juslin & Västfjäll, 2008; Koelsch, 2012, 2015; Thaut & Hoemberg, 2014; Zatorre, 2015). The most widely studied effects of music are the calming and stress reducing effects (Chanda & Levitin, 2013; Gillen et al., 2008; Juslin & Västfjäll, 2008; Koelsch, 2015). For decades, music has been used as an intervention for stress reduction, such as music activities (like singing or music making), music listening for a certain patient group ("music medicine"), and live music therapy offered by music therapists (Bradt et al., 2013; Gold et al., 2011).

In order to integrate the available knowledge on the effects of music interventions on stress, the present study is a systematic review and meta-analysis of experimental studies testing the effects of music interventions on both physiological and psychological stress-related outcomes in clinical, medical and work- or study-related settings.

1.1 The Stress System

The stress system can be considered as a highly important and preserved system in human beings. In physiology and medicine, the general definition of *stress* is introduced by Selye (1956): *"Stress is a general activation reaction to a stimulus that could mean both a challenge (in a positive way) and a threat (in a negative sense)"* (p. 32). Aldwin (2007) emphasized the negative part and defined stress as the quality of an experience, produced through a person-environment transaction that, through either overarousal or underarousal, results in psychological or physiological distress (Aldwin, 2007; Riley & Park, 2015). The responses to stress can be categorized as *physiological arousal* and *emotional responses* (e.g. Aldwin, 2007; Li & Goldsmith, 2012; Pelletier, 2004). Together, the underlying systems of these responses

regulate stress and affect each other during stress (e.g., Linnemann et al., 2017; McEwen & Gianaros, 2010).

The physiological response to stress implies the activation of the hypothalamic-pituitary adrenal (HPA) axis and, because of a release of adrenalin and noradrenalin, increased activity of the sympathetic nervous system resulting in increased physiological arousal, such as heart rate, blood pressure, and cardiac output (Bally et al., 2003; McCance et al., 2006; Pfaff et al., 2007). In a parallel process, involving the hypothalamus and the adrenal glands, cortisol is released. The emotional response to stress can be described as emotional states of subjective worry, such as state anxiety, restlessness or nervousness (Akin & Iskender, 2011; Cohen et al., 1983; Pittman & Kridli, 2011; Pritchard 2009). *State anxiety* has been defined as an emotional response to an individual's perception of a stressful experience (e.g., Hook et al., 2008; Koelsch et al., 2011a; Ng et al., 2016; Zhang et al., 2014). Therefore, state anxiety was operationalized for several decades as one of the psychological stress-related outcomes (Koelsch et al., 2011a; Lazarus, 1966; Pelletier, 2004). It is assumed that both the physiological and the emotional responses of stress may be reduced by music (Bradt & Dileo, 2014; Dileo & Bradt, 2007; Pelletier, 2004).

1.2 Effects of Music on Stress

Recent neuroscientific studies provide insights into how music interventions may lead to stress reduction and increased well-being. Firstly, music seems to be able to decrease *physiological arousal,* which is increased during stress. Music listening, and music making/singing, have been associated with decreases of physiological arousal, shown by reduction of cortisol levels or decrease in heart rate and blood pressure (Hodges, 2011; Koelsch et al., 2016; Kreutz et al., 2012; Leardi et al., 2007; Linnemann et al., 2015; Nilsson, 2009; Sokhadze, 2007). These three outcomes have been identified in neurobiology as distinct stress biomarkers (Cacioppo et al., 2007; Pfaff et al., 2007).

Music may also affect stress-related *emotional states*, such as subjective worry, anxiety, restlessness or nervousness (Akin & Iskender, 2011; Cohen et al., 1983; Pittman & Kridli, 2011; Pritchard, 2009). This is because music can modulate activity in brain structures that are known to be crucially involved in emotional processes. Recent neuroimaging studies on music and emotion showed that music may strongly influence the *amygdala*, a part of the limbic system, which is a section of the brain that plays a crucial role in the regulation of emotional processes by releasing endorphins. These neurotransmitters play an important role in enhancing a sense of well-being (Blood & Zatorre, 2001; Hodges, 2011; Koelsch, 2015; Koelsch et al., 2011b; Levitin, 2009; Moore, 2013; Thaut & Wheeler, 2010; Uhlig et al., 2013; Zatorre, 2015).

The systematic review of Moore (2013) on the neurological effects of music on emotional processes indicated that musical improvisation and music listening could deactivate the amygdala, which may decrease the intensity of stress-related emotional states and psychophysiological arousal. This in turn has been shown to evoke feelings of pleasure and

happiness (Blood & Zatorre, 2001; Koelsch et al., 2016; Koelsch et al., 2010; Limb & Braun, 2008). The (cognitive) behavioral framework is consistent with this and takes into account that music can serve as a distractor, diverting attention from a stressful event to something more pleasant, which reduces stress levels (Sendelbach et al., 2006; Vaajoki et al., 2011).

Many studies on the effects of music considered *state anxiety* to be a stress-related emotional state, examining relationships between state anxiety outcomes and physiological stress-related outcomes (e.g., Hook et al., 2008; Koelsch et al., 2011a; Ng et al., 2016; Zhang et al., 2014). Although the terms *state anxiety* and *stress* are used interchangeably in the psychology literature, different self-reporting questionnaires are used. It is therefore necessary to examine whether music has the same effects on stress and state anxiety.

1.3 Music Interventions

Music interventions can be regarded as purposeful musical exercises or methods in which music listening, music making, or singing is central. In both literature and practice there is a distinction between music interventions offered by a music therapist and music interventions offered by other healthcare professionals or without any support. First, music interventions can be defined as purposeful music activities if they concern listening to prerecorded music offered by medical or healthcare professionals, if the intervention is self-administered by the patient ("music medicine"), or if it concerns music making or singing without the involvement of a music therapist or a therapeutic context (American Music Therapy Association, 2018). Second, music interventions as a part of *music therapy* are offered by trained music therapists and are characterized by the presence of a therapeutic process and the use of personal music experiences (Bradt & Dileo, 2014; Bradt, Dileo, & Shim, 2013; Dileo, 2006; Gold et al., 2011; Kamioka et al., 2014). Music interventions in the practice of music therapy may concern music listening or music playing, but may also include composing, songwriting, or interacting with music (Leubner & Hinterberger, 2017).

It is assumed that specific characteristics of the music may have an impact on the stress-reducing effects of music interventions (e.g., Bradt & Dileo, 2014; Bradt, Dileo, & Potvin, 2013; Pittmann & Kridli, 2011). *Music tempo* can be considered as one of the most important moderators of music-related arousal and relaxation. Music with a slow tempo (60-80 bpm), for example meditative music, has often been associated with reductions in heart rate, resulting in greater relaxation (e.g. Bernardi et al., 2006; Bringman et al., 2009; Chlan, 2000; Hilz et al., 2014; Nomura et al., 2013). The use of *instrumental music*, instead of *music with lyrics*, would often lead to greater effects of music interventions on stress reduction. Several studies reported that music containing lyrics may be more distracting and activating instead of calming (Good et al., 2000; Halpern & Savary, 1985). However, Koelsch et al. (2011) reported that the use of music with lyrics may reinforce the positive effects of music interventions on stress reduction through the possible comforting effects of the lyrics. Another component of music interventions for stress reduction is the way the music is played (live music or prerecorded music). Music therapy consists mainly of live music interventions,

which are assumed to be more effective than "music medicine" interventions because music therapists individualize their interventions to meet patients' specific needs (Bradt & Dileo, 2014; Dileo, 1999, 2006). Notably, some studies measured differences in stress responses between participants receiving live music and those receiving prerecorded music, with live music appearing to be the most stress-reducing (e.g., Arnon et al., 2006; Bailey, 1983; Baker, 2001).

Many studies examining the effects of music interventions on stress-related outcomes in specific patient groups or settings have been published, such as cancer patients (Bradt et al., 2011), coronary heart disease patients (Bradt, Dileo, & Potvin, 2013), and patients undergoing endoscopic procedures (Rudin et al., 2007). Several studies reported positive effects of music-listening on stress-related outcomes. In medical settings, listening to tranquilizing music before, during, and after medical procedures has been reported to correlate with lower cortisol levels, associated with the reduction of stress and/or anxiety (e.g., Chanda & Levitin, 2013; Kamioka et al., 2014; Koelsch et al., 2016; Linnemann et al., 2015; Nilsson, 2008). However, the strength of the effect differs both within and between studies, and the particular impact of potential moderators – such as patient/client, setting, measurement and intervention characteristics – is largely unknown.

1.4 Rationale for the Present Study

The present study consists of two multilevel meta-analyses on the effects of music interventions on both physiological stress-related arousal (e.g., blood pressure, heart rate, hormone levels) and psychological stress-related experiences (e.g., state anxiety, restlessness or nervousness) in various populations and settings, and is a replication of the meta-analysis by Pelletier (2004), who reviewed 22 quantitative studies examining the effects of music interventions on stress reduction. Pelletier's (2004) meta-analysis showed that music alone and music-assisted relaxation significantly reduced stress-related arousal, with an overall medium-to-large effect size of d = .67, moderated by study characteristics, such as age, type of stress, musical preference and type of intervention. An important difference with the present study is that Pelletier included only studies in which the intervention consisted of listening to prerecorded music, often combined with relaxation techniques, whereas studies with live music interventions were excluded. Besides, studies examining *music therapy* for stress reduction were not included. The quality of the included studies was not assessed. Therefore, it is not clear whether the methodology of the studies did influence Pelletier's overall effects. In addition, we assume that the study quality of comparable studies has increased over the last 15 years.

In the last decade, music interventions were increasingly developed and used to reduce stress in a variety of settings (Chanda & Levitin, 2013; Heiderscheit et al., 2011; Koelsch, 2015; Uhlig et al., 2013), and to support physical and psychological health by creating an environment that stimulates relaxation and stress reduction (Bradt & Dileo, 2014; Kamioka et al., 2014; Koelsch & Stegemann, 2012; Nilsson, 2008). Therefore, it is timely to replicate

the meta-analysis by Pelletier (2004) by using new multilevel meta-analytic techniques that enable moderator analyses of both within and between study differences in outcomes, thus preventing loss of information and increasing statistical power (Assink & Wibbelink, 2016).

The present meta-analytic study includes all *randomized controlled trials* (RCTs) on the effects of music interventions on stress-related outcomes in adults, who are not suffering from dementia, that have been published. The first aim of this study is to examine whether, and the degree to which, music interventions are effective in reducing stress. The second aim is to examine possible moderator effects of study, sample and intervention characteristics, which may influence the strength of the effects of music interventions on stress-related outcomes. The results of this meta-analysis may be used to increase the effects of music interventions on stress by examining the conditions under which music interventions have the largest effects on both physiological and psychosocial dimensions of coping and stress.

2. Method

2.1 Inclusion Criteria

For the current meta-analysis, multiple inclusion criteria were applied. Firstly, only RCTs that examined the effect of music interventions on the experience of stress and/or (state) anxiety were included. Outcome measures related to quality of life (QoL) or pain were excluded, because these outcomes can be a response to stress, but are not measuring the primary stress-related outcome measures, upon which the present study is focused. The physiological effects of stress had to be measured by heart rate, blood pressure and hormone levels. Psychological effects of stress had to be measured by self-report instruments aiming at 'stress' or 'state anxiety'. Secondly, studies including participants younger than 18 years of age or examining people with dementia were excluded. Although many studies reported cognitive and emotional benefits in dementia patients when they were singing or when they were listening to familiar songs (Särkämo et al., 2008, 2014), such findings are not directly related to 'stress reduction'. Moreover, the regular stress measurement instruments, which are also used in the included RCTs of the present study, are not used in studies in which people with dementia are being examined.

2.2 Selection of Studies

We conducted a computer-based search of the psychological and medical electronic literature databases, including Medline, Academic Search Complete, Cochrane Library, Web of Science, Wiley Online Library, SpringerLink, PubMed, PiCarta, Academic Search Premier, ScienceDirect, PsychINFO and Google Scholar. Pelletier's meta-analysis (2004) can be seen as the starting point of current meta-analysis because Pelletier also included all kinds of settings and patient groups. All RCTs available until November 2017 that were in line with the inclusion criteria were included in this meta-analysis. The electronic databases were

searched using the following English search string: (music*) AND (stress* OR anxiety* OR arousal) AND ("randomized controlled trial" OR "randomised controlled trial" OR RCT). Furthermore, reference sections of review- and meta-analytic articles about the effect of music interventions on stress-related outcomes were inspected for qualifying studies. The initial search resulted in 2679 studies. Finally, 104 studies met all the inclusion criteria (Figure 1). An overview of the included studies and their main characteristics are presented in Table A1 (Appendix 1, see page 250).

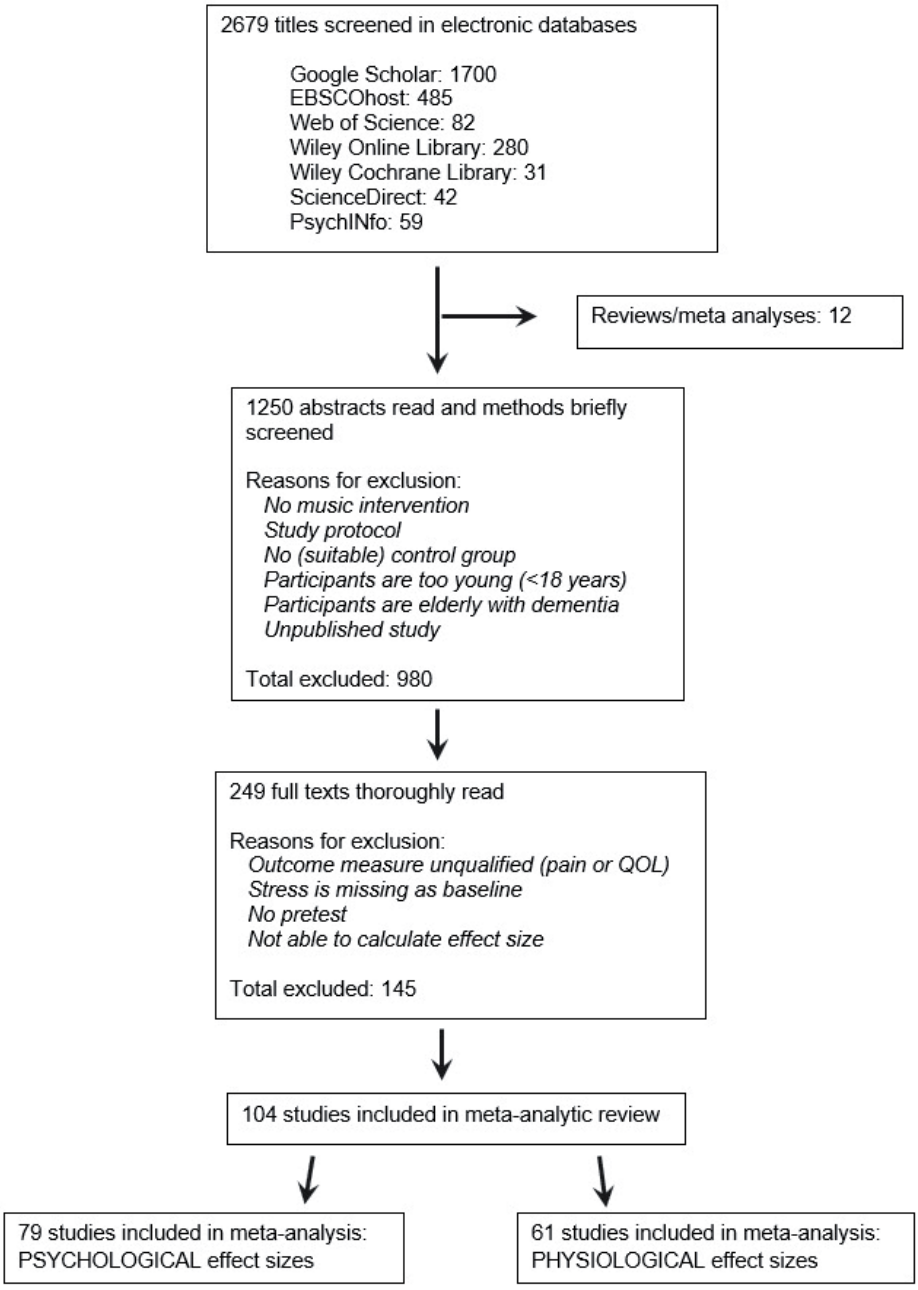
2.3 Coding and Moderators

The included studies were coded by the first author using a coding sheet according to the guidelines of Lipsey and Wilson (2001). *Stress* can be considered as the dependent variable and was coded into *physiological* or *psychological* stress-related outcomes, resulting in two meta-analyses. For each meta-analysis, various factors with a potential moderating effect on the relation between music interventions and stress were identified. These moderators were divided into outcome-, study-, sample-, and intervention characteristics.

Regarding the stress-related outcomes, three different physiological outcome measures were coded: *heart rate, blood pressure* and *hormone levels*. These are three different biomarkers and each biomarker must be measured differently (Cacioppo et al., 2007; Pfaff, et al., 2007). Regarding the psychological stress-related outcomes, it was coded whether the outcomes were assessed by means of questionnaires measuring *stress* or *state anxiety*. For measuring stress, the self-report questionnaires Perceived Stress Scale (PSS; Cohen et al., 1983) and the Visual Analogue Scale Stress (VAS-S) are widely used and 19% of the included studies measuring psychological stress-related outcomes have applied them. State anxiety is measured in multiple studies by the state version of the Spielberger State-Trait Anxiety Inventory (STAI-S; Spielberger et al., 1983), and is used in 59% of the included studies assessing psychological stress-related outcomes (k = 76). Notably, despite the positive psychometric features of the STAI-S, the measures have been criticized for their inability to adequately discriminate between the symptoms of anxiety and depression (Caci et al., 2003; Grös et al., 2007).

We coded whether the study quality was *strong, moderate* or *weak* with the 'Quality Assessment Tool for Quantitative Studies' (Effective Public Health Practice Project [EPHPP], 2009). This tool assesses the quality of a study by providing a comprehensive and structured assessment of the concept of study quality (Armijo-Olivo et al., 2012). The EPHPP has been reported to have content and construct validity (Jackson & Waters, 2005; Thomas et al., 2004). Low-quality studies negatively affect the internal (causal conclusion) validity, which can lead to a biased estimation of the overall effect estimate (Higgins & Green, 2011; Zeng et al., 2015). Regarding the setting in which the study was conducted, we coded whether the study was conducted in a *polyclinic medical setting*, *pre-, during or after medical surgery*, or whether it was a so-called *nonmedical setting* (e.g., work-related settings, research among students at universities or musical activities to improve health in chronic patients). Further, we coded

Figure 1
Flow Chart of the Search



whether the study was conducted in a *Western* country (European countries, Australia, USA, Canada, New Zealand) or in a non-Western country (mainly Asiatic countries). The cultural environment has been shown to influence the way people respond to and cope with stress (Lonner, 2007; Tweed et al., 2004), which could influence the effect of music on stress. The type of control condition was coded as care as usual (CAU) or another intervention. Different control conditions can lead to different effect sizes and recognizing this is crucial for drawing accurate conclusions about treatment efficacy (Finney, 2000; Karlsson & Bergmark, 2015).

Several sample characteristics were also coded, such as the percentage of men in each study. There are indications men and women tend to react differently to stress, both psychologically and physiologically, leading to substantiated gender differences in measured stress levels (Galanakis et al., 2009; Kajantie & Phillips, 2006; Verma et al., 2011). We also coded the average age of the samples per study because research on occupational stress shows the existence of several significant differences in stress levels between different ages (Galanakis et al., 2009).

Finally, we coded six music intervention characteristics. First, according to Bradt and Dileo (2014), we coded whether the music intervention was offered in the context of *music therapy* by a trained music therapist, or whether the music intervention was offered by a healthcare professional, the researcher, or by the patient himself, as a *music activity* (Dileo, 2006; Dileo & Bradt, 2007; Kamioka et al., 2014; Leubner & Hinterberger, 2017). Second, we coded whether the music intervention involved *live music* or *prerecorded music*, because differences in effects on stress-related outcomes were found (e.g., Arnon et al., 2006; Bailey, 1983; Baker, 2001). Third, regarding the selection of music, it was coded whether the music was self-selected by the participants and based on *own preference* (bringing their own music) or whether the music was *pre-selected* by the researcher or the (music) therapist. Some researchers have advised to allow participants to choose the music themselves, because this may have a greater stress reducing impact (Brannon & Fiest, 2007; Juslin et al., 2008), but this does not always mean that participants could bring their own music. On the other hand, the pre-selected music (by the researchers) is mostly based on the musical characteristics related to classical and soothing music, which are assumed to positively affect relaxation and stress reduction (Burns et al., 2002; Labbé et al., 2007). Fourth, we coded whether the music intervention concerned the music tempo of 60–80 bpm, or whether no specific music tempo was mentioned in the study. Fifth, it was coded whether the music contained lyrics or whether the music was purely instrumental. Lastly, we coded the number of music intervention sessions. The number of interventions is positively correlated to improvements in many outcomes including the regulation of stress and anxiety (Cassileth et al., 2003; Gold et al., 2009; Robb et al., 2011).

2.4 Calculation and Analyses

Data analysis was performed by the first and second author. The effect sizes were transformed into Cohen's *d* by use of Wilson's (2013) calculator and Lipsey and Wilson's (2001) formulae.

Negative effect sizes indicate that music interventions had a negative effect on stress-related outcomes. Most *d*-values were calculated based on reported means and standard deviations. To correct for pretreatment differences, pretest effects were subtracted from posttest effects. The effect size was coded as zero when a study mentioned that an effect was not significant without providing any statistics (Lipsey & Wilson, 2001). For both meta-analyses, the continuous moderators (age of the participants, gender of the participants, duration of the music intervention and frequency of the music intervention) were centered on their means. For categorical variables, dichotomous dummy variables were created. Extreme outliers in effect sizes were identified using box plots (Tabachnik & Fidell, 2013), and were winsorized (i.e., replaced by the highest or lowest acceptable score falling within the normal range) for both meta-analyses. Standard errors were estimated using Lipsey and Wilson's (2001) formulae.

In almost all the studies it was possible to calculate more than one effect size as most studies reported on multiple stress-related outcome variables, multiple scales or measurement instruments. It is possible that the effect sizes from the same study are more alike than effect sizes from other studies. The assumption of independent effect sizes underlying traditional meta-analytic methods was therefore violated (Hox, 2010; Lipsey & Wilson, 2001). In line with recently conducted meta-analyses, we applied a multilevel approach in both meta-analyses in order to deal with the interdependency of effect sizes (Assink et al., 2015; Cheung, 2014; Houben et al., 2015; Spruit et al., 2016; Ter Beek et al., 2018).

A three-level meta-analytic model was used to calculate the combined effect sizes and to perform the moderator analyses. Three sources of variance were modelled, including the sampling variance for each effect sizes (level-one), the variance between effect sizes within studies (level-two), and the variance between studies (level-three) (Assink & Wibbelink, 2016). The meta-analysis was conducted in R (version 3.4.3) with the metafor package, employing a multilevel random effects model (Houben et al., 2015; van den Bussche et al., 2009; Viechtbauer, 2010). This model is adequate and often used for multilevel meta-analyses and is, in general, superior to the fixed-effects approaches used in traditional meta-analyses (van den Noortgate & Onghena, 2003). We used likelihood ratio tests to compare the deviance scores of the full model and the models without variance parameters on level two or three to determine if the level-two and -three variances were significant, indicating heterogeneity of effect sizes. A heterogeneous effect size distribution indicates that the effect sizes cannot be treated as estimates of a common overall effect size. In that case, we conducted moderator analyses, because the differences among effect sizes may be explained by outcome, study, sample, and/or intervention characteristics.

2.5 Publication Bias

A common problem in conducting a meta-analysis is that studies with nonsignificant or negative results are less likely to be published than studies with positive and significant results. The studies included in this meta-analysis may therefore not be an adequate representation

of all studies that have been conducted. This phenomenon is called the "file drawer problem" (Rosenthal, 1995).

In order to check the presence of publication bias in the current meta-analysis, a trim and fill procedure was performed (Duval & Tweedie, 2000a, 2000b) by testing the asymmetry of the funnel plot according to Egger's method (Egger et al., 1997). In case of publication bias, the funnel plot of the distribution of effect sizes is asymmetric, resulting in a significant Egger's test. If Egger's method indicates publication bias, a trim and fill procedure is required. We tested if effect sizes were missing on the left side of the distribution, since publication bias would only be likely to occur in the case of nonsignificant or unfavorable (i.e., negative) results. In case of left-sided funnel plot asymmetry, we imputed estimations of effect sizes of missing studies through trim and fill analyses, and subsequently computed an overall effect size that would take the influence of publication bias into account (Duval & Tweedie, 2000b), providing an estimate of the degree to which publication bias might have affected the overall mean effect size.

3. Results

This meta-analytic review included 104 RCTs (all non-overlapping samples) with a total of *N* = 9.617 participants of whom *n* = 4.838 participated in a music intervention group or music therapy group, and *n* = 4.779 constituting the comparison group. Table A1 shows an overview of the most important characteristics of the included studies (see Appendix 1). Table 2 shows the overall effects of music interventions on both physiological stress-related outcomes and psychological stress-related outcomes.

Table 2
Overall Effects of Music Interventions on Physiological and Psychological Stress–Related Outcomes

Outcome	*s*	*k*	Mean *d*	95% CI	*p*	σ^2_{level2}	σ^2_{level3}	% Var. level 1	% Var. level 2	% Var. level 3
Physiological outcomes	61	197	0.380	0.296 – 0.465	<.00[1]**	0.024***	0.076***	32.44	16.09	51.47
Psychological outcomes	79	130	0.545	0.432 – 0.657	<.001***	0.119***	0.128***	15.38	40.76	43.87

Note. *s* = number of studies; *k* = number of effect sizes; CI = confidence interval; Mean *d* = mean effect size (*d*); CI = confidence interval; % Var = percentage of variance explained; σ^2_{level2} = variance between effect sizes within the same study; σ^2_{level3}= variance between studies.

3.1 Effect of Music Interventions on Physiological Stress-Related Outcomes

The meta-analysis on the effect of music interventions on physiological stress-related outcomes contained 61 independent studies (*s*), from which 197 effect sizes (*k*) were denied, and a total sample of *N* = 3.188 participants, of which *n* = 1.624 participants in the music intervention groups, and *n* = 1.564 participants in the comparison groups.

3.1.1 Overall effect on physiological stress-related outcomes

A significant small-to-medium effect (*d* = 0.380, *p* = ≤ .001) of music interventions on physiological stress-related outcomes (heart rate, blood pressure, stress-related hormones) was found, indicating that music interventions reduced physiological stress symptoms. According to the trim-and-fill plot, the presence of publication bias was unlikely, as there were no imputed effect sizes on the left side of the funnel (see Figure 2). The likelihood ratio test showed that significant variance was present at the between-study level (level 3) and the within studies level (level 2). In cases of heterogeneous effect size distributions, moderator analyses are advised to assess whether the variance between the effect sizes can be explained by moderators. Therefore, we conducted moderator analyses on type of outcome, study, sample and music intervention characteristics to examine the effect of music intervention on physiological stress-related outcomes.

3.1.2 Results of moderator analyses on physiological stress-related outcomes

The moderator analyses of the physiological stress-related outcomes were justified by significant overall heterogeneity for all moderator variables, including the variables with missing values. The results of these moderator analyses are presented in Table 3.

Outcome characteristics. We found a significant moderating effect (*p* = ≤ .05) on the different types of physiological stress-related outcomes (heart rate, blood pressure or stress-related hormones). The strongest effects of music interventions on the physiological stress-related outcomes were measured by heart rate (*d* = 0.456*)* compared to blood pressure (*d* = 0.343) and hormone levels (*d* = 0.349).

Study characteristics. No significant moderating effects of study characteristics were found. More specifically, the type of setting (non-medical settings, pre- or post-medical surgery or during medical procedures), the continent where the study was conducted (Western or non-Western countries), and the quality of the study (weak, moderate or strong) showed no significant moderating effects. The continent where the study had been carried out showed a trend (*p* = 0.089), indicating that non-Western studies yielded larger effects on physiological stress-related outcomes than studies conducted in Western countries.

Sample characteristics. The age and gender of the samples did not have a moderating effect on the physiological stress symptoms.

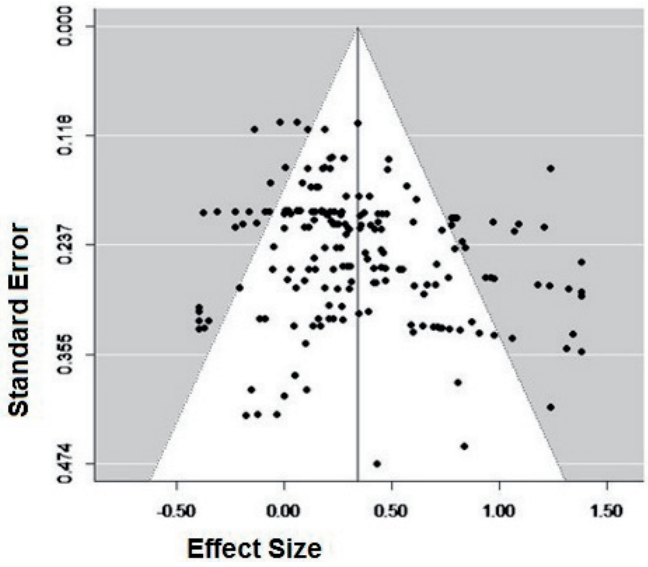
Intervention characteristics. No significant moderating effect was found in the type of intervention (music therapy or music intervention), type of control condition (CAU or another

Table 3
Moderator Effects of Music Interventions on Physiological Stress–Related Outcomes

Moderator variables	*s*	*k*	β_0 (mean *d*)	t_0	β_1	t_1	*F*(df1, df2)
Outcome characteristics	61	197					$F(2, 193) = 3.581^{*}$
Bloodpressure (RC)	47	104	0.343	7.141^{***}			
Hormones	13	22	0.349	3.949^{***}	0.006	0.067	
Heart rate	53	70	0.456	8.944^{***}	0.113	2.617	
Study characteristics							
Setting	61	197					$F(2, 194) = 1.099$
Surgery (RC)	27	110	0.393	6.370^{***}			
Nonmedical	10	20	0.523	4.299^{***}	0.130	0.953	
Polyclinical procedures	24	67	0.320	4.677^{***}	−0.073	−0.794	
Continent	61	197					$F(1, 195) = 2.914$
Western (RC)	30	108	0.306	5.097^{***}			
Non–Western	31	89	0.450	7.659^{***}	0.143	1.707	
Study quality	61	197					$F(2, 194) = 0.586$
Strong (RC)	28	92	0.405	6.413^{***}			
Moderate	20	52	0.313	4.052^{***}	−0.093	−0.928	
Weak	13	53	0.426	4.596^{***}	0.021	0.190	
Intervention characteristics							
Type of music intervention	61	197					$F(1, 195) = 0.094$
Music activity (RC)	54	183	0.379	8.346^{***}			
Music therapy	7	14	0.423	2.931^{**}	0.046	0.307	
Type of control condition	61	197					$F(1, 195) = 2.784$
CAU (RC)	46	130	0.417	8.641^{***}			
Other intervention	18	67	0.284	3.948^{***}	−0.133	−1.668	
Music selection	61	197					$F(1, 195) = 1.040$
Own preference (RC)	24	90	0.329	4.985^{***}			
Selection by researcher/therapist	37	107	0.415	7.580^{***}	0.087	1.020	
Music induction	61	197					$F(1, 195) = 1.065$
Prerecorded music (RC)	54	184	0.367	8.287^{***}			
Live music	7	13	0.525	3.571^{***}	0.159	1.032	
Music style	58	186					$F(1, 184) = 1.336$
Relaxation (RC)	46	152	0.393	8.689^{***}			
Own choice	12	34	0.285	3.417^{***}	−0.109	−1.156	
Music with lyrics	61	197					$F(2, 194) = 0.560$
No (RC)	47	143	0.372	7.488^{***}			
Yes	2	6	0.189	0.789	−0.183	−0.747	
Both	12	48	0.446	4.604^{***}	0.074	0.678	
Music tempo	61	197					$F(1, 195) = 0.304$
60–80 beats p/m (RC)	36	130	0.361	6.449^{***}			
No specific tempo	25	67	0.409	6.029^{***}	0.049	0.551	
Frequency	60	193					$F(1, 195) = 0.091$
One session (RC)	49	146	0.375	8.003^{***}	0.027	0.301	
More sessions	11	47	0.402	4.845^{***}			
Duration (continuous)	49	161	0.318	8.215^{***}	0.003	1.479	$F(1, 159) = 2.187$
Sample characteristics							
Proportion of males (continuous)	55	181	0.366	7.871^{***}	−0.094	−0.549	$F(1, 179) = 0.301$
Age (continuous)	55	182	0.386	8.228^{***}	−0.001	−0.223	$F(1, 180) = 0.050$

Note. *s* = number of independent studies; *k* = number of effect sizes; β_0 = intercept/mean effect size (d); t_0 = difference in mean *r* with zero; β_1 = estimated regression coefficient; t_1 = difference in mean r with reference category; *F*(df1, df2) = omnibus test; (RC) = reference category. * $p < .05$, ** $p < .01$,*** $p < .001$

Figure 2
Trim-and-Fill Plot for the Effects of Music Interventions on Physiological Stress-Related Outcomes.



intervention), music induction (prerecorded music or live music), music style (predetermined relaxing music or patient preferred music), music tempo (60-80 bpm or another tempo), or the use of music with lyrics in contrast to the use of purely instrumental music.

3.2 Effect of Music Interventions on Psychological Stress-Related Outcomes

The meta-analysis on the effect of music interventions on physiological stress-related outcomes contained 79 independent studies (*s*), from which 130 effect sizes (*k*) were denied, and a total sample of *N* = 6.800 participants, of which *n* = 3.373 participants in the music intervention groups and *n* = 3.427 participants in the comparison groups.

3.2.1 Overall effect on psychological stress-related outcomes

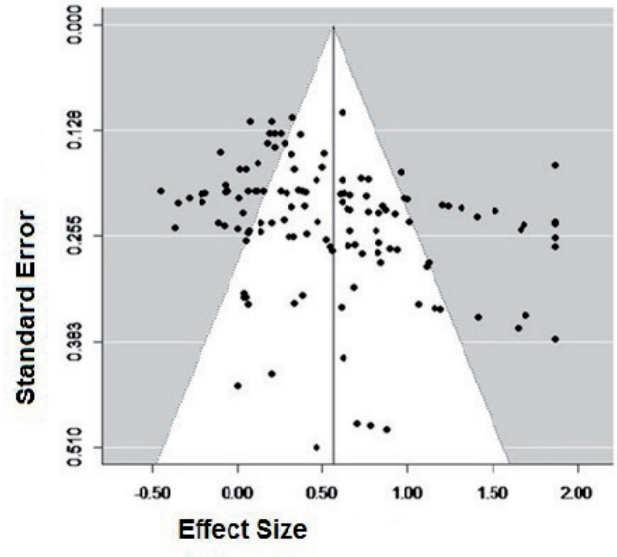
A significant medium-to-large effect (*d* = 0.545, *p* = ≤ .001) of music interventions on psychological stress-related outcomes was found, indicating that music interventions reduced psychological stress-related symptoms (state anxiety, nervousness, restlessness, and feelings of worry). The trim-and-fill plot did not show lack of effect sizes on the left side of the funnel (see Figure 3), therefore publication bias was unlikely. The likelihood ratio test showed that significant variance was present at the between (level 3) and within (level 2) study level. Moderator analyses on type of outcome, study, sample, and music intervention characteristics were conducted to examine the effect of music intervention on psychological stress-related outcomes.

Table 4
Moderator Effects of Music Interventions on Psychological Stress-Related Outcomes

Moderator variables	*s*	*Kk k*	β_0 (mean *d*)	t_0	β_1	t_1	*F*(df1, df2)
Outcome characteristics	79	130					*F*(1, 128) = 0.118
State anxiety (RC)	70	101	0.553	9.008***			
Stress	19	29	0.512	4.592***	−0.041	−0.344	
Study characteristics							
Setting	79	130					*F*(2, 127) = 2.353
Surgery (RC)	23	51	0.669	6.808***			
Nonmedical	12	19	0.694	4.553***	0.025	0.137	
Polyclinic procedures	44	60	0.433	5.707***	−0.236	−1.897	
Continent	79	130					*F*(1, 128) = 0.765
Western (RC)	40	70	0.496	6.206***			
Non–Western	39	60	0.596	7.318***	0.100	0.874	
Study quality	79	130					*F*(2, 127) = 1.573
Strong (RC)	42	71	0.628	8.175***			
Moderate	21	28	0.387	3.423***	−0.240	−1.757	
Weak	16	31	0.518	4.142***	−0.110	−0.749	
Intervention characteristics							
Type of music intervention	79	130					*F*(1, 128) = 0.013
Music intervention (RC)	67	110	0.548	8.838***			
Music therapy	12	20	0.529	3.539***	−0.019	−0.115	
Type of control condition	79	130					*F*(1, 128) = 2.795
Other intervention (RC)	23	35	0.592	9.252***			
Care as usual	63	95	0.402	3.900***	−0.190	−1.672	
Music selection	79	130					*F*(1, 128) = 0.953
Own preference (RC)	30	62	0.611	6.911***			
Selection by researcher/therapist	49	68	0.498	6.709***	−0.113	−0.976	
Music induction	79	130					*F*(1, 128) = 0.385
Prerecorded music (RC)	66	109	0.560	8.991***			
Live music	13	21	0.464	3.269**	−0.096	−0.621	
Music style	71	120					*F*(1, 118) = 0.512
Own preference (RC)	34	47	0.521	5.655***			
Relaxation	37	73	0.609	7.296***	0.089	0.716	
Music with lyrics	79	130					*F*(2, 127) = 0.872
No (RC)	52	81	0.556	7.891***			
Yes	5	8	0.264	1.186	−0.292	−1.257	
Both	22	41	0.581	5.471***	0.026	0.201	
Music tempo	79	130					*F*(1, 128) = 3.132
60–80 beats p/m (RC)	49	82	0.625	8.789***			
No specific tempo	30	48	0.413	4.578***	−0.212	−1.871	
Frequency intervention	79	130	0.601	9.316***	−0.222	−1.776	*F*(1, 128) = 3.156
One session (RC)	61	90	0.379	3.487***			
More than one session	18	40					
Duration (continuous)	65	109	0.547	8.662***	0.001	0.547	*F*(1, 107) = 0.299
Sample characteristics							
Proportion of males (continuous)	78	129	0.549	9.480***	0.017	0.061	*F*(1, 127) = 0.004
Age (continuous)	73	123	0.537	9.023***	0.003	0.629	*F*(1, 121) = 0.395

Note. *s* = number of independent studies; *k* = number of effect sizes; β_0= intercept/mean effect size (*d*); t_0 = difference in mean *d* with zero; β = estimated regression coefficient; t_1 = difference in mean *d* with reference category; $F(df_1, df_2)$ = omnibus test; (RC) = reference category. * $p < .05$, ** $p < .01$,*** $p < .001$

Figure 3
Trim-and-Fill Plot for the Effects of Music Interventions on Psychological Stress-Related Outcomes.



3.2.2 Results of moderator analyses on psychological stress-related outcomes

The moderator analyses of the psychological stress-related outcomes were justified by significant overall heterogeneity for all moderator variables, including the variables with missing values. The results of these moderator analyses are presented in Table 4.

Outcome characteristic. The type of psychological outcome (stress- or state anxiety) did not moderate the effect of music interventions on psychological stress-related outcomes.

Study characteristics. No significant moderating effects were found in the study characteristics. More specifically, the continent where the study was conducted, type of setting, and quality of the study, did not moderate the effect of the music intervention on physiological stress-related outcomes.

Sample characteristics. No moderating effects of the age and gender composition of the samples were found.

Intervention characteristics. We found no significant moderating effects of the intervention characteristics, which included the type of intervention (music therapy or music intervention), music induction (prerecorded music or live music), music style (predetermined relaxing music or self-selected music by the patient), and the use of music with lyrics in contrast to the use of purely instrumental music. The music tempo showed a trend (p = .064), indicating that music with 60–80 bpm yielded larger effects than music with another or unspecified tempo. Another trend concerned the type of control condition (p = .097). If a non-established intervention aimed at stress reduction was used as a control condition, effects were somewhat larger than if the control condition concerned CAU (i.e., regular medical

care). Finally, the number of interventions showed a trend (p = .078) indicating that one single session generated larger effects than two or more sessions.

4. Discussion

4.1 Overall Effects

By conducting two separate multilevel meta-analyses, the current study aimed to assess the strength of the effect of music interventions on both physiological and psychological stress-related outcomes. Furthermore, the study aimed to examine which outcome, study, sample or intervention characteristics moderated the strength of the effect on physiological and psychological stress-related outcomes. Overall, we found a significant small-to-medium effect of music interventions on physiological stress-related outcomes (d = .380) and a medium effect of music interventions on psychological stress-related outcomes (d = .545), indicating that music intervention groups benefited more than the comparison groups. We conclude that music interventions are effective in reducing physiological and psychological stress-related symptoms in different kinds of settings (mental healthcare, polyclinic medical settings, during medical surgery and in daily life situations). There were no indications of publication bias.

The overall findings of the current study are consistent with findings of previous meta-analyses (Bradt & Dileo, 2014; Bradt, Dileo, & Potvin, 2013; Bradt, Dileo, & Shim, 2013; Bradt et al., 2011; Gillen et al., 2008; Kim et al., 2015; Pelletier, 2004; Rudin et al., 2007). There is a growing body of evidence that music interventions yield positive, moderate effects on stress reduction. Considering the demands of today's society, the need for stress reduction interventions are large. Millions of people around the world use tranquilizing medications, such as tricyclic antidepressants and benzodiazepines, to cope with life stressors or anxiety (e.g., Bandelow et al., 2015; Olfson et al., 2015; Puetz et al., 2015). Not only do these types of medication have considerable negative side effects, including substance dependence and abuse, research also indicates that the effects of pharmacological treatment on stress-related problems are not much larger than the effects of music interventions found in the current meta-analytic study (Olfson et al., 2015). Besides, a common argument for starting pharmacological treatment instead of psychological treatment for stress reduction is that its effects occur immediately or faster (Bandelow et al., 2015; Fedoroff & Taylor, 2001). However, the results of this meta-analysis were based on mainly short-term music interventions, most of the time on a single occasion, having a direct stress reducing effect in various contexts. The current study therefore indicates the relevance of brief music interventions for stress reduction in all kinds of settings. Also, our search revealed that the effects of long-term music interventions have been seldom examined.

4.2 Psychological Effects of Music

Results of our meta-analysis show a medium effect of music interventions on psychological stress-related outcomes, including emotional states of subjective worry, state anxiety, restlessness and nervousness. Music not only reduces *physiological arousal*, but also affects *emotional states.* This may be attributed to the effect of music on brain areas, such as the amygdala, which are responsible for emotional processes. A related explanation for the positive effects of music interventions on psychological stress-related outcomes concerns the positive influence of listening to pleasant music on *emotional valence*, which can be explained by the degree of attraction that an individual feels towards a specific object or event (Jäncke, 2008; Juslin & Västfjäll, 2008). Music experienced as pleasant increases the intensity of emotional valence (the felt happiness), which has a stress-reducing effect (Jiang et al., 2016; Rohner & Miller, 1980; Sandstrom & Russo, 2010; Witvliet & Vrana, 2007). An increased dopamine activity in the mesolimbic reward brain system has been shown to be associated with these feelings of happiness in response to high-valence music (e.g. Blood & Zatorre, 2001; Salimpoor et al., 2013; Salimpoor et al., 2011; Zatorre, 2015).

Another explanation for the positive effect of music interventions on psychological stress-related outcomes may be that listening to music can provide 'distraction' from stress-increasing thoughts or feelings (Bernatzky et al., 2011; Chanda & Levitin, 2013). Indeed, the beneficial property of music to distract people from aversive states has been supported by short-term music interventions for acute stress reduction (Fancourt et al., 2014; Linnemann et al., 2015). These findings are therefore in line with the current meta-analysis, which primarily included studies involving short-term music interventions.

Music listening in the presence of others may strengthen the stress-reducing effect of the music intervention, which is believed to be caused by increased emotional well-being (Juslin et al., 2008), and increased feelings of social cohesion (Boer & Abubakar, 2014; Linnemann et al., 2016; Pearce et al., 2015). There is also empirical evidence showing that people synchronize in movement (auditory-motor synchronization) with each other when engaging in music therapy group interventions, which evokes positive feelings of togetherness and bonding, and decreases stress levels (Linnemann et al., 2016; Tarr et al., 2014). Group music-making or singing together may result in social bonding, which may be explained by the release of the neurotransmitters *endorphin* and *oxytocin* (e.g., Dunbar et al., 2012; Freeman, 2000; Tarr et al., 2014; Weinstein et al., 2016). These neurotransmitters play a role in the defensive response to stress (Amir et al., 1980; Dief et al., 2018; Myint et al., 2017). It should be noted that nearly all studies included in our meta-analysis examined participants listening to music alone (e.g., by using headphones), because most studies examined 'music medicine' instead of music therapy, which is applied by a music therapist, and can be delivered both in a group and individual format.

4.3 Effect Moderating Variables

Results indicate that moderating variables may explain differences in the strength of the effect sizes. Significant stronger moderating effects were found in studies in which physiological arousal, as a result of stress, was measured by heart rate, compared to studies in which physiological arousal was measured by blood pressure or stress-related hormone levels. These results are consistent with the large body of knowledge concerning the immediate effects of psychological stress on the sympathetic responses in the autonomic nervous system, meaning increases of heart rate and decreases of heart rate variability (Chandola et al., 2010; Föhr et al., 2016). The results are also in line with the assumption that music with a slow steady rhythm provides stress reduction by altering inherent body rhythms, such as heart rate (Thaut & Hoemberg, 2014; Thaut et al., 1999).

None of the other possible moderators of interventions effects proved to be significant, but results showed some noteworthy trends of the intervention characteristics. First, the *music tempo* seemed to influence the strength of the effect of music interventions on the psychological stress-related outcomes. Larger effect sizes were found in music with a tempo of 60–80 bpm, where tempo represents slow and soothing music. This corresponds with previous research, which suggests that music with a slow tempo can be considered to be one of the most significant determinants of audio-related effects on stress reduction (Bernardi et al., 2006; Björkman et al., 2013; Iwanaga et al., 2005; Jiang et al., 2016; Nilsson, 2008). Second, the frequency of the music intervention sessions did moderate the effect of music interventions on the psychological stress-related outcomes, indicating that only one session is needed for achieving effects on stress reduction. This is in line with previous research, which implies that music has an immediate positive effect on stress reduction (e.g., Koelsch, 2015; Zatorre, 2015). However, Leubner and Hinterberger's (2017) review of the effectiveness of music interventions on depression showed that more than one session yielded larger effects (at least, within the first 6 weeks of treatment).

Contrary to our expectations, we did not find a significant moderating effect of some intervention characteristics. Firstly, type of intervention did not moderate the effects on physiological stress-related outcomes. Studies in which *music therapy* is offered (by a trained therapist) did not yield significantly larger effect sizes than studies in which music interventions were offered as so-called purposeful *music activities* (by the researcher, healthcare professionals or self-administered by the patient). However, Dileo (2006) stated that music therapy is more effective than 'music medicine' interventions and attributed this difference to the fact that music therapists individualize their interventions to meet patients' specific needs (Bradt & Dileo, 2014; Dileo, 1999, 2006). A possible explanation for different findings of Dileo's review (2006) compared to the current meta-analytic review is that, in the past 14 years, the number of RCTs examining the effects of 'music medicine' interventions has grown substantially, and with that, both the associated research methods and intervention protocols were improved. However, experimental research on the effects of *music therapy* is still in its infancy, while considerable diversity is evident in the interventions that have

been examined so far. Secondly, results show that the way the music was selected did not influence the effect of music interventions on stress-related outcomes, while many previous studies report that self-selected music is the most effective in terms of stress reduction (e.g. Brannon & Fiest, 2007; Jiang et al., 2013; Jiang et al., 2016; Juslin et al., 2008). A possible explanation is that the term 'self-selected music' is used differently in studies included in this meta-analytic review. The term 'self-selected music' was both used when the patient brings their own favorite music *and* when the patient could choose the music from a pre-selected list of musical styles (e.g. Bradt & Dileo, 2014; Cepeda et al., 2006; Helsing et al., 2016; Lee et al., 2005; Nilsson, 2008). Notably, where self-selected music also means that patients have to choose their music from a preselected list, the researcher can preselect only music with specific characteristics (nonlyrical music with a tempo of 60–80 bpm and a sound intensity level of 60 dB) which contributes to a stronger effect on physiological stress-related outcomes (Bernardi et al., 2006; Björkman et al., 2013; Dileo, 2007; Gan et al., 2016; Good et al., 2000). Besides, previous research shows that listening to soothing music, pre-selected by the researcher, lowers the stress-levels significantly in contrast to energetic up-tempo music (Iwanaga et al., 2005; Jiang et al., 2013; Sandstrom & Russo, 2010). This differs from the findings of many studies, which attribute these positive effects to the ability of participants to self-select music (Juslin et al., 2008). However, this often means that the participants can choose one of the researcher's pre-selected playlists instead of actually bringing their own music. Because of the uncertainty about what the effect actually causes, in the present study we coded 'own preference' when patients could bring their own music, and 'selection by researcher/therapist' when patients had to choose the music that was pre-selected by the researcher or therapist.

Results showed no moderating effect for the settings in which the music intervention was conducted (nonmedical, surgery, polyclinic procedures), which might indicate that the effects of music interventions do not depend on the type of setting. This is in line with the assumption that stress is a general activation response to any stimulus that could mean both a threat and a challenge, resulting in heightened arousal of the autonomic nervous system (Pfaff et al., 2007; Selye, 1976, 1973). Also, neurological evidence provides insight in to the positive effects of music on the stress response with respect to arousal regulation, and these effects appear to be independent of context or setting (e.g. Casey, 2017; Koelsch, 2015; Koelsch et al., 2016; Linnemann et al., 2015; Thaut & Hoemberg, 2014).

4.4 Limitations of the Present Study

The current study has some limitations that need to be mentioned. Firstly, we operationalized the concept of stress in terms of physiological and psychological outcomes, which resulted in two meta-analyses. Because the moderator analyses were performed on both psychological and physiological data, the impression can be that it concerns two independent outcomes, while the outcomes always affect and strengthen each other during stress. This is because the underlying systems of the physiological and psychological responses are both, and mostly at

the same time, responsible for the experience of stress (e.g., Linnemann et al., 2017; McEwen & Gianaros, 2010). Secondly, some categories of relevant variables included in the moderator analyses contained only a few effect sizes, which reduces statistical power and sets limits to the generalizability of the study findings. This especially may be the case for the moderators *music induction* (prerecorded music vs. live music) and *music style* (relaxation vs. own choice) in the meta-analysis of the physiological stress-related outcomes and the moderators *music tempo* (60–80 bpm vs. another tempo), and the *frequency* of the intervention (a single session vs. more than one session) in the meta-analysis of the psychological stress-related outcomes.

4.5 Implications for Research and Practice

Despite the limitations, this study has important implications for future research and the practical use of music interventions in stress reduction. First and foremost, this meta-analytic study indicates that music interventions can be effective in the reduction of stress. Many people suffer from stress-related symptoms, both in their daily lives and in specific settings (e.g., medical settings, mental health care settings, work-related settings). Considering the fact that music interventions are very easy and inexpensive to integrate in both daily lives and in medical settings, it is important to recognize the effects of music interventions. Another implication is that future research should focus on the specific characteristics of the music intervention on stress reduction, for example, the music tempo, the style of the music, the use of live music or prerecorded music, the way the music is selected, or the frequency of the music intervention sessions.

The vast body of neurological evidence regarding the influence of music on arousal, stress and emotional processes is still growing, but the specific practical implications have not yet been sufficiently investigated. In included RCTs, music interventions are still considered like one and the same, while the specific characteristics of music show different effects, such as the music tempo (60–80 bpm). To identify the most effective music intervention for stress reduction, it is important that the possible moderators of the music interventions be better tested. Therefore, it is recommended for future trials to describe all aspects of the music intervention, with both the specific characteristics involved in these two meta-analyses as well as the musical characteristics, such as timbre, melodic and harmonic aspects, and rhythmic accentuation. These characteristics were not reported in the included RCTs, but can nonetheless moderate the effects of music interventions on stress reduction (e.g. Leubner & Hinterberger, 2017; Moore, 2013; Thaut & Hoemberg, 2014). There are many RCTs examining the effect of *music medicine interventions*, but there is definitely a lack of RCTs examining the effect of *music therapy interventions* on stress-related outcomes, and it is therefore impossible to establish at this time whether these interventions are more effective than the 'music medicine' interventions. Although the use of prerecorded music may be preferred by researchers as a standardized stimulus, it is recommended to also develop specific *music therapy* protocols that will adhere to the research standards of RCTs.

The relationship between the frequency/duration of the music intervention and the effects on stress reduction is unclear. Future research should examine this topic, because apart from the musical characteristics, such as the music style and music tempo, these general characteristics of the music intervention may also moderate the effects on stress reduction. It seems that the influence of the music selection method on the effects of the music intervention is interpreted differently. In new trials, we strongly recommend reporting clearly if the researcher/therapist did choose the music of the intervention, or if the participants had to choose one of the researchers' or therapists' pre-selected music playlists, or if the participants brought their own music. Finally, it is recommended to compare the effects of music (therapy) interventions on stress-related outcomes with pharmacological treatment, but also with other experiential interventions, such as yoga or mindfulness.

4.6 Summary

The current meta-analytic review provides high-level evidence that music interventions can be effective in reducing stress and provides justification for the increasing use of music interventions for stress reduction in both medical and mental health care practice. Considering the low costs and lack of side effects of music interventions, the moderate tranquilizing effects of music are very significant for the prevention and treatment of stress-related problems. However, the development of music (therapy) intervention protocols are necessary to set up more robust research into the effects of music interventions, and to gain more insight into the effect moderating characteristics of music intervention for stress reduction.

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CHAPTER 3
3

Music Therapy for Stress Reduction: A Systematic Review and Meta-Analysis

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Abstract

Music therapy is increasingly being used as an intervention for stress reduction in both medical and mental healthcare settings. Music therapy is characterized by personally tailored music interventions initiated by a trained and qualified music therapist, which distinguishes music therapy from other music interventions, such as 'music medicine', which concerns mainly music listening interventions offered by healthcare professionals. To summarize the growing body of empirical research on music therapy, a multilevel meta-analysis, containing 47 studies, 76 effect sizes and 2.747 participants, was performed to assess the strength of the effects of music therapy on both physiological and psychological stress-related outcomes, and to test potential moderators of the intervention effects. Results showed that music therapy showed an overall medium-to-large effect on stress-related outcomes (*d* = .723, [.51-.94]). Larger effects were found for clinical controlled trials (CCT) compared to randomized controlled trials (RCT), waiting list controls instead of care as usual (CAU) or other stress-reducing interventions, and for studies conducted in Non-Western countries compared to Western countries. Implications for both music therapy and future research are discussed.

Keywords: music therapy, arousal, stress, state anxiety, music intervention, multilevel meta-analysis

1. Introduction

Stress is a well-known risk factor for the onset and progression of a range of physical and emotional problems, such as cardiovascular diseases, cancers, anxiety disorders, depression, and burnout (American Psychological Association [APA], 2017; Australian Psychological Society [APS], 2015; Steptoe & Kivimäki, 2012). To cope with stress and the demands of today's society, millions of people over the world use tranquilizing medications, which have a lot of negative contraindications and side effects, including substance dependence and abuse (Bandelow et al., 2015; Olfson et al., 2015; Puetz et al., 2015; World Health Organization [WHO], 2010). Therefore, it is important to examine the effects of non-pharmacological therapeutic interventions for the prevention and management of stress (De Witte, Spruit, et al., 2020; Kamioka et al., 2014; Martin et al., 2018; Raglio et al., 2015).

For decades and all over the world music has been used to provide calmness and relaxation. These stress reducing qualities are the most widely studied effects of music (Chanda & Levitin, 2013; de Witte, Spruit, et al., 2020; Juslin & Västfjäll, 2008; Koelsch, 2015; Mehr et al., 2019). Therefore, music therapy interventions are increasingly being used to reduce stress and enhance the well-being of clients across a variety of clinical populations (Agres et al., 2021; Bainbridge et al., 2021; Juslin & Västfjäll, 2008; Kemper & Danhauer, 2005; Koelsch, 2012, 2015; Landis-Shack et al., 2017; Thaut & Hoemberg, 2014).

Music therapy is specifically characterized by using the specific qualities of music in a therapeutic relationship with a music therapist. This distinguishes music therapy from other music interventions, mostly offered by medical or healthcare professionals and referred to as *music medicine* (Agres et al., 2021; Bradt, Dileo, & Shim, 2013; Gold et al., 2011; Magee, 2019; de Witte, Spruit, et al., 2020). The body of research on music therapy is much smaller compared to the amount of research on music listening interventions. Our previous meta-analytic review (de Witte, Spruit, et al., 2020) was focused on the effects of music interventions in general and included mainly music listening interventions. In none of these studies a trained music therapist was involved.

In order to integrate the available knowledge on the effects of music therapy on stress, we conducted a systematic review and meta-analysis of quantitative studies testing the effects of music therapy on both physiological and psychological stress-related outcomes in mental and medical healthcare settings.

1.1 The Influence of Music on the Stress Response

Stress can be regarded as the quality of an experience, produced through a person-environment transaction that may result in physiological or psychological distress (Aldwin, 2007). Responses to stress can be related to both increased physiological arousal and specific emotional states, while the underlying systems of those responses regulate and affect each other during stress (e.g., de Witte, Spruit, et al., 2020; Linnemann et al., 2017; McEwen & Gianaros, 2010). The stress reducing effect of music therapy interventions is explained by

music itself as well as the continuous attunement of music by the music therapist to the individual needs of a patient.

Music listening is strongly associated with stress reduction by the decrease of physiological arousal as indicated by reduced cortisol levels, lowered heart rate, and decreases in mean arterial pressure (e.g. Burrai et al., 2016; Koelsch et al., 2016; Kreutz et al., 2012; Linnemann et al., 2015). Music can also reduce negative emotions and feelings, such as subjective worry, state anxiety, restlessness or nervousness (Akin & Iskender, 2011; Cohen et al., 1983; Pittman & Kridli, 2011; Pritchard, 2009), and increase positive emotions and feelings, such as happiness (Jäncke, 2008; Juslin & Västfjäll, 2008). This is in line with studies showing that music modulates activity in brain structures, such as the amygdala and the mesolimbic reward brain system, which are known to be involved in emotional and motivational processes (Blood & Zatorre, 2001; Koelsch et al., 2016, 2016; Levitin, 2009; Moore, 2013; Salimpoor et al., 2013; Zatorre, 2015). Furthermore, it is assumed that the systematic application of music in therapy in response to the needs of the patient(s) can strengthen the impact of music (Agres et al., 2021; Bradt & Dileo, 2014). In addition, empirical evidence shows that music activities in a group may result in synchronization among group members, which leads to positive feelings of togetherness and bonding (Linnemann et al., 2016; Tarr et al., 2014). These feelings of togetherness and bonding may be explained by the release of the neurotransmitters endorphin and oxytocin, which both play an important role in the defensive response to stress (e.g., Amir et al., 1980; Dief et al., 2018; Myint et al., 2017). Lastly, music listening can help to lower stress levels through its quality to provide 'distraction' from stress-increasing feelings or thoughts (Bernatzky et al., 2011; Chanda & Levitin, 2013).

1.2 Music Therapy

Music therapy can be defined as the clinical and evidence-informed use of music interventions to accomplish individualized goals within a therapeutic relationship in order to achieve physical, emotional, mental, social and cognitive needs (Aalbers et al., 2019; Agres, et al., 2021; American Music Therapy Association [AMTA], 2018; de Witte, Spruit, et al., 2020). Music therapy has been applied as a therapeutic intervention in a wide spectrum of health care contexts, such as mental health care, forensic care, nursing homes, rehabilitation, and oncology (e.g. Agres et al., 2021; Martin et al., 2018; Kamioka et al., 2014). Although the term 'music therapy' sometimes refers to any kind of use of music as an intervention in health care settings, music therapy should be offered by a trained music therapist, who is a licensed and qualified therapist with the required knowledge in psychology, medicine, and music (Agres et al., 2021; AMTA, 2018; Bradt et al., 2015; Magee, 2019).

Music therapists use the unique qualities of music (e.g., melody, rhythm, tempo, dynamics, pitch) within the therapeutic relationship to access patient's emotions and memories, to address social experiences or influence behavior (Bruscia, 1987; Wheeler, 2015). This specific kind of responsivity to the patient's needs can be regarded as the key competencies of the music therapist, referring to the processes that take place between

therapist and patient supporting coordination, empathy and shared perspectives (Agres et al., 2020). More specifically, during music therapy the music therapist attunes to the patient by adjusting the music created as an immediate response to the patient's needs (Aalbers et al., 2019; Magee, 2019).

To work on patient-therapist attunement, the music therapist synchronizes with the patient moment-by-moment, which may be considered as a mirroring technique. This means that the (musical) actions of the music therapist and the patient can become simultaneous and regulated through time, yielding a similar expression in movement, matching pulse, rhythm, dynamics and/or melody (Aalbers et al., 2019; Bruscia, 1987; Schumacher & Calvet, 2008). For example, the music therapist may influence patients' perceived stress during musical improvisation by synchronizing with the patient's music-making, subsequently changing the musical expression by playing slower and less loudly. This specific way of patient-therapist attunement is commonly used in music therapy practice and refers to the so-called Iso Principle (e.g., Altshuler, 1948; Heiderscheit & Madson, 2015). Literature shows that the tempo and loudness are important for the experienced intensity of the music (Gabrielsson & Lindström, 2010), and music with a slow steady rhythm may provide stress reduction by altering inherent body rhythms, such as heart rate (Thaut & Hoemberg, 2014; Thaut et al., 1999). The music therapist uses several types of interventions, which can be offered to a group of patients as well as individually. The number, frequency, and duration of the music therapy session may vary widely, and depend on the targeted outcome, patients' preferences, and/or the setting in which the music therapy is offered (Agres et al., 2021; AMTA, 2018). Furthermore, music therapy interventions can be subdivided in two broad categories: *active* and *receptive* interventions (Magee, 2019; Magee et al., 2017; Wheeler, 2015).

Active interventions involve the patient doing something with the music during the music therapy sessions, such as musical improvisation, composing music or songs, movement to music, or singing or vocalizing. According to both literature and clinical practice, it seems that musical improvisation is the most used intervention within music therapy, meaning that patient(s) and therapist improvise on musical instruments they have chosen and play together freely or with a given structure (Gold et al., 2009; Wigram, 2004). In receptive music therapy interventions, the patient is not actively making music, but rather responds to music provided by the music therapist, such as listening to live or prerecorded music (Bruscia, 1998; Magee, 2019; Wheeler, 2015). The patient listens to the music and may process verbally their own emotions and/or experiences. During both active and receptive music interventions music therapists make specific use of the unique qualities of music (also known as 'musical components'), such as rhythm, pitch, tempo, dynamics, melody and harmony, to facilitate and promote personal contact, communication, learning, mobilization, expression and other relevant goals (Agres et al., 2021; Câmara et al., 2013; Taets et al., 2019; Thaut & Hoemberg, 2014; Wheeler, 2015).

Summarized, whereas *music medicine* does not involve a personal therapeutic process, *music therapy* requires such a process, characterized by personally tailored music

interventions initiated by a trained/qualified music therapist (de Witte, Spruit, et al., 2020; Leubner & Hinterberger, 2017). These music therapy interventions can be divided in receptive music therapy interventions (music listening) or active music therapy interventions (live music-making), and are specifically characterized by musical attunement, facilitated by the music therapist, which distinguishes music therapy from other music interventions.

1.3 Music Therapy versus Music Medicine

Research on music therapy is fast-growing (de Witte, Spruit, et al., 2020). The effects of music listening interventions, such as 'music medicine', are mainly caused by the general influence of music on the stress response, whereas the effects of music therapy may also be explained by the therapeutic relationship through patient-therapist attunement by the use of music. Dileo (2006) stated that music therapy is more effective than 'music medicine' interventions, and attributed this difference to the fact that music therapists individualize their interventions to meet patients' specific needs (Bradt et al., 2010; Dileo, 1999, 2006).

In a Cochrane review of Bradt et al. (2016) it was shown that 'music medicine' interventions and music therapy were equally effective in decreasing (state) anxiety. Bradt et al. (2016) compared 'music medicine' with music therapy, and also found that both types of interventions were equally effective for anxiety and stress reduction, although 77.4% of the participants expressed a preference for music therapy for future treatments. This patient's preference for music therapy was related to quality of therapeutic relationships, interactive music making and the possibility of emotional expression, which is precisely what music therapy distinguishes from music listening interventions (Bradt et al., 2016; Gutgsell et al., 2013).

In our previous meta-analytic review on Randomized Controlled Trials (RCTs) examining the effects of music interventions on stress-related outcomes (de Witte, Spruit, et al., 2020), we showed that music therapy did have at least as much effect on physiological stress-related outcomes (d = .423) as 'music medicine' (d = .379). However, only 7 studies on music therapy were included against 54 studies examining music medicine. Therefore, findings on music therapy were compromised by low generalizability and lack of statistical power to examine factors that might affect the effectiveness of music therapy by means of moderator analyses. Notably, most effectiveness studies on music therapy are quasi-experimental, because it is often difficult to meet the requirements for randomization and/or masking procedures (Bradt, Dileo, & Shim, 2013, de Witte, Spruit, et al., 2020; Magee et al., 2017).

1.4 The Present Study

The present study is a systematic review and meta-analysis on the effects of music therapy on both physiological stress-related arousal (e.g., blood pressure, heart rate, hormone levels) and psychological stress-related experiences (e.g., state anxiety, restlessness or nervousness) in clinical health care settings. In our previous meta-analysis, we examined the effect of music interventions on stress-related outcomes. The included studies primarily used prerecorded

music offered by medical professionals, whereas music therapy involves a trained music therapist who is responsive to the needs of the patient and can influence emotions and/or behavior of the patient by the use of music.

In the present meta-analysis, we included both RCTs and quasi-experimental designs with a control condition (Clinical Controlled Trials [CCT]), accounting for the effect of study design and quality in moderator analyses. The inclusion of quasi-experimental studies, which have been conducted under clinically representative conditions, increases external validity of meta-analytic findings and substantially increases statistical power of a meta-analysis (Shadish et al., 2002, 2008).

The methodology of the present meta-analytic study is in line with our recent three-level meta-analysis (de Witte, Spruit, et al., 2020), in which 104 randomized controlled trials were included. Results showed a significant small-to-medium effect of music interventions on physiological stress-related outcomes (d = .380; 61 trials), and a medium effect of music interventions on psychological stress-related outcomes (d = .545; 79 trials), indicating that groups receiving music intervention benefited more than the comparison groups. In the present meta-analysis, we examine the overall effect of music therapy on stress reduction, accounting for differences in physiological and psychological stress-related outcomes, and we aim to gain more insight into study, sample, outcome and intervention characteristics that might moderate the effects of music therapy on stress reduction.

2. Methods

2.1 Inclusion Criteria

For the current meta-analysis, multiple inclusion criteria were formulated. First, only Randomized Controlled Trials (RCTs) and Clinical Controlled Trials (CCTs) that examined the effect of music therapy on the experience of stress and/or state anxiety were included. The type of intervention concerned important inclusion criteria for this meta-analysis. Only studies that offered music therapy by an educated and certified music therapist were included in this meta-analysis. Outcome measures related to quality of life (QoL) or pain were excluded, because in this study only the primary outcome measures of stress were included. The physiological effects of stress had to be measured by heart rate (HR), heart rate variability (HRV), blood pressure and hormone levels. The psychological effects of stress had to be measured by self-report instruments aiming at '*stress*' or '*state anxiety*'. Second, studies examining people with dementia or participants younger than 18 years of age were excluded. Although many studies showed cognitive and emotional benefits in dementia patients when they sing or listen to familiar songs (Särkämö et al., 2008; 2014), these findings are not directly related to 'stress reduction'. In addition, the stress measurement instruments which are used in the included studies are not used in studies examining people with dementia or young participants.

2.2 Selection of the Studies

All randomized controlled trials (RCTs) and clinical controlled trials (CCTs) available until the 8th of May 2019 that met the inclusion criteria were included in this meta-analytic review. Multiple systematic searches were performed with the help of an independent medical librarian, as librarian engagement is significantly associated with higher quality of reported search strategies (Rethlefse et al., 2015). We conducted a computer-based search of the psychological and medical electronic literature databases, including Medline, Academic Search Complete, Cochrane Library, Web of Science, Embase, Wiley Online Library, Springerlink, PubMed, PiCarta, Academic Search Premier, ScienceDirect, PsycINfo and Google Scholar. The search string comprised three elements: a *music therapy* element, a *stress-related outcome* element and a *study design* element. For the music therapy element, the following keywords were used: 'music therapy', 'musical therapy' or 'music-based therapy'. For the stress-related outcome element, the following keywords were used: 'stress', 'anxiety', 'arousal', 'psychological stress', 'occupational stress', 'physiological stress', 'mental suffer', 'anguish', 'hypertension', 'relaxation', 'heart rate', 'blood pressure', 'nervousness', 'cortical vigilance', 'distress', 'cortisol', 'intravascular pressure', 'vascular pressure' or 'STAI'. Concerning the study design element, the keywords: 'randomized controlled trial', 'randomised controlled trial', 'clinical controlled trial', 'randomised', 'randomized', 'RCT', 'review' or 'meta-analysis' were used. Furthermore, reference sections of review- and meta-analytic articles about the effect of music (therapy) interventions on stress-related outcomes were inspected for qualifying studies. The search protocol of this meta-analytic review is registered at the international prospective register of systematic reviews (ref.no. CRD42020160222).

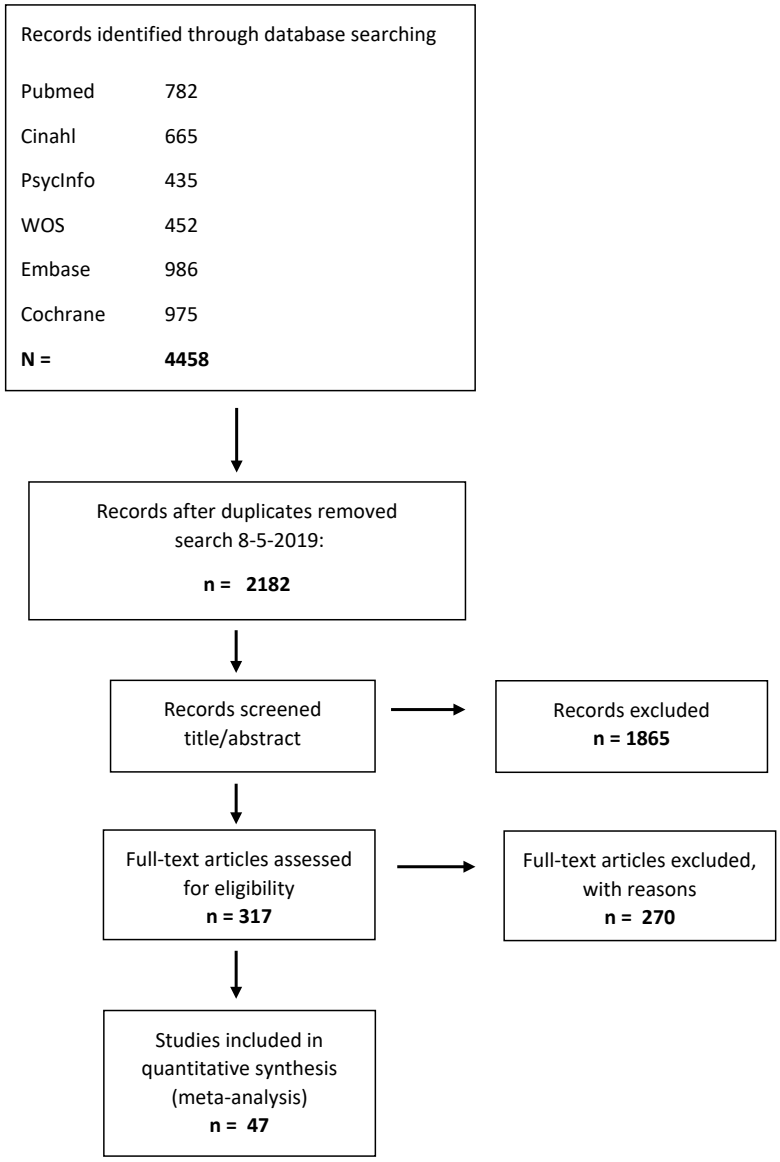
The initial search resulted in 2.180 individual studies and was conducted by an independent librarian and the first author. The first selection on *title* and *abstract* resulted in 317 individual studies that were, posteriorly, *full text* screened based on the inclusion criteria. Finally, 47 studies met all the inclusion criteria (see Figure 1). All the steps of the selection process were conducted by two different authors, who selected the studies blindly from each other. Concerning the selection conflicts, a third author was involved and made the final decision. Table A2 provides an overview of the included studies and their main characteristics (see Appendix 2, page 253).

2.3 Coding and Moderators

The included studies were coded by the first and second author using a coding sheet according to the guidelines of Lipsey and Wilson (2001). *Stress* can be considered as the dependent variable and was coded into *physiological* or *psychological* stress-related outcomes, resulting in one meta-analysis. Multiple variables with a potential moderating effect on the relation between music therapy and stress were identified. These moderators were divided into outcome-, study-, sample-, and intervention characteristics.

Regarding the psychological stress-related outcomes, it was coded whether the psychological outcomes were assessed by means of questionnaires measuring stress or (state)

Figure 1
Flow Chart of the Search



anxiety. *State anxiety* can be seen as a psychological stress-related outcome, because many studies (e.g., de Witte, Spruit, et al., 2020; Hook et al., 2008; Ng et al., 2016; Zhang et al., 2014) considered state anxiety to be a result of stress and outcome measures related to state anxiety or stress. Therefore, in the literature these concepts are used interchangeably (Bradt & Dileo, 2014; Lazarus & Folkman, 1984; Ozer et al., 2013; Pittman & Kridli, 2011; Wetsch

et al., 2009). This is in line with the results of our previous meta-analysis, which showed no significant differences in effect sizes between state-anxiety self-report scales (*d* = .553) and stress self-report scales (*d* = .512). In the present study, 30% of the studies used Visual Analog Scales (VAS) to measure perceived stress or state anxiety. Overall, stress is often measured by the Perceived Stress Scale (PSS) (Cohen et al., 1983), the Quick Mood Scale (Woodruffe-Peacock et al., 1998), and the Profile of Mood States (POMS) (McNair et al., 1981), which instruments are used in 19% of the included studies. State anxiety is predominantly measured by the state version of the Spielberger State-Trait Anxiety Inventory (STAI) (Spielberger et al., 1983) and the anxiety version of the Hospital Anxiety and Depression Scale (HADS-A), which are used in 45% of the included studies.

Regarding the study characteristics, we coded the design, study quality, type of setting, type of control condition and whether the study was conducted in Western- or non-Western countries. Studies with prospective group design, such as RCTs and CCTs were considered relevant for the current research. Therefore, we coded study design as RCT when participants were allocated to treatment conditions through randomization (e.g., computer-generated randomization lists), and CCT design when authors did not explicitly mention randomization, or quasi-randomized studies. The quality of the study was coded as *strong, moderate* or *weak* after assessment with the 'Quality Assessment Tool for Quantitative Studies' (Effective Public Health Practice Project [EPHPP], 2009). This tool measures the quality of a study by providing a comprehensive and structured assessment of study quality (Armijo-Olivo et al., 2012). The EPHPP has been reported to have high content and construct validity (Jackson & Waters, 2005; Thomas et al., 2004). Low quality studies negatively affect the internal (causal conclusion) validity, which can lead to a biased estimation of the overall effect estimate (Higgins & Green, 2011; Zeng et al., 2015).

Regarding the setting in which the study was conducted, we coded whether the study was conducted in a *mental healthcare setting* or in a *medical setting* (e.g., during polyclinic treatments, before or after surgery, palliative care). Furthermore, the type of control condition was coded, because different control conditions can yield different effect sizes (Finney, 2000; Karlsson & Bergmark, 2015). We coded *care as usual* (CAU) when no stress-reducing intervention was offered, but patients did receive regular care within medical or mental healthcare, *waiting list* when there was no care or intervention offered, or *stress intervention* when another stress-reducing intervention was delivered, such as listening to prerecorded music, verbal support, or mindfulness-based therapy. Further, we coded whether the study was conducted in *Western* countries (European countries, Australia, USA, Canada, New Zealand) or whether the study was conducted in countries designated as *non-Western* countries (mainly Asiatic countries). The cultural environment has been shown to influence the way people respond to and cope with stress (Lonner, 2007; Tweed et al., 2004), which could influence the effect of music on stress. In our previous meta-analysis, the country in which the study was conducted just failed to reach the conventional level of

statistical significance (p = 0.089), indicating that non-Western studies yielded larger effects on physiological stress-related outcomes than studies conducted in Western countries.

Sample characteristics were also coded, such as the percentage of men in each study. There are indications that men and women react differently to stress, both psychologically and physiologically, leading to substantiated gender differences in measured stress levels (Galanakis et al., 2009; Kajantie & Phillips, 2006; Verma et al., 2011). We also coded the average age of the participants per study, because research on occupational stress revealed several differences in stress levels between different age groups (Galanakis et al., 2009).

Additionally, we coded nine music therapy characteristics. First, we coded whether the music therapy was offered to an individual patient or whether it concerned a group music therapy. Empirical evidence shows that during group music therapy interventions people synchronize with each other, which evokes positive feelings of togetherness and bonding, and decreases stress levels (Linnemann et al., 2016; Tarr et al., 2014). Second, we coded music therapy interventions as 'protocolized' or 'non-protocolized'. Music therapy protocols not only enable researchers to compare and replicate studies, but also to understand consistencies and strategies used by music therapists across sessions with participants (de Witte, Spruit, et al., 2020; Vink & Hanser, 2018). Both structure as well as strategies used during the therapy session may have impact on participants' outcomes, such as stress levels. Third, the quality of the intervention description was coded in *reported detailed* or *reported briefly and poor*. We considered the description of the therapy as detailed if authors mentioned or elaborated on components of a session of music therapy, such as the number or duration of the sessions, listening to live or recorded music, or which musical instruments or music therapeutic techniques were used. If authors did not explicitly report on most of the characteristics of the delivered music therapy (as mentioned above), the description was regarded as brief/poor.

Fourth, music style was divided into three categories: classical music offered by the music therapist, relaxation music, and selection of own-preference music by patient. Fifth, we made a distinction between the way the music was offered: whether the music therapist used live music alone, pre-recorded music alone or both. Sixth, with regard to music selection, we coded whether the music was selected based on the preferences of the patient, on the choice of the music therapist himself, or whether a pre-selected choice of music was offered. In some studies, in which the effects of music listening on stress-related outcomes was examined, it was advised to allow the subjects to choose the music themselves, because this may have a greater stress reducing impact (Brannon & Fiest, 2007; Juslin et al., 2008). However, our previous meta-analytic review showed that the term 'self-selected music' is used both in studies where the patient could bring her/his own preference music and in studies where the patient had to choose from a pre-selected list of music styles or songs (de Witte, Spruit, et al., 2020). Therefore, we coded as such in the present study.

Seventh, we coded whether the tempo of the music was 60-90 bpm or whether the music had another tempo. Tempo can be considered as one of the most significant

moderators of music-related arousal and relaxation effects. In the previous meta-analytic review (de Witte, Spruit, et al., 2020) of the effects of music interventions on stress-related outcomes, larger effect sizes were found in music with a tempo of 60-90 bpm compared to music with another tempo. Music with a slow tempo, such as meditative music, has often been demonstrated to initiate reductions in heart rate, resulting in greater relaxation (e.g. Bernardi et al., 2006; Bringman et al., 2009; Chlan, 2000; Hilz et al., 2014; Nomura et al., 2013). Lastly, we coded the number of music intervention sessions and the frequency of the sessions per week. The number of interventions has been shown to be positively correlated with stress and anxiety regulation (Cassileth et al., 2003; Gold et al., 2009; Robb et al., 2011).

2.4 Calculation and Analyses

The effect sizes were transformed into Cohen's *d* by using the calculator of Wilson (2013) and formulas of Lipsey and Wilson (2001). Negative effect sizes indicate that music therapy had a negative effect on stress-related outcomes. Most *d*-values were calculated based on reported means and standard deviations. To correct for pre-treatment differences, pre-test effects were subtracted from post-test effects. The effect size was coded as zero when a study reported that an effect was not significant without providing any statistics (Lipsey & Wilson, 2001). For both meta-analyses, the continuous moderators (age of the participants, gender of the participants, duration of the music intervention and frequency of the music van intervention) were centered on their means. For categorical variables, dichotomous dummy variables were created. Extreme outliers in effect sizes were identified using box plots (Tabachnick & Fidell, 2013), and were winsorized (i.e., replaced by the highest or lowest acceptable score falling within the normal range) for both meta-analyses. Standard errors were estimated using formulas of Lipsey and Wilson (2001).

In some of the studies, it was possible to calculate more than one effect size, as most studies reported on multiple stress-related outcome variables, multiple scales or measurement instruments. It is possible that the effect sizes from the same study are more alike than effect sizes from other studies. The assumption of independent effect sizes underlying traditional meta-analytic methods was therefore violated (Hox, 2010; Lipsey & Wilson, 2001). We applied a multilevel approach to meta-analysis in order to account for the interdependency of effect sizes (see Assink et al., 2015; Cheung, 2014; de Witte, Spruit, et al., 2020; Houben et al., 2015; Spruit et al., 2016).

A three-level meta-analytic model was used to calculate the combined effect sizes and to perform the moderator analyses. Three sources of variance were modeled, including the sampling variance for each effect sizes (level-one), the variance between effect sizes within studies (level-two), and the variance between studies (level-three) (Assink & Wibbelink, 2016). The meta-analysis was conducted in R (version 3.4.3) with the metafor-package, employing a multilevel random effects model (Houben et al., 2015; van den Bussche et al., 2009; Viechtbauer, 2010). This model is often used for multilevel meta-analyses and, in general, it is superior to the fixed-effects approaches used in traditional meta-analyses (van

den Noortgate & Onghena, 2003). We used likelihood-ratio-tests to compare the deviance scores of the full model and the models without variance parameters on level two or three to determine if the level-two and -three variances were significant, indicating heterogeneity of effect sizes. A heterogeneous effect size distribution indicates that the effect sizes cannot be treated as estimates of a common overall effect size. In that case, we conducted moderator analyses, because the differences among effect sizes may be explained by outcome, study, sample, and/or intervention characteristics.

2.5 Publication bias

A common problem in conducting a meta-analysis is that studies with non-significant or negative results are less likely to be published than studies with positive and significant results. The studies included in this meta-analysis may therefore not be an adequate representation of all studies that have been conducted, which is called the 'file drawer problem' (Rosenthal, 1995).

In order to check the presence of publication bias in the current meta-analysis, a trim and fill procedure was performed (Duval & Tweedie, 2000a, 2000b). In case of publication bias, the funnel plot of the distribution of effect sizes is asymmetric. We tested if effect sizes were missing on the left and right side of the distribution. Publication bias would only be likely to occur in case of non-significant or unfavorable (i.e., negative) results, resulting in left-sided funnel plot asymmetry. Right-sided funnel plot asymmetry is indicative of selection bias. We imputed estimations of effect sizes of missing studies through trim and fill analyses in the case of left or right-sided asymmetry, and subsequently computed an overall effect size that would take the influence of publication bias or selection bias into account (Duval & Tweedie, 2000a, 2000b), providing an estimate of the degree to which publication bias or selection bias might have affected the overall mean effect size.

3. Results

3.1 Overall effect of music therapy on stress-related outcomes

The present meta-analytic review on the effects of music therapy on both physiological and psychological stress-related outcomes, included 47 independent studies (*s*), reporting on 76 effect sizes (*k*), and a total sample of N = 2.747 subjects, of which n = 1.405 subjects in the music therapy groups, and n = 1.342 subjects in the comparison groups. Table A2 shows an overview of the most important characteristics of the included studies. Table 2 shows the overall effect of music therapy on both physiological stress-related outcomes and psychological stress-related outcomes. We found a significant medium-to-strong effect (d = .723, [.51-.94]) of music therapy on stress-related outcomes in mental healthcare and medical settings.

According to the trim-and-fill plot, the presence of publication bias was unlikely (see Figure 2), because studies were lacking on the right side of the funnel instead of the left side of the funnel. A trim and fill analysis yielded a marginally larger effect size of *d* = .783 compared to the observed effect size of *d* = .723. The likelihood ratio test showed that significant variance was present at the between-study level (level 3) and the within study level (level 2). We therefore conducted moderator analyses on type of outcome, study, sample, and music intervention characteristics to examine the effect of music intervention on physiological stress-related outcomes. The results are presented in Table 3.

Table 2
Overall Effects of Music Therapy on Stress-Related Outcomes

Outcome	*s*	*k*	Mean *d*	95% CI	*p*	σ^2_{level2}	σ^2_{level3}	% Var. level 1	% Var. level 2	% Var. level 3
Stress-related outcomes	47	76	0.723	0.510 – 0.936	<.001***	0.145***	0.649***	13.98	4.11	81.91

Note. s = number of studies; *k* = number of effect sizes; CI = confidence interval; Mean *d* = mean effect size (*d*); CI = confidence interval; % Var = percentage of variance explained; σ^2_{level2} = variance between effect sizes within the same study; σ^2_{level3}= variance between studies.

Figure 2
Trim-and-Fill Plot

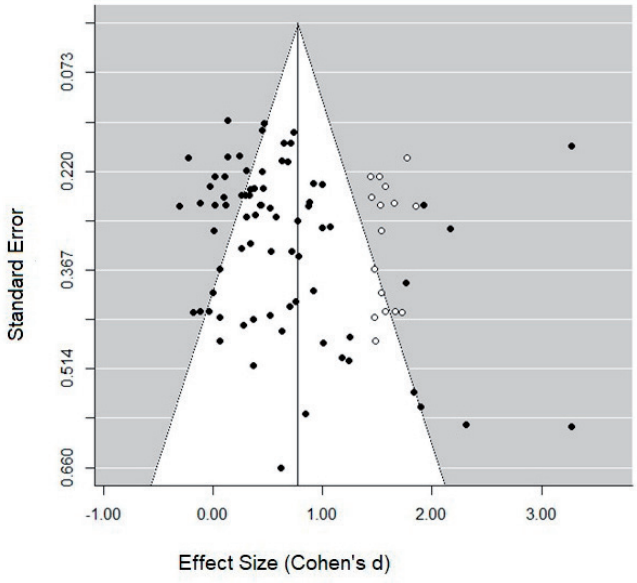


Table 3
Moderator Effects of Music Therapy on Stress-Related Outcomes

Moderator variables	*s*	*k*	β_0 (mean *d*)	t_0	95%CI lb, ub	β_1	t_1	*F*(df1, df2)
Outcome characteristics								
Domain of outcome	47	76						*F*(1, 74) = 0.270
Psychological outcomes (RC)	45	59	0.732	6.790***	0.52, 0.95			
Physiological outcomes	9	17	0.658	4.071***	0.34, 0.98	0.074	0.519	
Type of psychological outcome	47	76						*F*(1, 74) = 0.000
Stress (RC)	26	47	0.724	5.746***	0.47, 0.98			
State-anxiety	26	29	0.722	5.559***	0.46, 0.98	0.003	0.018	
Study characteristics								
Design	47	76						*F*(1, 74) = 13.344***
RCT (RC)	38	67	0.555	5.392***	0.35, 0.76			
CCT	9	9	1.449	6.52o*****	1.01, 1.89	0.895	3.653***	
Clinical setting	45	74						*F*(1, 72) = 1.030
Mental health care (RC)	12	14	0.880	4.153***	0.46, 1.30			
Medical health care	33	60	0.630	5.043***	0.38, 0.88	0.250	1.015	
Continent	47	76						*F*(1, 74) = 6.129**
Western countries (RC)	39	66	0.611	5.508***	0.39, 0.83			
Non-Western countries	8	10	1.306	5.066***	0.79, 1.82	0.869	2.747	
Study quality	47	76						*F*(2, 73) = 3.056+
Strong (RC)	11	15	0.444	2.077*	0.02, 0.87			
Moderate	20	33	0.589	3.730***	0.28, 0.90	0.145	0.545	
Weak	16	28	1.056	6.109***	0.71, 1.40	0.611	2.222*	
Intervention characteristics								
Therapy setting	42	71						*F*(2, 69) = 1.088
Individual (RC)	27	52	0.679	4.949***	0.41, 0.95			
Group	15	19	0.927	4.772***	0.54, 1.32	0.248	1.043	
Type of control condition	47	76						*F*(2, 73) = 5.400***
Waiting list (RC)	8	12	1.415	6.039***	0.95, 1.88			
CAU	28	44	0.561	3.891***	0.33, 0.79	-0.854	-3.265**	
Stress intervention	12	20	0.594	3.855***	0.29, 0.90	-0.820	-2.926**	
Treatment protocol used	47	76						*F*(1, 74) = 0.082
Yes (RC)	17	26	0.683	3.821***	0.33, 1.04			
No	30	50	0.747	5.512***	0.48, 1.02	0.064	0.286	
Description intervention	47	76						*F*(1, 74) = 0.482
Detailed	30	49	0.775	5.938***	0.52, 1.03			
Brief and poor	17	27	0.637	3.891***	0.31, 0.96	0.137	0.694	
Music style	47	76						
Own preference (RC)	18	40	0.688	4.803***	0.40, 0.97			*F*(2, 73) = 0.375
Relaxation	24	29	0.826	4.959***	0.49, 1.16	0.138	0.673	
Classical	6	7	0.562	1.775+	0.07, 1.19	0.136	0.349	
Music induction	47	76						*F*(2, 73) = 0.069
Live music (RC)	27	38	0.726	5.491***	0.46, 0.99			
Prerecorded music	10	18	0.664	3.258**	0.26, 1.07	0.062	0.285	
Both	10	20	0.767	3.849***	0.37, 1.16	0.040	0.180	
Music selection	44	73						*F*(2, 73) = 0.292
By therapist (RC)	15	25	0.695	4.232***	0.37, 1.02			
Choice of the patient	20	31	0.766	5.219***	0.47, 1.06	0.023	0.108	
Preselected choice	10	17	1.059	5.221***	0.66, 1.46	0.195	0.661	

Moderator variables	*s*	*k*	β_0 (mean *d*)	t_0	95%CI lb, ub	β_1	t_1	*F*(df1, df2)
Music tempo	47	76						*F*(1, 74) = 1.425
60-90 beats p/m (RC)	16	29	0.900	4.930***	0.54, 1.26			
No specific tempo	31	47	0.631	4.803***	0.37, 0.90	0.268	1.194	
Number of sessions (continuous)	38	60	0.748	5.663***	0.48, 1.01	0.016	1.175	*F*(1, 58) = 1.381
One or more sessions	38	60						*F*(1, 58) = 1.249
One session (RC)	14	32	0.594	2.824**	0.17, 1.02			
More than one session	24	28	0.894	5.346***	0.56, 1.23	0.300	1.117	
Frequency per week (continuous)	39	62	0.746	5.937***	0.49, 1.01	0.153	1.292	*F*(1, 60) = 1.668
Sample characteristics								
Proportion of males (continuous)	47	76	0.728	6.716***	0.51, 0.94	0.209	0.513	*F*(1, 74) = 0.263
Age (continuous)	46	75	0.718	6.431***	0.50, 0.94	0.001	0.1001	*F*(1, 73) = 0.010

Note. s = number of independent studies; k = number of effect sizes; β_0 = intercept/mean effect size (d); t0 = difference in mean r with zero; CI = confidence interval; lb = lower bound; ub = upper bound; β_1 = estimated regression coefficient; t1 = difference in mean r with reference category; F(df1, df2) = omnibus test; (RC) = reference category. + p < .10, * p <.05, ** p <.01,*** p < .001

3.2 Results of moderator analyses of music therapy on stress-related outcomes

Outcome characteristics. Both the domain of outcomes (physiological or psychological stress-related outcomes) and the type psychological measure (stress or state-anxiety measurements) did not influence the effects of music therapy on stress-related outcomes.

Study characteristics. Firstly, the strongest effects of music therapy on stress-related outcomes were measured by CCTs (*d* = 1.449, [1.01-1.89]) compared to RCTs (*d* = .555, [.35-.76]). Secondly, the continent in which the study was conducted did also moderate the overall effect. Studies from non-Western countries had a stronger influence on the overall effect of music therapy on stress-related outcomes (*d* = 1.306, [.79-1.82]) compared to studies from Western countries (*d* = .611, [.39-.83]). Thirdly, a significant moderating effect was found for type of control condition. Studies with a waiting list control condition yielded a larger effect (*d* = 1.415, [.95-1.88]) than studies with CAU (*d* = .561, [.33-.79]) or another stress-reducing intervention (*d* = .594, [.29-.90]). The clinical setting in which the study was conducted, did not moderate the effect. No significant differences were found between the effects of music therapy on stress-related outcomes in mental health care settings and medical settings. Furthermore, we observed that study quality moderated the overall effect with low quality studies (*d* = 1.056, [.71-1.40]) yielding larger effects compared to studies with a moderate (*d* = .589, [.28-.90]) or strong (*d* = .444, [.02-.87]) study quality .

Sample characteristics. The age (*d* = .718, [.50-.94] and gender (*d* = .728, [.51-.94] of the samples did not show to have a moderating effect on stress symptoms.

Intervention characteristics. Music tempo between 60 and 90 bpm yielded a larger effect (*d* = .900 [.54-1.26]) compared to music with no specific tempo (*d* = .631 [.37-.90]). Similarly, more than one session of music therapy had a larger effect (*d* = .894 [.56-1.23]) than one session (*d* = .594 [.17-1.02]). The effect size of preselected choice was larger (*d* = 1.059 [.66-1.46]) than music selection by the music therapist (*d* = .695 [.37-1.02]) and by the patient (*d* = .766 [.47-1.06]). With respect to music style, relaxation had a greater effect (*d* = .826 [.49-1.16]) compared to own preference music (*d* = .688 [.40-.97]) and classical music (*d* = .562 [-.07 to 1.19]). Additionally, group music therapy yielded a larger effect (*d* = .927 [.54-1.32]) than individual music therapy (*d* = .679 [.41-.95]). However, due to the small number of studies in certain categories (see Table 3), these differences were not statistically significant, and further studies are necessary to estimate these differences with more precision. Other differences had similar effect sizes (e.g., treatment protocol (*d* = .683 [.33-1.04]) or not (*d* = .747 [.48-1.02]); detailed intervention description (*d* = .775 [.52-1.03]) or brief/poor description (*d* = .637 [.31-.96]); and the way the music therapist induced the music – live music (*d* = .726 [.46-.99]), prerecorded music (*d* = .664 [.26-1.07]) and both (*d* = .767 [.37-1.16]). Lastly, the effect of frequency of sessions per week (*d* = .746 [.49-1.01]) was not significant.

4. Discussion

4.1 Overall Effects

Overall, we found a significant *medium-to-strong* effect (*d* = 0.723, [0.51, 0.94]) of music therapy on stress-related outcomes, indicating that participants receiving music therapy benefited more than controls. We conclude that music therapy is effective in reducing stress-related symptoms in both mental healthcare and medical settings. In our previous meta-analytic review, we found positive *small-to-medium* effects of music interventions on stress-related outcomes (see for more details: de Witte, Spruit, et al., 2020), while the findings of the present study demonstrate that music therapy yields a *medium-to-strong* effect on stress reduction. The difference in the strength of overall effect sizes may be explained by the different way both types of interventions are offered. The active involvement of a music therapist who is specifically trained to tailor interventions to the needs of patients and their musical preferences might give a reasonable explanation for the larger effect size for music therapy compared to music interventions (Bradt & Dileo, 2014; Dileo, 1999; 2006; Magee, 2017, 2019; Stegemann et al., 2019). Music therapists are especially trained to deliver music therapy sessions to meet participants/patients' needs at the individual or group level (Rafieyan & Ries, 2007).

The overall findings of the present meta-analysis are consistent with the findings of previous reviews and/or meta-analyses on the effects of *music therapy* on stress- and anxiety- related outcomes (Bradt & Dileo, 2014; Bradt, Dileo, & Potvin, 2013; Bradt, Dileo,

& Shim, 2013; Bradt et al., 2016; Carr et al., 2013; de Witte, Spruit, et al., 2020; Gold et al., 2009; Kamioka et al., 2014). In addition, the promising results of music therapy established in the current meta-analysis are in line with the findings of previous systematic reviews and meta-analyses on the effects of music interventions on the reduction of stress and/or (state) anxiety (Bradt & Dileo, 2014; Bradt, Dileo, & Potvin, 2013; Bradt, Dileo, & Shim, 2013; Bradt et al., 2016; de Witte, Spruit, et al., 2020; Gillen et al., 2008; Kim et al., 2015; Pelletier, 2004; Rudin et al., 2007).

Both the present meta-analysis and previous reviews show a growth in controlled clinical studies testing the effects of music therapy and/or music interventions on stress-related outcomes, which is important in order to formulate valid conclusions on the effects of non-pharmaceutical interventions for stress reduction (Casey, 2017; de Witte, Spruit, et al., 2020). The demand for more non-pharmaceutical interventions, such as music therapy, may be explained by the increasing awareness of the negative side effects of tranquilizing medication, such as substance dependence and abuse (Casey, 2017; World Health Organization [WHO], 2010). Although a considerable number of people around the world use tranquilizing medications to cope with daily life stressors or anxiety (e.g., Bandelow et al., 2015; Olfson et al., 2015; Puetz et al., 2015), previous studies show no convincing evidence for the short-term effectiveness of pharmacological treatment in the reduction of stress-related problems (Donovan et al., 2019; Olfson et al., 2015).

4.2 Effect Moderating Variables

Results of the present meta-analysis indicate that moderators explain differences in the strength of the effect size. Significant larger effects were found for studies using quasi-experimental CCT's compared to RCT's. Regardless of ethical concerns about the randomization of patients, the results of RCT's are still considered to provide the most robust evidence, because RCT designs can better exclude alternative explanations for established intervention effects than non-randomized designs. Selection bias in non-randomized effect studies can lead to overestimations of treatment effects (Page et al., 2018; Valentine & Thompson, 2013). In addition, we also found a significant moderating effect on the type of control condition: comparisons with a waiting list control group showed larger effects than comparisons with CAU or another stress-reducing intervention. This finding is in line with our expectation that CAU or another intervention would lead to reduction of stress, and thus to more stress reduction compared to a waiting list group. In addition, participants on a waiting list may also show a reduction in the stress level of symptomatology, which is shown in previous research in psychiatric populations (Arrindell, 2001; Haeyen et al., 2018). Specifically, Crawford et al. (2013) found that in spite of the positive scores of subjects of the music therapy group on stress reduction compared to subjects of the wait-list control group, the control participants also showed an improvement on stress reduction compared to baseline.

The overall effect size proved to be strongly moderated by the country in which the study was conducted (i.e., non-Western versus Western countries). Larger effects were

found in non-Western studies (*n* = 8), including studies conducted in Asian countries, such as China, Korea, and Taiwan, but also studies conducted in Iran, Brazil and Nigeria. It has been shown that the cultural environment influences how people respond to stress (Lonner, 2007; Tweed et al., 2004), which might explain differences between Western and non-Western countries. On the other hand, the non-Western countries show great heterogeneity in culture, socioeconomic characteristics or topographical region. Moreover, Western countries with large proportions of immigrants – such as the USA, Canada, and Australia – make it difficult to equate country with culture (Morales & Ladhari, 2011). Additionally, post-hoc analyses showed a weak correlation between Non-Western countries and study design (*r* = .34*, p* = <.01*)*, which indicates that the CCT design was more frequently used in Non-Western countries than in Western studies. Further research is needed to test particular explanations for cultural differences in effects between studies in Western and non-Western countries. Not only culture should be taken into account in future research, but also socioeconomic characteristics of study samples and the delivery of care in different health care systems, because of great heterogeneity both within and between countries.

Contrary to our expectations, we did not find evidence for a moderating effect of studies using a specific therapy protocol compared to studies without such a protocol. This can be explained by the fact that most studies included in our meta-analysis did not report on the use of such a therapy protocol, but still showed an adequate and rich description of the content of music therapy interventions. From the perspective of the music therapist, who is trained to tune in to the patient by adjusting the way of music-making as an immediate response to the patient's needs (Aalbers et al., 2019; Magee, 2019), music therapy protocols might often equal the flexibility of non-protocolled treatment in order to deliver personalized treatment, which increases the comparability or sameness of protocolled and non-protocolled treatment.

Notwithstanding we believe that there is a need for developing music therapy protocols and intervention descriptions that facilitate further replication of music therapy interventions and, subsequently, will better inform clinicians and practitioners in both mental health care and medical settings (de Witte, Spruit, et al., 2020). In addition, in future trials we strongly recommend examining treatment integrity as well, because music therapists may choose not to offer some of the elements specified in the protocol or to add new treatment elements. Having information on treatment integrity allows for the examination of the degree to which the implemented intervention approximates the intended intervention, and possible effects of treatment integrity on client outcomes (Perepletchikova, 2011; Vermilyea et al., 1984).

Study quality just failed to reach the conventional level of statistical significance, which indicates that low quality studies may yield larger effects compared to studies with a moderate or strong study quality. An explanation for this result could be that the degree of 'masking' was an important factor in assessing study quality. Masking of participants in music therapy studies is usually not possible unless two types of music therapy interventions are compared, such as receptive music therapy versus active music therapy (Bradt, Dileo, &

Shim, 2013). Masking procedures in which only the investigator is masked to the allocation of the intervention is much more feasible in music therapy trials (Day & Altman, 2000). Nevertheless, the present meta-analysis contained several studies in which the way of masking was not reported at all (*n* = 12), which is in line with the findings of Magee et al. (2017), who conclude in their Cochrane review that in future research reporting on the masking of participants and outcome assessors requires improvement. The lack of participant masking is problematic when studies examine subjective outcomes, such as mood or quality of life. Masking of therapists is often not possible in music therapy studies when active music-making is examined. When due to setting constrains the interventions cannot assure masking procedures, they should at least be masked to the purpose of the study where possible. In either case, masking procedures should be reported or discussed (Bradt, Dileo, & Shim, 2013; Magee et al., 2017).

Statistical analyses showed that the selected music therapy characteristics do not seem to moderate the overall effects of music therapy on stress-related outcomes. This could be explained by the diversity of the music therapeutic approaches and/or the applied interventions of the included music therapy studies, which is also mentioned in several previous reviews (Carr et al., 2013; Gold et al., 2009; Mössler et al., 2011; Silverman, 2003). On the other hand, this diversity in the content of music therapy can also be related to the core competence of a qualified music therapist, which means that the interventions are often tailored to what the patient needs or shows at that moment. Precisely this aspect of music therapy is the main difference with music interventions without a music therapist and could therefore have resulted in a larger effect size.

The selected intervention characteristics did not have a statistically significant impact on the effectiveness of music therapy. However, some substantial differences in effect sizes were found (*d* = .30 or larger) in moderator analyses that did not reach the conventional level of significance due to lack of statistical power, mostly caused by an unequal distribution of studies (and effect sizes) among moderator categories (see Table 2). We discuss some of these findings because they may be of particular theoretical interest, and probably should be addressed in future research.

First, there was a difference of *d* = .30 between the impact of only one session of music therapy (*d* = .594, [.17–.1.02]) and more than one session (*d* = .894, [.56–1.23]), indicating that the effect of music therapy on stress-related outcomes increases with the use of multiple sessions . The larger effect size for more than one session is in line with the study of Gold et al. (2009), which showed more substantial benefits in patients who took a longer course of music therapy or more frequent sessions. However, Gold's study examined the effects of music therapy in patients with severe mental disorders, whereas the present meta-analysis mostly included studies with patients suffering from much milder mental problems or patients with stress due to medical conditions. This does not diminish the importance of stress reduction, since stress is globally recognized as a major risk factor for the development of serious health

problems (American Psychological Association [APA], 2017; Australian Psychological Society [APS], 2015).

Notably, the effect of the number of sessions seems to be related to the type of setting (medical healthcare versus mental healthcare). In our meta-analysis, only studies conducted in medical healthcare settings measured one-session effects of music therapy on stress-related outcomes, which of course does not exclude the possibility that positive effects can just as well be measured within mental healthcare settings after only one session of music therapy. Nevertheless, our meta-analysis found empirical evidence for the short-term effectiveness of music therapy (i.e., a single session of music therapy) in reducing stress, and therefore puts the assumption that pharmacological treatment should be started due to its immediate and rapid effect in a critical light (Bandelow et al., 2015; de Witte, Spruit, et al., 2020; Fedoroff & Taylor, 2001). Moreover, the shown efficacy of only one single session may facilitate the implementation of music therapy in cases where for logistic reasons or in more complicated settings (e.g., during chemotherapy in the treatment of cancer, before or after surgery, or in palliative care) multiple sessions of music therapy would not be possible.

Second, the large effect of music with a tempo of 60-90 bpm (d = .900, n = 16) is worth mentioning. It is larger than the effect obtained in our previous meta-analytic review (de Witte, Spruit, et al., 2020), showing a medium effect (d = .625, n = 36) . The larger effect size found in the current meta-analysis may be ascribed to a lower amount of studies using prerecorded music than in our previous meta-analysis, which included mostly 'music medicine' interventions. Interestingly, a post-hoc analysis showed a significant strong correlation between music tempo of 60-90 bpm and prerecorded music (r = .61, p = <.01). Unfortunately, several studies did not report on the tempo used due to the fact that (1) interventions could vary across the music therapy session depending on participants' needs, and (2) the use of musical instruments varied considerably within and across the music therapy sessions. Moreover, the music tempo is usually not measured during a music therapy session of live improvised music are. We strongly recommend to investigate the influence of music tempo as a component of music therapy interventions, especially when targeting stress reduction. Moreover, literature also shows that music with a slow tempo and steady rhythm may provide stress reduction by altering inherent body rhythms, such as heart rate (Thaut et al., 1999; Thaut & Hoemberg, 2014).

Lastly, the moderator 'therapy setting' revealed a large effect for group music therapy (d = .927, [.54–1.32]). There is empirical evidence showing that group music activities stimulate the release of the stress-reducing neurotransmitters endorphin and oxytocin as a result of positive feelings of togetherness and bonding among group members (Linnemann et al., 2016; Tarr et al., 2014). In group music therapy, feelings of togetherness and bonding may be the result of non-verbal synchronization with each other by making music or listening to music, which offers a different experience of communicating and relating to others in a medium that has been shown to be motivating for people who otherwise find it difficult to share or engage (Carr et al., 2017; Gold et al., 2013; Stern, 2010). Moreover, research

shows that achieving synchronization by musical attunement is considered one of the most important (pre-)conditions in music therapy for eventually reaching stress reduction (Aalbers et al., 2019; de Witte, Lindelauf, et al., 2020). Facilitating synchronization as the basis for further interventions in music therapy is therefore regarded as one of the key competencies of a music therapist (e.g., Aalbers et al., 2019; Bruscia, 1987; Schumacher & Calvet, 2008; Wheeler, 2015). Finally, a post-hoc analysis showed a significant moderate correlation (r = .45, p = <.01) between individual music therapy and medical settings, which indicates that individual music therapy is relatively more used in medical setting compared to other settings.

4.3 Limitations of the Present Study

The current study has some limitations that need to be mentioned. Firstly, a significant number (n = 16) of the studies included in this meta-analysis had a small sample size (10-25 participants). Studies with small sample sizes are fairly common in meta-analyses (Davey et al., 2011), particularly when studies are conducted in medical or palliative settings where time and logistic constraints occur. It is important to highlight that small sample sizes in primary studies may result in great heterogeneity in treatment effects due to relatively large standard errors. Studies with small samples may also show greater clinical heterogeneity among patients compared to studies with large sample sizes, which may affect the outcome of the experimental treatment (IntHout et al., 2015; Schwarzer et al., 2015). Furthermore, the findings from small sample size studies tend to be less generalizable compared to studies with large number of participants. Furthermore, a limitation of any meta-analysis is that there is not a completely satisfactory way to test the presence of publication bias (Carter et al., 2019). The presence of publication bias can therefore never be ruled out, even if formal tests indicate that publication bias is unlikely. In fact, it is imperative that all clinical trials be preregistered in effectiveness research, including publication of the research protocols. In the present study, we chose to conduct a funnel-plot-based trim and fill method (Duval & Tweedie, 2000a, 2000b), which is commonly used in three-level meta-analyses in the domain of psychological studies (see Assink et al., 2019; Assink & Wibbelink, 2016; Zeegers et al., 2017), which seems a sufficiently sensitive method to detect publication bias in the current meta-analysis given the substantial number of studies and effect sizes, the magnitude of the effect sizes, and the degree of level 2 (within studies) and level 3 (between studies) heterogeneity of the overall effect size (See Assink & Wibbelink, 2016; Carter et al., 2019).

Although a clear search strategy to identify relevant studies has been performed, by for instance excluding observational and retrospective studies, most studies included in our meta-analysis lack masking procedure to participants. This particularly occurred in medical settings where due to the clinical condition of the participants the treatment group was disclosed. The majority of studies with small sample sizes and without a masking procedure were conducted in medical care. This might have influenced some of the study outcomes. Specifically, the lack of masking may have contributed to therapist expectancy (leading to therapist bias) and/or a patient expectancy-effect (also known as placebo effect), eliciting a

desirable therapeutic outcome (Tambling, 2012). In future studies, efforts need to be made to reduce expectancy or placebo effects, for example, by measuring expectation and/or adopting alternative experimental designs to control for these effects (Atwood et al., 2020; Boot et al., 2013). Notwithstanding the ethical reasons to refrain from a masking procedure and waiting-list design in anxiety and stress studies, it is important to further improve study quality and use larger samples. Next, we strongly recommend that future trials report on power analyses.

4.4 Concluding Remarks

The current meta-analytic review provides evidence that *music therapy* can be effective in reducing stress and provides justifications for the increasing use of music therapy carried out by a qualified music therapist in both mental health care practice and medical settings. Given the added value of the presence of a well-trained and qualified music therapist who offers music therapy, it is advisable to carefully consider whether music therapy is needed, or whether music listening interventions, mostly offered by healthcare professionals, are sufficient. In addition, low costs and lack of side effects of music therapy, and the moderate-to-strong stress-relieving effects of music therapy are very important for the prevention and treatment of stress-related problems. Nevertheless, with respect to the methodology of future trials, we strongly recommend reducing the risk of selection bias by aligning with the conditions of RCTs. Finally, the development of standardized music therapy protocols is necessary to conduct more robust research on the effects of music therapy, and to gain more insight into the moderating effects of characteristics of music therapy for stress reduction.

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CHAPTER 4

4

Music Therapy Interventions for Stress Reduction in Adults with Mild Intellectual Disabilities: Perspectives from Clinical Practice

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Abstract

Stress is increasingly being recognized as one of the main factors that is negatively affecting our health, and therefore there is a need to regulate daily stress and prevent long-term stress. This need seems particularly important for adults with mild intellectual disabilities (MID) who have been shown to have more difficulties coping with stress than adults without intellectual disabilities. Hence, the development of music therapy interventions for stress reduction, particularly within populations where needs may be greater, is becoming increasingly important. In order to gain more insight into the practice-based knowledge on *how* music therapists lower stress levels of their patients with MID during music therapy sessions, we conducted focus group interviews with music therapists working with adults with MID (*N* = 13) from different countries and clinical institutions in Europe. Results provide an overview of the most-used interventions for stress reduction *within* and *outside* of music. Data-analysis resulted in the further specification of therapeutic goals, intervention techniques, the use of musical instruments, and related therapeutic change factors. The main findings indicate that music therapists used little to no receptive (e.g. music listening) interventions for stress reduction, but preferred to use active interventions, which were mainly based on musical improvisation. Results show that three therapy goals for stress relief could be distinguished. The goal of "synchronizing" can be seen as a sub goal because it often precedes working on the other two goals of "tension release" or "direct relaxation," which can also be seen as two ways of reaching stress reduction in adults with MID through music therapy interventions. Furthermore, the tempo and the dynamics of the music are considered as the most important musical components to reduce stress in adults with MID. Practical implications for stress-reducing music therapy interventions for adults with MID are discussed as well as recommendations for future research.

Keywords: relaxation, mild intellectual disabilities, stress release, stress, music therapy, improvisation (music), mechanisms of change, change factors

1. Introduction

Stress reduction has become increasingly important in health care practices of many professional fields, including music therapy. Continuous high levels of stress have been shown to negatively affect our health (e.g., American Psychological Association [APA], 2017; Australian Psychological Society [APS], 2015). Although many people have found ways to adequately cope with stress and its possible negative consequences, it still appears to be difficult for a substantial number of people to cope with daily stressors, and the demands of contemporary life (Casey, 2017; Holahan et al., 2005; World Health Organization [WHO], 2010; de Witte, Pinho et al., 2020). This especially may be the case for adults with mild intellectual disabilities (MID[2]), who have been shown to experience much more stress in daily life than people without intellectual disabilities (Emerson, 2003; Hatton & Emerson, 2004; Schuengel & Janssen, 2006), having fewer resources to cope with daily life stress (Hartley & MacLean, 2009; Lunsky & Benson, 2001; Scott & Havercamp, 2014). Therefore, it is important to gain more insight into the effectiveness and applicability of therapeutic interventions for stress reduction for adults with MID.

Music therapy is often applied to reduce stress for adults with MID (Moonen & Didden, 2014; de Witte, Pinho et al., 2020, de Witte, Spruit, et al, 2020). The experience-based approach in music therapy is believed to be well suited to the needs of adults with MID. Although more and more studies are published on the *effects* of music therapy on stress-related outcomes (see for an overview: de Witte, Pinho et al., 2020), more information is needed regarding specific techniques, instruments, supposed change factors, requisites, and procedures within these interventions.

1.2 Music Therapy

Music therapy is defined as the clinical and evidence-informed use of music interventions[3] to accomplish individualized goals within a therapeutic relationship in order to meet physical, emotional, mental, social and cognitive needs (Agres et al., 2021; American Music Therapy Association [AMTA], 2018; de Witte, Pinho et al., 2020). In contrast to health care staff providing prerecorded music listening interventions (often referred to as music medicine), music therapy interventions are offered by a qualified music therapist in relation to assessment and treatment planning. Both music medicine as well as music therapy can be useful and important. According to the music therapy literature, music therapy interventions can be divided in two broad categories: active and receptive interventions (Bruscia, 2014; Magee, 2019; Wheeler, 2015).

2 By common Dutch definition adults with an IQ 50-70 as well as with an IQ 70-85 in combination with a disability in adaptive skills are considered to have a MID. In this article the abbreviation MID is therefore used to include people with what is often referred to as "borderline intellectual disability" (Kaal et al., 2015).

3 In this study, the term "intervention" refers to a specified therapeutic action or a process of intervening characterized by a structured and coherent collection of therapeutic actions.

Active interventions involve the patient making music during the music therapy session, while receptive interventions mean that the patient is only receiving the music, such as listening to live or prerecorded music. Whereas "music medicine" does not involve a therapeutic process, music therapy requires the presence of such a process through personally tailored music experiences, including listening to music (Bradt et al., 2015; Bruscia, 2014; Grocke & Wigram, 2006). Bruscia (2014) described a more refined classification of music therapy interventions/methods, resulting in four categories: improvisation, recreating (or performing), and composing, which are active interventions, and music listening, which refers to receptive interventions.

Music therapists are specifically trained to use the unique qualities of music, also known as musical components (e.g., melody, rhythm, tempo, dynamics, pitch) within the therapeutic relationship to work on patient's treatment goals (Bruscia, 1987; Wheeler, 2015; de Witte, Pinho et al., 2020). During the music therapy session, the music therapist attunes to the patient by adjusting the way of music-making as an immediate response to the patient's needs (Aalbers et al., 2019; Magee, 2019). In music therapy literature, this way of patient-therapist attunement is often related to the term "synchronization," which means that the music therapist and the patient interact simultaneously and are regulated through time, yielding a similar expression in movement, matching pulse, rhythm, dynamics and/or melody (Aalbers et al, 2019; Bruscia, 1987; Schumacher & Calvet, 2008; de Witte, Pinho et al., 2020). More specifically, synchronization in music therapy may refer to both the way music therapists intervene within different kinds of interventions, and to a specific music therapy technique for musical improvisation developed by Bruscia (1987), which can help music therapists to resonate emotions and/or expressions in the music or resonate non-verbal behavior. In this study, we consider synchronization in a broader sense.

In addition, music therapists often perform interventions based on rich experiential, tacit knowledge. Tacit knowledge refers to practice-based knowledge developed from the direct experiences within clinical practice, which is mainly subconsciously understood and applied (McAdam et al., 2007; Petri et al., 2020; Polanyi, 2009). Using tacit knowledge is common in professionals in clinical practice and guides the professional in treating patients based on professionals' previous experiences. However, making tacit knowledge explicit may stimulate transferability of valuable clinical practices, and may stimulate a different way of thinking about the relationship between clinical practice, theory, and research (Aigen, 1999; Smeijsters & Vink, 2006; Stige, 2015); furthermore, making tacit knowledge explicit may lead to the development of intervention descriptions derived directly from clinical practice, such as intervention manuals or therapy protocols, for which there is still a great need (Aalbers et al., 2019). From the scientific point of view, clear intervention descriptions are needed to further investigate what exactly is effective in music therapy interventions or to replicate outcome studies (Hoffmann et al., 2014). Our previous meta-analysis on the effects of music therapy on stress reduction showed, for example, that 63% of the included studies provided important details of the music therapy interventions examined, but only 35% of the studies

reported on the use of therapy protocols or manuals (de Witte, Pinho et al., 2020). Therefore, it is important for both clinical practice and future research to systematically illuminate the knowledge already present in clinical practice, such as the tacit knowledge of music therapists.

1.3 Experience of Stress in People with MID

Stress can be defined as the quality of an experience, produced through a person-environment transaction that, through either overarousal or underarousal, results in psychological and/or physiological distress (Aldwin, 2007; Riley & Park, 2015). Adults with MID, also defined as a neurodevelopmental disability characterized by deficits in intellectual and adaptive functioning skills, may demonstrate difficulties in coping with stress (American Psychiatric Association [APA], 2013). As in the general population, (continuous) high levels of stress experienced by adults with MID is associated with many negative mental health outcomes (Hartley & MacLean, 2009a; 2009b; Hulbert-Williams & Hastings, 2008; Scott & Havercamp, 2014).

In general, social support and self-efficacy can be considered as important factors for stress resistance (Everly & Lating, 2019). These factors are less developed in adults with MID (Abbaszadeh & Sardoie, 2016; Seyed et al., 2017). The relatively high impact of daily stressors, such as taxing social interactions, may also be explained by the lack of control over both minor daily and major life decisions (Dulin et al., 2013; Hartley & MacLean, 2009b; Scott & Havercamp, 2014). When stress experiences continue in adults with MID, it can lead to an increase of maladaptive coping strategies and serious health issues, such as depression (Hartley & Maclean, 2009a, 2009b), impaired cognitive functions (Heyman & Hauser-Cram, 2015), physical health problems (Lunsky, 2008), and substance abuse (Didden et al., 2009).

In clinical practice, the content of psychosocial approaches to interventions for adults with MID varies from strict cognitive-behavioral to that which is more experiential. Experiential approaches refer to those instances where the patient can learn through action-based experiences. Experiential approaches to intervention may allow for adults with MID to more readily address the reduction of stress. Experiential approaches to intervention focus on the “here and now” while guided by a therapist through stress responses and real-time emotional regulation. Through safely structured active experiences, stress inducing situations can be co-navigated, and stress reducing strategies can be developed and/or practiced (de Witte et al., 2017; de Witte, Pinho et al., 2020).

The efficacy of action-based experiences in interventions for adults with MID has been demonstrated in a study on the effects of the different components of cognitive-behavioral interventions for adults with MID. Studies have shown that behavioral activation strategies alone, such as roleplay exercises, resulted in the same outcome as the full cognitive therapy package (Cuijpers et al., 2007; Didden et al., 2016; Hamelin, et al., 2013). The cognitive-behavioral approach often results in patients being able to recognize that their thinking is not logical, but they do not feel emotionally different afterward (Patterson, et al., 2019). This lack of emotional address suggests the need for the further development of interventions

based on experiential approaches to intervention, including those that occur within music therapy processes.

1.4 Music Therapy for Adults with MID

Despite the lack of clinical effectiveness studies on music therapy for adults with MID, clinical practice shows that music therapists can meet the needs of adults with MID very well (Hooper, et al., 2011; Hoyle & McKinney, 2015; Watson, 2002). A scoping review (Hooper et al., 2008a, 2008b) provides a comprehensive summary of published studies about music interventions/activities for adults with learning and/or intellectual disabilities documented from 1943 to 2006; the researchers primarily located practice-based evidence from small sized studies of music interventions. Studies that specifically focused on music therapy with adults with MID were lacking. The majority of the published studies involved children with intellectual disabilities in a school setting (Meila, 2017), or concerned case study reports of music therapy processes in which stress regulation was not the targeted therapeutic goal (e.g., Brennand, et al., 2011; Hoyle & McKinney, 2015).

Since 2006, no studies have been conducted in which the content of music therapy interventions aimed at stress reduction for adults with MID was described clearly and systematically. However, there is a growing body of neuropsychological evidence showing the positive influence of music on lowering people's stress levels (Chanda & Levitin, 2013; Koelsch et al., 2016; Thaut & Hoemberg, 2014). Two meta-analytic studies showed positive overall effects of music listening interventions on stress reduction in different kinds of settings (Pelletier, 2004; de Witte, Spruit, et al., 2020). A recent meta-analytic review of 47 studies shows medium-to-strong effects (d = .723) of specifically *music therapy* on psychological and physiological stress-related outcomes (de Witte, Pinho et al., 2020).

1.5 Purpose of the Present Study

The purpose of this study is to systematically collect practice-based knowledge on both the most efficient music therapy interventions, such as therapeutic methods or exercises, and elements of those interventions, such as musical *techniques*, used by music therapists to reduce stress in patients with MID. This information may contribute to the body of knowledge on *how* music therapists lower the stress levels of their patients with MID during music therapy sessions. To be more specific, we wanted to know which interventions both within and outside of the music are used by the music therapists to lower the patients' stress levels, and which factors may influence the choice of the applied intervention(s).

2. Materials and Methods

To collect this practice-based knowledge, we set up multiple focus groups (Kitzinger, 2013; Liamputtong, 2011; Litosseliti, 2003) with music therapists from three different countries

and different types of clinical institutions who practice music therapy with adults with MID. Focus group methodology is characterized by guided group discussions to generate a rich understanding of participants' experiences and their tacit knowledge (Morgan, 1997; van Bruggen-Rufi et al., 2018). A focus group discussion is aimed at eliciting perceptions, attitudes, and ideas from participants about specific topics (Vaughn et al., 1996), and to enable them to react and build on the responses of others in the group about the topics that were broached (Kitzinger, 2013; Liamputtong, 2011; Litosseliti, 2003; Morgan & Scannell, 1998).

2.1 Participants

A total of 13 music therapists (male *n* = 6, female *n* = 7) based in The Netherlands (*n = 7*), Belgium (*n* = 3), and Germany (*n* = 3), who practice music therapy in institutions exclusively providing clinical treatment to adults with MID took part in three focus group interviews. Two music therapists were working in the same clinical institution, but in different departments. The age of the participating music therapists varied from 25 to 63 years, and overall, they were trained at different universities in The Netherlands, Belgium, and Germany. We chose to include music therapists from different countries, as results related to the specific context of one country may not be automatically generalizable to other countries. The study was approved by the Ethical Research Committee of the HAN University of Applied Sciences in the Netherlands (ref. no: 198.09/20).

2.2 Procedure

Purposive sampling was applied during the recruitment process. Purposive sampling allows for the selection of participants based on specific study driven variables or characteristics (Patton, 2014; Valerio et al., 2016). This sampling strategy was considered appropriate to ensure a group of participants that would be representative of the overall population of music therapists (Dörnyei, 2007; Etikan et al., 2016). Therefore, participants were recruited from a variety of clinical institutions, taking into consideration age, gender, country of origin, and educational background. The intention for diversity sought to ensure that overall findings would not be influenced by over-representation in these areas. Once selected, music therapists were invited by email to participate in this focus group study. The music therapists who asked for more information received a second email in which the general theme of the study was described. Since the aim of the present study is to gather practice-based knowledge, no in-depth information or topic related literature was shared prior to the focus group meetings. Before the focus group meetings started, the participants were asked to sign an informed consent form, in which they clarified their willingness to participate, and their spoken data could be used anonymously for the purpose of this study.

Eligible focus group participants were (a) qualified and board-certified music therapists (b) who apply music therapy to patients with MID, (c) within a clinical or healthcare setting in which (d) patients were specifically referred to music therapy by a psychologist or psychiatrist. Music therapists without significant experience with patients with MID, such as those who

had just recently started working as music therapists or those who have only treated a small number of patients with MID, were not approached. Furthermore, we did not include music therapists working in a private practice, because in this setting it is not always clear whether a patient meets the diagnostic criteria of MIDs.

2.3 Data Collection

Prior to the focus group interviews, an interview guide was developed in which the research question, the purpose of the study, the topics and preconditions of the focus group were set up. Subsequently, a time-bound questioning route was developed, in which questions and sub-questions were formulated regarding each topic. In order to stimulate a rich discussion about which interventions music therapists use to reduce stress in adults with MID, we included questions about both interventions *within* the music, as well as interventions *outside* the music, and possible effect moderating conditions (the full questioning route used can be requested from the first author). Although the questioning route was followed to keep the discussion on track without inhibiting the flow of ideas, the focus group promoted an open character by stimulating all participants to give their views on the subject. For example, by using follow up questions, researchers could give participants the space to respond to each other's views.

The focus group was led by a moderator with advanced research skills and a Master's degree in music therapy. An assistant moderator was also present to ensure that the equipment for the audio recordings worked correctly, to manage the time and take notes during the focus groups, and to join the conversation if warranted. A general introduction of the research project was given, before starting the focus group discussion. After each focus group session, the moderator and the assistant moderator evaluated the notes taken. The first focus group session included the participants who were working in The Netherlands. Subsequently, two identical focus group sessions were held, one with Belgium-based participants and one with participants based in Germany.

2.4 Data Analysis

After data-saturation was established, researchers analyzed the information using the coding principles of qualitative content analysis. Qualitative content analysis is frequently applied to answer questions such as *what*, *why,* and *how*, whereby the common patterns in the data are deduced by using a consistent set of codes to organize text into identified categories of similar meanings (Cho & Lee, 2014; Moretti et al., 2011; Nandy & Sarvela, 1997). Qualitative content analysis is based on naturalistic inquiry, which entails identifying themes and patterns, and involves rigorous coding (Cho & Lee, 2014; Moretti et al., 2011).

In preparation of the coding process, the audio recordings of the focus group sessions were fully transcribed. Then, consistent with the principles of content analysis, we applied three successive coding steps (i.e., open, axial, and selective coding) to analyze the transcripts (Corbin & Strauss, 2008). The first open-coding step was conducted by two researchers, who

independently coded relevant text fragments based on "in vivo codes". The relevant text fragments were labeled with the literal terms used by the participants (Corbin & Strauss, 2008). In case of disagreement between the two researchers, the topics were discussed by a larger group of researchers (the co-authors of this article) and resolved by consensus. The second step involved axial coding, where the open codes were grouped into categories based on their more overarching similarities to the property and dimension levels (Corbin & Strauss, 2008). During this axial-coding process, it became possible to define preliminary analytical (sub)categories and compare these. The third step concerned selective coding, in which the categories formed during the axial coding procedure were connected in order to create and refine an integrating model (Charmaz, 2003).

The cycle of data-analysis (the three coding steps: open, axial and selective coding) was executed by two different researchers in which an iterative process and "constant comparison" were the leading principles (Corbin & Strauss, 2008). To ensure that the findings were grounded in the initial data, every step of the data-analysis was continuously audited/coached by both a professor specialized in music therapy and a professor specialized in mental health issues of adults with MID.

2.5 Criteria of Trustworthiness

To meet the criteria for trustworthiness, several techniques were applied to enhance the quality of the present focus group study (e.g., Creswell, 1998; Denzin & Lincoln, 2005). Transferability was strengthened by the fact that the included music therapists were employed in a variety of mental health care centers that specialize in the treatment of patients with MID.

To minimize possible bias of the interviewer, member checking took place, which met the criteria for credibility. In preparation of the analysis, the co-moderator summarized the content of each focus group discussion, which subsequently was member-checked by the participants individually by email. All the participants agreed with this report, and none of them suggested any additional comments or changes. The fact that every step of data-analysis was continuously audited by other research experts, helped to ensure that the findings were not biased by the main investigator's motivation or interest, thus meeting the criteria of confirmability.

3. Results

The open coding initially showed a mixture of many different types of music therapy interventions, in which a distinction could be made between interventions within the music and interventions outside of the music.

3.1 Therapy Goals for Stress Reduction

Three types of therapeutic goals were mentioned by the music therapists related to stress reduction in adults with MID: synchronizing with the patient (as a starting point for stress reduction), releasing stress or tension (by self-expression), and stimulating relaxation. The music therapists stated that in general these goals do not stand alone but are used in succession of or in combination with each other. This means that the goal of achieving synchronization with the patient within the music can be seen as a sub goal, and often precedes working on the goals related to the release of tension or direct relaxation.

"I think when treating patients with MID, they first have to come into their normal tension level before start working on other treatment goals. So, every single music therapy session is starting with the lowering of the patient's tension level and after that we continue with the common therapy activities [participant 4, music therapist in the Netherlands]."

Table 1 shows a detailed overview of all the mentioned interventions and related characteristics. Because the data did not provide a detailed description of every mentioned intervention, not all columns could be filled for all the interventions mentioned. Nevertheless, in order to provide a realistic representation of the results we chose to show all the findings, even if this meant that some areas of the table would be left blank.

Table 1
Overview of interventions

Music therapy interventions within the music		Goal	Instruments	Specification of the intervention	Assumed change factors promoting stress reduction
Active interventions	Free Improvisation	Synchronizing with the client (as a starting point for relaxation or releasing tension)		• Attuning to the client's tempo • Following the client's dynamic • Using the same pulse	
		Relaxation	Piano Monochord Guitar Percussion - Drum kit - Rainmaker - Ocean drum Singing bowls Wind chimes	• Decreasing music tempo • Playing simple harmonies or chord progressions (third, fifth, octave) • Using a low tonal register • Playing a simple chord progression on piano • Inducing playfulness • Working towards musical form • Incorporating patterns • Tapping/moving along to the music • Structuring the music expression • Playing in a slow tempo • Inducing minimal changes in tempo • Inducing minimal changes in melodic intonation • Using specific Bruscia techniques: - Pacing - Rhythmic grounding - Tonal centring	Simple instruments and harmonies may impart support, the use of complicated instruments may cause stress Triggering and synchronizing of multiple senses Music may support the sense of containment. Percussion (drum kit) may impart delimitation and physical distance from the therapist
		Releasing tension	Piano Drums	• Accelerating tempo • Alternating between increasing and decreasing of certain musical components • Gradually increasing musical tension • Building up dynamics • Increasing the musical tension • Collectively playing an unstructured drumroll	Shared experience by playing the piano together After an increase of tension, a decrease may be experienced more intense or more vivid

Music therapy interventions within the music			Goal	Instruments	Specification of the intervention	Assumed change factors promoting stress reduction
Active interventions	Structured improvisation	Freestyling	Releasing tension	Singing and/or rap	• Playing an aggressive tight beat • Playing a heavy bass	Physically experiencing the beat, may lead to become aware of body signals and tension Freestyling may offer space for the direct expression of frustration
		6/8-meter Improvisation	Relaxation	Drums	• Using a steady pulse of 60 bpm • Using 6/8 meter • Playing the same for the duration of 5 to 10-minutes • Initiating no increases in tempo	6/8 meter is soothing and relaxing, and can elicit related emotions 6/8 meter can be experienced as a mantra (meditative) 6/8 meter consists of a clear pulse/rhythm, which is associated with calmness
		Interplay on piano		Piano	• Harmonic basis offered by the therapist • Dictating client's tempo and pulse • Letting the client choose minor or major scale • Instructing the client to start with single key and later to extend play with additional keys	Experiencing intrinsic rhythm
		Taking turns				
		Expressing feelings in music		Instrument of own choice Cymbal (as a stop signal)		
		Singing mantras		Singing with the use of singing bowls		The repetitive nature of mantra singing
		Playing intrinsic tempo		Drums	• Playing client's own intrinsic tempo • Playing each other's intrinsic tempo	Experiencing your own intrinsic tempo

Music therapy interventions within the music		Goal	Instruments	Specification of the intervention	Assumed change factors promoting stress reduction
Active interventions	Singing and/or playing existing songs: Greek Sirtaki	Externalising clients: Releasing tension Internalising clients: Promoting self-expression	Therapist plays accordion Clients play a variety of percussion instruments	• Starting at a low tempo • Increasing music tempo • Shouting: 1234 HOPPA • Playing rhythm collectively • Implementing changes in tempo	Structure may promote stress release for externalising clients Coaxing may promote stress release for internalising clients
	Singing/ playing well-known songs			• Making variations of well-known songs • Starting with children's songs • The use of age-appropriate songs	Familiarity of well-known songs
	Using the same opening song each session				Repetition may create sense of safety
	Song writing and/or composing: Recording self-composed songs (and taking home)	Releasing tension transferred to outside the music therapy session			The use of self-composed stress relieving songs outside of the music therapy sessions (transfer)
	Song writing	Releasing tension			Writing songs about stressful events, may offer exposure
Receptive interventions	Listening to pre-recorded music: Recording and/or curating personal relaxation music	Relaxation outside the music therapy session			

3.2 Interventions Within the Music

The open coding indicated that the interventions within the music could be categorized in two domains: active and receptive music therapy interventions. The axial coding led to several different types of active interventions, and two types of receptive interventions.

3.2.1 Active Interventions

The active interventions mentioned were categorized in a number of subcategories (see Table 1). For each of these subcategories, the coding led to several intervention characteristics, such as associated therapeutic goals, the musical instruments that were used, and the change factors that may possibly clarify the effect of the chosen intervention. Improvisation-based interventions concerned the first type of active interventions. Other types of active interventions included "singing and/or playing existing songs" and "songwriting and/or composing". Lastly, the music therapists mentioned a variety of active interventions that referred to a certain music therapy method or approach. These were recognized as "other interventions" (see Table 1).

Improvisation. The music therapists agreed that improvisational method was one of the most often used active interventions. The second step of data-analysis showed that these improvisation-based interventions could be distinguished from each other by the level of structure, the applied improvisation techniques, the goal of the improvisation, and/or the used musical instruments. With respect to the process of selective coding, improvisation-based interventions for stress reduction could be subdivided in "free improvisation" and "structured improvisation". Structured improvisation interventions refer to "freestyling," "6/8-m improvisation," "interplay on piano," "expressing feelings in music," "taking turns," "mantra singing," and "playing intrinsic tempo" (see Table A3 for an extended description of each of these interventions). Most music therapists mentioned that they mainly preferred to use *free improvisation*, meaning that they start improvising with patients without using a predetermined musical structure or certain rules for the music-making.

Participants appeared to identify two main therapeutic goals within free and structured improvisation: tension release and reaching direct relaxation. Synchronization is also mentioned as a goal, but the participants explained that they used it more as a sub goal. Specifically, free improvisation often starts with *synchronizing with the patient*, which can be achieved by attuning to the tempo and following the dynamics and intensity of the patients' music-making, as can be seen in the quotation below. Subsequently, the music therapist tries to implement repetition and/or structure to allow the patient to slow down and 'get a grip' on their stress experience. Concerning synchronization as a sub goal, no specific musical instruments were mentioned.

"I often try to lower the stress level of the patient by first playing along with the patient, and then gradually lower the music tempo. As a therapist you hope that they synchronize with the music [participant 11, music therapist in Belgium]."

"For example, when the patient is playing the drums and is playing well enough, at least when you hear a rhythm, you can play along on the piano with only 4 chords [participant 3, music therapist in the Netherlands]."

The first therapeutic goal for improvisation-based interventions concerned *releasing experienced tension*, which the music therapist tries to achieve by increasing the intensity of the music. One music therapist mentioned tension release as a goal for "freestyling," which is classified as a more structured improvisation intervention (see Table 1) because of using a fixed beat and/or musical structure on which the freestyling (often through voice work) takes place. According to most of the participants, music improvisational methods focusing on tension release are characterized by starting with building up (accelerating) the music tempo and dynamics until a peak/climax is reached, and then slowly decreasing the tempo/dynamics. A potential change factor that was mentioned, which possibly contributes to achieving the goal, is that after an increase of (musical) tension, a decrease of this tension is experienced more "vividly" by the patient. Piano and percussion instruments were named as the musical instruments most often used in free improvisation interventions aiming at tension release.

"I first try to structure all the musicality that is there, a clear pulse, say, a clear rhythm. From there I often continue with the release of tension by playing faster and faster through differences in dynamics and in tempo [participant 9, music therapist in Germany]."

"I often use a form where you build up very slowly by making loud sounds and vocals, as a counterpart to a very structured intervention. Just a very free and fast way of expressing, in which the patient can connect to his/her feelings and then eventually release and discharge [participant 10, music therapist in Germany]."

Participants also noted *direct relaxation* as a therapeutic goal in improvisation-based interventions. Free improvisation aiming at relaxation is mainly characterized by the simplicity of the musical structures, such as playing in a low tonal register, using simple chord progressions, making minimal changes in music tempo or melodic intonation, and using easy-to-play musical instruments (e.g., monochord, wind chimes, ocean drum, rainmaker). In contrast to free improvisation aiming at the release of tension, free improvisation for direct relaxation is characterized by the use of a consistently low tempo, while changes in melody, harmonics, and tempo are kept to a minimum. The music therapists stated that it is precisely this simplicity of the musical improvisation that provides the patient with feelings of support and containment, both of which can be regarded as certain *change factors* for achieving stress reduction in adults with MID. To reach relaxation by using free improvisation, three improvisation techniques (Bruscia, 1987) were regarded as helpful by some of the music therapists, i.e., "pacing," "rhythmic grounding," and "tonal centering." The music therapists

mentioned that they mainly use harmonic instruments (e.g., piano and guitar) and (melodic) percussion instruments (such as chimes and singing bowls).

"After a while, you lower the music tempo, and you will see that the patient is almost often following you in this and you see his stress level is lowering"[participant 3, music therapist in The Netherlands]."

"So, a solid beat is needed. That is how I see it. When patients with MID are highly tensioned, they have to experience themselves more, their own body, otherwise they lose it. By using a steady and solid beat, they feel it in their body, which is an important experience for them and helps them [participant 8, music therapist in Germany]."

Performing existing songs. Besides improvisation, "singing and/or playing of existing songs" was the type of active intervention most often mentioned by the music therapists. Interventions concerning singing by patients were singing well-known European-based cultural songs, singing the Greek Sirtaki (mentioned by one music therapist) and the use of the same opening song at the start of every session. The Greek Sirtaki intervention focuses on releasing tension and stimulating self-expression and is characterized by a (slow) increase of the music tempo, and clear rhythmic structures. The music therapist who often used this intervention clarified that the clearly defined rhythmic structure particularly promotes stress release in MID patients with externalizing behavioral patterns and can be seen as a possible change factor. With respect to the use of both "well-known songs" and "always using the same opening song," the music therapists noted that it may provide a patient with feelings of safety due to potential linkages to cultural musical representations for patients. The presence of a repeating musical structure and familiarity can be regarded as possible change factors related to these effects (see Table 1).

Songwriting and composing. The active intervention category "song writing and/or composing" was mentioned only in the focus group held with the Dutch music therapists (n = 7). One of the participating music therapists clarified that music therapeutic songwriting can stimulate exposure to stress-causing situations and thereby help patients to release stress. Several other music therapists added that recordings of self-composed songs could also be helpful for patients to reduce stress by listening to the recordings outside of the music therapy session.

Other interventions. Some of the active interventions mentioned by the music therapists did not fit any of the previously described categories, but were based on a certain methodic framework (Hegi, Schuhmacher, or Ronnie-Gardiner) or the more protocol-oriented music therapy interventions (Neurologic Music Therapy [NMT]). One of the interventions mentioned was called "Eentoonssymphonie [One note symphony]," which originated from the clinical practice of the Dutch music therapist Berman (2016), and concerns music-making with just one note (see Table A3 for more detailed descriptions of these active interventions).

3.2.2 Receptive Interventions

In addition to the active interventions, the music therapists of this study also mentioned one receptive intervention, namely creating a personalized playlist for relaxation. Music therapists in all three focus groups referred to this receptive intervention for stress reduction. The personalized playlists can include both recorded musical improvisations and compositions made during the music therapy session(s), as well as pre-existing music of personal preference. The main goal focuses on "transfer," meaning that patients can use this prerecorded music to lower their stress levels through relaxation outside of the music therapy sessions.

3.3 Interventions Outside of the Music

The music therapists mentioned some stress-reducing interventions outside of the musical context, which are related to the therapeutic attitude of the music therapist. A majority of the Dutch focus group participants stated that creating a framework may offer support to the patient. This framework can be seen as a solid base of structure visible in both the interventions within the music and the way the music therapists interact with their patients. The music therapists indicated that after offering a structure or framework during a session, they simply "let the rest happen" and let the patient respond. One of the therapists stated that in some cases the music therapy session started with "doing nothing," because often a starting point arises naturally, with which one can then continue working. The Belgian music therapists explained that they always base the starting point of their interventions on the patient's needs.

Most of the music therapists considered the level of autonomy (self-control) they offered their patients, as an important aspect of the therapeutic attitude for reaching stress relief in patients with MID. The degree of autonomy provided to the patient depended on their individual needs and their capability to handle their self-control. One of the Dutch music therapists noted how important it is to take into account the patient's strengths and limitations, such as not to have them feel overwhelmed or over-questioned. This sentiment was echoed by the German participants.

3.4 Factors Influencing the Choice of the Intervention

Participants of two focus groups mentioned some factors that could possibly influence the stress-reducing effect of the applied music therapy interventions. To keep the results as close as possible to the initial data, we report a summary of the responses of the music therapists to this specific question.

The degree of intellectual impairment was considered as a factor for determining the choice of interventions. The Belgian music therapists stated that the degree of intellectual impairment (i.e., the IQ score) influences the effects of certain music therapy interventions, and therefore also influences their choice of intervention. They explained that when working with patients with MID from the upper segment of the IQ range, they often use rhythm-oriented interventions, such as drumming in the same beat or tapping along with the music

on small percussion instruments. This is in contrast to interventions they offered to patients with MID in the lower segment of the IQ-range, which were characterized by a more multi-sensory approach and the use of tonal instruments.

Another factor possibly influencing the choice for a certain intervention was the mood or preference of the music therapist himself. For example, the music therapist may choose interventions that he/she is comfortable with and that makes him/her more relaxed and grounded as a therapist, which in turn is considered to have a positive influence on a patient.

Lastly, the patient's preferences (like a specific music style, song, or instrument) were also mentioned as a factor that influenced the choice of intervention. Almost all the music therapists mentioned that they always tried to take the patient's preferences into account. On the other hand, they tried to avoid the use of existing songs that were "too familiar," as they noticed that patients tended to stick too much to the familiar lyrics instead of being aware of the musical experience.

4. Discussion

In this study, we tried to explicate practice-based knowledge, also known as "tacit-knowledge," about *how* music therapists can reduce stress in adults with MID during the music therapy session. The purpose of the present study was to describe music therapists' experiences and perceptions on what works in their own practice, and provide an overview of music therapy interventions to generate an immediate stress-reducing effect in adults with MID. We discuss the implications of the results of this study by comparing them with the literature at large. Furthermore, we explore the facets of stress reduction, the musical components connected with such, and consider limitations of this study while providing implications for future practice, theory, and research.

4.1 Two Ways to Reduce Stress in Music Therapy

Results of the present study distinguished two ways of intervening to reduce stress in adults with MID, which are related to the therapy goals mentioned by the music therapists. The achievement of (musical) *synchronization* with the patient, which can be regarded as a sub goal, often precedes working on one of the other two goals: *release of tension* or *direct relaxation*, which clearly leads to two different ways of intervening during music therapy.

The first way of intervening is related to the release of tension and aimed at attuning musically to the patient's perceived tension/stress, which often results in music with an increased tempo and dynamics, and fast rhythms. After this, the music therapist further increases the intensity of the music, for example by accelerating the music tempo, so that the patient can release the felt tension. When the music intensity has come to a climax, the music therapist starts to slowly decrease the intensity of the music and actively guides the patient to decrease the music tempo to 60-80 bpm (beats per minute). This corresponds with

previous research, which suggests that the tempo and dynamics of music are important for the experienced intensity of the music (Gabrielsson & Lindström, 2010). The second way of intervening is related to the achievement of direct relaxation. In contrast to the first way of intervening, the music therapist's interventions are only focused on providing relaxation by synchronizing with the client's music, and gradually decreasing the music tempo to 60-80 bpm and the volume to silence.

Both ways of intervening, aiming at the *release of tension* or *direct relaxation*, have common ground with the so-called "ISO-principle", which refers to a music therapy technique by which the music is matched with the mood of a patient, then gradually altered to affect the desired mood state. This technique can also be used to affect physiological responses, such as heart rate and blood pressure (Davis et al., 2008).

4.2 Musical Components

Within these two ways of stress reduction during music therapy, the *tempo* and the *dynamics* (also known as "loudness") of the music can be considered as the most important musical components to reduce stress or tension in adult patients with MID. Studies suggest that music with a slow tempo can be considered to be one of the most significant determinants of audio-related effects on stress reduction (e.g., Bernardi, et al., 2006; Björkman, et al., 2013; Iwanaga et al., 2005; Jiang et al., 2016; Nilsson, 2008). Tempo and changes in tempo can influence different physiological and neurological responses, such as arousal, motor activity and motivation (Roth, 2014), this means that music with a slow tempo and steady rhythm may provide stress reduction by altering inherent body rhythms, such as heart rate (Thaut et al., 1999; Thaut & Hoemberg, 2014). Therefore, an increase in music tempo can lead to the increased activation of the nervous system, muscle tension and heart rate, whereas a decrease of the tempo can lead to muscle relaxation and a lowered heart rate resulting in more relaxation (Bernardi et al., 2006; Bringman et al., 2009; Juslin & Västfjäll, 2008; Nomura et al., 2013).

4.3 Active Music Therapy Interventions

The present study makes a clear distinction between active and receptive music therapy interventions, which is in line with the music therapy literature (e.g., Bruscia, 2014; Magee, 2019; Wheeler, 2015). The results show that the participants of this study prefer the use of active music therapy interventions to reduce stress in adults with MID, compared to receptive interventions. It is preferred to involve the patient in making music and/or singing to decrease the level of stress instead of offering receptive interventions, in which the patient only receives the music, such as listening to live or prerecorded music (Bruscia, 2014; Magee, 2019; Wheeler, 2015). A possible explanation could be that the interaction between patient and therapist during active interventions may lead to "affect attunement" because of the possibility to (co)-modulate, and (co)-regulate emotional states through the different musical components, such as rhythm, tempo or dynamics (Aalbers et al., in press; Raglio et al., 2016).

Similarly, findings of the present study suggest that synchronizing with patients' music making and facilitating changes in musical expression by reducing tempo and volume, can lead to stress reduction. This is consistent with previous studies and literature, which describe music therapists' use of synchronization techniques to attune to the patient for the purpose of change (e.g., Aalbers et al., 2019, 2020; Bruscia, 1987, 2014; Dvir et al., 2020).

Surprisingly, active improvisation interventions for stress reduction were used and few to none receptive interventions, whilst in reviews and meta-analyses on the effects of music or music therapy interventions on stress-related outcomes mainly receptive music listening interventions could be included (e.g., Bradt et al., 2013; Pelletier, 2004; de Witte, Pinho et al., 2020; de Witte, Spruit, et al., 2020). The effects of music listening interventions are mainly related to the general influence of music on the stress response, whereas the effects of music therapy also can be explained by means of the therapeutic relationship, namely, through patient-therapist attunement by the use of music and the fact that music therapists individualize their interventions to meet the patients' needs (Bradt & Dileo, 2014; Wheeler, 2015; de Witte, Pinho et al., 2020). Although none of these reviews and meta-analyses included adult patients with MID, it raises the question of whether music therapists should use receptive interventions more frequently because of their effects on stress reduction. On the other hand, there is evidence showing that action-based interventions are very suitable to adults with MID (Cuijpers et al., 2007; Didden et al., 2016), which could favor active music therapy interventions.

4.4 Music Therapeutic Improvisation for Stress Reduction

Results show that the participants of this study commonly used active improvisation interventions for stress reduction, while their use of receptive intervention processes was either limited or non-existent. Literature suggests that improvisational methods are highly common in music therapy; within musical improvisation, patient(s) and therapist improvise on musical instruments they have chosen and play together freely or with a given structure (Gold et al., 2009; MacDonald & Wilson, 2014; Wigram, 2004). Gold et al. (2009) emphasize in their review that improvisational music therapy techniques can be regarded as the most important interventions for achieving social interaction in active music therapy. In addition, musical improvisation can also be seen as a mode of self-expression, where the expressive character of musical interactions enables the release of difficult or repressed emotions (Burns et al., 2001; Gilboa et al., 2006; MacDonald & Wilson, 2014), which corresponds with the therapy goal of achieving tension release, found in this study. However, researchers reported that the use of improvisation may also evoke stressful experiences, necessitating the presence of an educated and qualified music therapist to attune from moment to moment to the patient's needs (Aalbers et al., in press; Magee, 2019; de Witte, Pinho et al., 2020). Thereby, the understanding of the links between the music-related mechanisms and synchronization may help music therapists to make informed decisions on how to use musical improvisation to

lower the patient's stress levels (Aalbers et al., in press; MacDonald & Wilson, 2014; Moore, 2013; de Witte, Pinho et al., 2020).

4.5 Change Factors

Results of this study identified some factors that may contribute to the explanation of the perceived effects of specific music therapy interventions for stress reduction in adults with MID. In the body of literature, these factors are often described as "change factors (Cuijpers et al., 2019; Lambert, 2013)." An important change factor found for promoting stress release during music therapy in adults with MID concerns the amount of simplicity involved in the chosen interventions. Examples of this are the use of mantra techniques, which have a repetitive character, or the choice for a simple harmonic structure, which seems to provide support. This fits the concept of the technique Vocal Holding, in which the music therapist is vocally following, supporting, and mirroring the patient while accompanying with only two piano chords, to create an atmosphere that is safe and predictable during improvising (Monti & Austin, 2018). Besides, music therapy literature indicates that "lullabies" with characteristics very similar to the mantra-based interventions, also generate stress-reducing effects, which is attributed to the simplicity of the musical components, such as a slow and steady tempo, a repetitive and simple rhythm, and just a few different notes (Friedman et al., 2010; Standley, 2003).

The importance of implementing a solid structure is also emphasized when reviewing the interventions outside of the music, which are focused on establishing a clear, predictable and above all a safe framework for the patient, giving the patient the support and confidence to play, experiment and express themselves within the offered framework.

Strong similarities can be seen between the possible change factors found in this study and the general characteristics that can positively influence the quality of care or treatment of people with MID. Literature shows that psychological treatment for people with MID may benefit from a clear structure and predictability (Došen, 2007; Elias et al., 2009; ten Wolde et al., 2006). Creating favorable environmental conditions, such as a predictable course of events in daily life or a clear structure of the therapeutic session based on small feasible steps, can enhance feelings of safeness and reduce stress in people with MID, which will benefit treatment (Didden, 2007). These important factors, which can be seen as treatment guidelines for adults with MID, are in line with the possible change factors found in this study, such as the creation of a predictable musical structure.

4.6 Strengths and Limitations of the Present Study

In order to show international practice-based knowledge, the current study was conducted in three different countries, since results related to the specific context of one country may not be relevant to other countries. Firstly, international definitions of MID and subsequent clinical care for adults with MID differs between countries (Moonen, 2017). Secondly, music therapy education differs among countries with regard to music therapy methods and general

theories. As only three countries were involved, each with close proximity to each other, results are unlikely to be generalizable. Replication of this study across different continents is strongly recommended.

With respect to some of the other results, more research is needed to be able to generalize findings, especially regarding change factors. The initial research questions focused on how and in what way music therapists could lower stress levels of patients with MID instead of exploring *why* interventions can lead to stress reduction. Data-analysis revealed some of these possible change factors, and we decided to report them (see Table 1). In future research, we suggest including research questions specifically addressing "change factors" or "mechanisms of change," because this may yield important knowledge on both the development and the evaluation of music therapy interventions for stress reduction in adults with MID.

Several techniques were applied to enhance the quality of the present study. The use of an interview guide, a codebook, the iterative process and the 'constant comparison' method during data analysis can be seen as a strength of this study, because it helps to ensure that the results stay as close to the original data as possible. We found this particularly important because this is the first study in which music therapists in clinical practice were interviewed about their music therapy interventions to reduce stress in patients with MID.

4.7 Implications for Clinical Practice

The findings of this study contribute to both the body of practice-based knowledge on which specific interventions can be used by music therapists to reduce stress in patients with MID, and to the knowledge base of experiential interventions for people with MID in addition to the cognitive-oriented approaches, such as "cognitive behavioral therapy" [CBT].

Firstly, results show two main ways to reach stress reduction. Although both ways are intended to lead to the same result (stress reduction), the order of intervention steps and/or the way in which the music therapist intervenes on the basis of musical components, may be different. Therefore, it is important that music therapists, prior to the intervention, have a clear understanding of the needs of their patient and the most fitting goal (stress release or direct relaxation), so that the most suitable interventions can be applied.

Secondly, the present study shows that improvisation is the most commonly used intervention by music therapists to reduce stress in adults with MID (see Table 1 for an overview of the different types of the mentioned improvisation-based interventions). When using improvisation-based interventions to reduce stress in patients with MID, according to the present study, there are some key elements that can positively influence the effect of the intervention. Namely, the use of simple musical structures, which means that the improvisation has a repetitive and predictable character. Also, synchronizing with the patient during improvising, whether as a stand-alone goal or as a step-in working on the goals "stress release" or "direct relaxation," helps the music therapist to attune to the patient's needs from moment-to-moment. This may enhance feelings of safety, which is especially important for

the clinical treatment of adults with MID (Didden, 2007) and can therefore increase the effect of improvisation.

Thirdly we believe that the results of the present study may provide a useful basis for the further development of more explicit music therapy intervention descriptions for stress reduction in patients with MID. According to the previous work of Robb et al. (2011), intervention descriptions of music therapy should be tailored to the target population, and realistic therapy goals as well as desired outcomes should be defined. Moreover, Hanson-Abromeit (2015) adds the importance of defining the purpose and intention of each musical element in the descriptions of music therapy interventions, such as the specific use of musical techniques within musical improvisation.

Fourthly, with the findings of the current study, we kindly recommend that other health care professionals responsible for referral to therapy (e.g., psychologists, physicians, health care coordinators) will consider experience-based interventions, such as music therapy, since both literature and results of the present study show that they are well suited to the needs and capabilities of adults with MID (Didden et al., 2016; de Witte et al, 2017). Indeed, from neurological perspectives, music is intrinsically motivating, drives motor function and elicits emotional responses (Koelsch et al., 2016; Thaut & Hoemberg, 2014). Moreover, according to the music therapists of the present study, it is the structure of music which provides a sense of safety, which benefits treatment in adults with MID. However, although psychologists often recognize that the use of music in daily life helps to manage stress, they still appreciate additional knowledge about why and when a music therapist should be involved in the treatment of their patients (Aalbers et al., in press; Magee, 2019). Our findings not only provide more insight into the different types of music therapy interventions for stress reduction, but also into the related goals, techniques, and change factors. This helps to understand when, why, and how music therapy can be effective for stress reduction, which reinforces referral to music therapy on the basis of substantive grounds. In addition, the results of the present study demonstrate the need for a clear and thorough assessment of patient with MID before any therapeutic intervention should be initiated.

4.8 Recommendations for Future Research

The findings of this study may help to implement music therapy interventions for stress reduction in adults with MID, but more research is needed to assess the effectiveness and applicability of these interventions. It would be relevant to replicate this and future studies in other parts of the world. In future research it is recommended to add research questions on therapeutic change factors, which may help to explain *why* the interventions used possibly lead to stress reduction. We also strongly recommend a study employing a systematic review methodology – such as a meta-synthesis – to analyze music therapy literature regarding possible change factors, as the number of empirical studies has increased in recent years. Finally, we would welcome the development of standardized intervention descriptions, like music therapy protocols, to enhance treatment fidelity, enabling more robust research on

the effects of music therapy. Detailed intervention descriptions within research are essential for replication and translation of music therapy interventions to clinical practice (Hoffmann et al., 2014; Robb et al., 2011; Stouffer et al., 2007).

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CHAPTER 5

The Validity and Reliability of Self-Report Stress Measures and their Suitability for Assessing Stress in Adults with Mild Intellectual Disabilities

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Abstract

Around the world, stress is one of the major factors that negatively impacts the development of psychopathology and leads to severe physical conditions. Timely recognition of stress-related problems is therefore becoming increasingly important, especially in client populations that appear to be more vulnerable to stress, such as people with mild intellectual disabilities (MID). Recent research conducted on the use of physiological measures to assess people's stress levels shows that, in addition to these measures, self-report measures are necessary to gain a correct understanding of an individual's perception of stress. However, there is no overview of self-report stress measures that focus on experiencing stress in the present moment or in daily life. In order to provide an overview of the existing self-report stress measures, a scoping review was conducted. In addition, to advise clinical professionals on the correct use of the identified self-report measures of stress in people with MID, an expert consultation was held to refine the preliminary findings. Results showed a total of 13 self-reported stress measures that met the final inclusion criteria, of which three were developed specifically for assessing stress in adults with MID (GAS-ID, LI, and SAS-ID). For each included self-report stress measure, the psychometric quality, assessment procedures, and suitability for adults with MID were reported, supplemented by the findings from the expert consultation. Implications for clinical practice on the use of self-report stress measures, particularly in people with MID, are discussed, as well as recommendations for future research.

Keywords: stress, state anxiety, self-report measures, stress assessment, stress scale, scoping review, mild intellectual disabilities, borderline intellectual functioning

1. Introduction

Recognizing a person's stress-related problems is becoming increasingly important, as stress is believed to be one of the major factors negatively affecting health and well-being. High stress levels are regarded as an important risk factor for the onset and progression of a wide range of physical and emotional problems, such as cardiovascular diseases, cancer, anxiety disorders, depression, and burnout (American Psychological Association [APA], 2017; Australian Psychological Society [APS], 2015; Steptoe & Kivimäki, 2012). Nevertheless, the literature reports that it is difficult for many people to both understand the destructive impact of daily life stress experiences (Casey, 2017; de Witte, Spruit, et al., 2020) and to reduce or cope with stress without any professional support (World Health Organization [WHO], 2010). This is especially the case for adults with mild intellectual disabilities (MID), as they experience stress more frequently in daily life than people without intellectual disabilities (Emerson, 2003; Hatton & Emerson, 2004; Schuengel & Janssen, 2006; World Health Organization [WHO], 2010). In addition, people with MID have also been found to have fewer resources to cope with daily life stress experiences (Hartley et al., 2009a; Lunsky & Benson, 2001; Scott & Havercamp, 2014).

1.1 Measuring the Concept of Stress

When we use the term "stress" in the present study, we are referring to negative stress experiences, defined by Aldwin (2007) as the quality of an experience produced by a person-environment transaction that, through either overarousal or underarousal, results in psychological or physiological distress (Aldwin, 2007; Riley & Park, 2015). Responses to stress are related to physiological arousal and emotional states, and the underlying systems of both these responses regulate and affect each other in times of stress (Linnemann et al., 2017; McEwen & Gianaros, 2010; de Witte, Spruit, et al., 2020). The physiological response to stress implies the activation of the hypothalamic-pituitary adrenal (HPA) axis and, due to the release of adrenalin and noradrenalin, increased activity of the sympathetic nervous system. This in turn results in increased *physiological arousal*, such as heart rate (HR), blood pressure, and cardiac output (Bally et al., 2003; Pfaff et al., 2007). Stress-related *emotional states* can be defined in terms of subjective worry, nervousness, and restlessness (Akin & Iskender, 2011; Cohen et al., 1983; Pittman & Kridli, 2011; Pritchard, 2009), and has many similarities with "state anxiety" as an outcome. Accordingly, many researchers describe state anxiety as an emotional response to an individual's perception of a stressful experience (e.g., Hook et al., 2008; Koelsch, Fuermetz, et al., 2011). In this review, we therefore regard state anxiety as a stress-related outcome. Stress-related outcomes can be measured by means of biomarkers related to physiological arousal (physiological measures) and/or by measuring people's emotional states related to stress experiences (psychological measures). Previous empirical studies confirm this and show that stress-related outcomes have been measured

with physiological and /or with psychological measurement methods (proxy or self-reports; Kim et al., 2018).

Although there is a large body of knowledge concerning the immediate effects of stress on physiological arousal, as indicated by several biomarkers like HR, blood pressure, heart rate variability (HRV) and hormone levels (Chandola et al., 2010; Föhr et al., 2017; Kim et al., 2018), an increased physiological arousal does not automatically refer to the perception of stress. It can also mean, for example, that a person is positively excited or deeply focused (Csikszentmihalyi, 2000; Pfaff et al., 2007; Rheinberg & Engeser, 2018). When examining the degree of stress, many researchers therefore emphasize the importance of measuring stress outcomes related to both physiological arousal as well as to people's emotional states.

Both proxy-reported and self-reported information is used to examine psychological stress-related outcomes, such as people's emotional states (Crawford et al., 2006). Proxy reports refer to information about an individual given by others, like relatives or caretakers. These are often used as an alternative when obtaining self-reported information is not a viable option, for instance when the respondent is not able to communicate verbally (Emerson et al., 2013; Miller & Tucker, 1993; Moore, 1988). Evidence suggests that proxy reports may be less accurate and less sensitive, compared to self-reported information (Scott & Havercamp, 2018; Moss et al., 1996). Self-report measures originally refer to data obtained by questionnaires or interviews in which respondents are asked to report about their personal experiences, values, feelings or thoughts, related to certain contexts and/or circumstances (Chan, 2009). Self-report data are commonly collected on a wide variety of topics in both medical and psychological research on topics such as pain, emotions, and personal preferences (Scott & Havercamp, 2018; Gerald & George, 2010).

1.2 Perceived Stress in Adults with Mild Intellectual Disabilities

MID is a neurodevelopmental disability characterized by deficits in intellectual and adaptive functioning skills (American Psychiatric Association [APA], 2013). In the Netherlands, the term MID mostly refers to people with limited intellectual and adaptive skills with IQ scores of 50-85, thus including people otherwise referred to as "people with borderline intelligence" (Kaal et al., 2015; Wieland & Zitman, 2016). As in the general population, stress experienced by adults with MID is linked to many negative mental health outcomes (Hartley et al., 2009a, 2009b; Hulbert-Williams & Hastings, 2008; Scott & Havercamp, 2014). Persistent stress in adults with MID can lead to maladaptive coping strategies and serious health issues like depression (Hartley et al., 2009a, 2009b), impaired cognitive functions (Heyman & Hauser-Cram, 2015), physical health problems (Lunsky, 2008), and substance abuse (Didden et al., 2009).

Adults with MID experience more stress in daily life than people without intellectual disabilities (Bramston & Mioche, 2001; Casey, 2017; Emerson, 2003; Hatton & Emerson, 2004; Schuengel & Janssen, 2006; de Witte, Spruit, et al., 2020; World Health Organization [WHO], 2010). Moreover, they have been shown to have more difficulties coping with their daily

stress than adults without intellectual disabilities (APA, 2013), which fits with the theory of Cohen et al. (1983) who defined psychological stress as the extent to which persons perceive that demands exceed their ability to cope. Various explanations have been proposed for this increased risk of stress, including experienced difficulties in social interactions, which appears to be one of the main stressors in their daily lives. This in turn can be explained by their lack of control over minor daily and major life decisions (Dulin et al., 2013; Hartley et al., 2009b; Scott & Havercamp, 2014). Furthermore, adults with MID often seem to lack social support and self-efficacy, important factors for coping with stress (Abbaszadeh & Sardoie, 2016; Everly & Lating, 2019; Seyed et al., 2017).

1.3 Psychological Stress Measures for Adults with MID

Research suggests that self-reported information may be more accurate and sensitive compared to proxy measures, so researchers in the field of MID also prefer self-reporting measures above proxy measures to better understand how and to which degree adults with MID experience stress (Lindsay & Skene, 2007; Scott & Havercamp, 2018). The value of proxy instruments is perceived to be limited as a proxy can never reliably report on another person's internal mental state (Emerson et al., 2013; Scott & Havercamp, 2018). This is in line with results of studies which show that, when compared to self-reported outcomes, proxies tend to overestimate impairment and underestimate health-related quality of life of people with (M)ID (Andresen et al., 2001; Vlot-van Anrooij et al., 2018). However, Scott and Havercamp (2018) found that ratings on stress by family members did significantly correlate with self-report ratings, whilst staff ratings did not. A plausible explanation provided by the researchers is that participants for whom family members completed the survey were likely to be living with their family, perhaps giving family members better insights into the day to day stressors impacting these adults with MID.

In general, there is a need for high quality self-report measures on stress in adults with MID (Glenn et al., 2003; Kooijmans et al., submitted for publication; Sams et al., 2006). Gaining an understanding of someone's personal thoughts, attitudes and feelings can lead to an enriched knowledge base from which opinions can be formed and interventions for stress reduction implemented (O'Keeffe et al., 2019). However, there are many challenges when collecting self-reported data from people with MID associated with the nature of the disability, including problems with reasoning, verbal expression, reading, abstract thinking, and judgment (APA, 2013; Schalock et al., 2010). To accommodate for these challenges, adaptations have to be made to 'standard' instrument language, lay-out, and assessment procedures. However, only a few self-report measures are available that incorporate these adaptations to better suit individuals with intellectual disabilities (Lindsay & Skene, 2007; Scott & Havercamp, 2018). Wieland et al. (2012) have identified a number of self-report measurement instruments developed for use in the general population which are suitable for adults with MID.

1.4 Purpose of the Present Study

As persistent stress can lead to the development of psychopathology and severe physical conditions, it is becoming increasingly important to recognize stress-related symptoms in populations known to be more vulnerable to stress, like people with MID. It is therefore critical to gain more insights into the way stress can be assessed in this group. In recent years, more and more research has been conducted on the use of physiological measures to assess people's stress levels. Although this has added substantial value to stress research, it is no substitute for the importance of self-report measures, since the individual's perception of stress is directly related to the emotional states an individual is in. And as stated before, physiological and emotional stress are not necessarily directly related (e.g., de Witte, Spruit, et al., 2020; Linnemann et al., 2017; Scott & Havercamp, 2018). In order to provide an overview of the existing self-report stress measures and to provide more information about their suitability for adults with MID, we conducted a scoping review. Moreover, in order to advise clinical professionals on how to correctly use the identified self-report stress measures, expert consultations were held to refine our preliminary findings. Our findings can be applied to research in which stress-related outcomes are measured in both adults with MID as well as those without intellectual disabilities. Results of this scoping review will provide guidance to clinical practitioners to assess perceived stress in adults with MID.

2. Method

In order to provide an overview of existing stress self-report measures, we performed a scoping review. A scoping review follows a *systematic approach* to map evidence or to bundle scientific findings on a topic to identify concepts, theories, sources, and knowledge gaps (Arksey & O'Malley, 2005; Munn et al., 2018; Tricco et al., 2018). This approach matches our research questions, which aim to provide more insights into the different types of self-report measurements and their characteristics, and how they can be used in adults with MID. Scoping review methods differ from systematic reviews which are used to address more specific, and mostly clearly predefined questions, based on particular criteria of interest (Peters et al., 2015). Scoping reviews can be seen as a hypothesis-generating exercise, while systematic reviews often focus on hypothesis-testing (Tricco et al., 2015; 2018). Results of a scoping review thus provide in-depth information for further orientation, define preliminary working hypotheses, set research agendas, and identify implications for decision-making (von Elm et al., 2019; Munn et al., 2018; Tricco et al., 2016).

2.1 Search and Selection Process

2.1.1 Search Terms and Sources

Multiple systematic searches were performed with the help of a medical librarian, as previous research showed that librarian engagement is significantly associated with higher quality

of reported search strategies (Rethlefsen et al., 2015). We conducted a computer-based search of the psychological and medical electronic literature databases, including Medline, Academic Search Complete, CINAHL, Cochrane Library, Web of Science, Wiley Online Library, SpringerLink, PiCarta, Academic Search Premier, ScienceDirect, PsycINFO and Google Scholar. Appropriate key words were identified through exploring the literature on "stress assessment", "stress questionnaires", and "stress measures".

Many previous studies have examined the relationship between state anxiety outcomes and physiological stress-related outcomes (e.g., Hook et al, 2008; Koelsch, Fuermetz, et al., 2011; de Witte, Pinho et al., 2020; de Witte, Spruit, et al., 2020) and defined state anxiety as a stress-related emotional state (Lazarus, 1966; Meijer, 2001; Yang et al., 2011; de Witte, Pinho et al., 2020; de Witte, Spruit, et al., 2020). We have therefore included state anxiety as a stress-related outcome in our current study. In addition, we note that in the literature, the concepts of stress and state anxiety are used interchangeably (Bradt & Dileo, 2014; Lazarus & Folkman, 1984; Ozer et al., 2013; Pittman & Kridli, 2011; Wetsch et al., 2009).

We then combined multiple search terms related to *stress* or *state anxiety* with terms referring to *psychological testing*. An exemplary search string used for the PsycINFO database can be requested from the first author. Searches were limited to publication dates from 1980 to April 2020. This time frame is consistent with the consensus within the literature that research concerning psychological measures of stress and / or state anxiety commenced in the 1980s (e.g., Cohen et al., 1983; Spielberger et al., 1983). In addition to the online databases, forward and backward searches were conducted by screening the reference lists of included studies, visiting a university testing library, and consulting research experts for "grey" literature. The initial search resulted in the screening of a total of 3451 studies and an additional 20 measures from forward and backward searches.

2.1.2 Selection of the Self-Report Stress Measures

To identify the self-report stress measures that fit the aims of the present study, we applied several selection criteria in two different selection steps. The first step concerned the screening of the studies found. Titles and abstracts of all the English-language peer-reviewed studies were screened for relevance, which means they had to include the terms "stress" or "state anxiety" related to psychological measures. Psychological measures that did not purely target *general* stress or state anxiety or stress in daily life were excluded, such as measures specifically assessing work stress, long term stress, parenting stress, or stress within the context of a specific medical diagnosis. At this stage, studies were also included in cases where the abstracts did not explicitly state whether the scale used was specifically a self-report stress measure, or whether the outcome measure concerned stress or state-anxiety in general or in daily life. Studies on self-report stress measures in non-English languages were excluded. This selection step ultimately resulted in 26 self-report measures assessing stress or state anxiety in adults. This reduced the number of studies to 75, which were then full-text screened by

at least one author. Table A4 (Appendix 4, page 261) contains the complete overview of the self-report stress measures that resulted from this step one selection.

The second selection step concerned the final inclusion of the self-report stress measures. Therefore, we applied the following criteria: instruments had to (1) be available for order in English, (2) have been applied in (clinical) outcome studies published in peer-reviewed scientific journals and, (3) instructions for assessment of the instrument are available. This selection step was performed by the first three authors (MdW, RK and MH) independently. Discrepancies were resolved through discussion. This resulted in consensus on the inclusion of 13 self-report measures for further analysis (see Figure 1: Flow Diagram of the Selection Process).

2.2 Evaluation of Included Self-Report Stress Measures

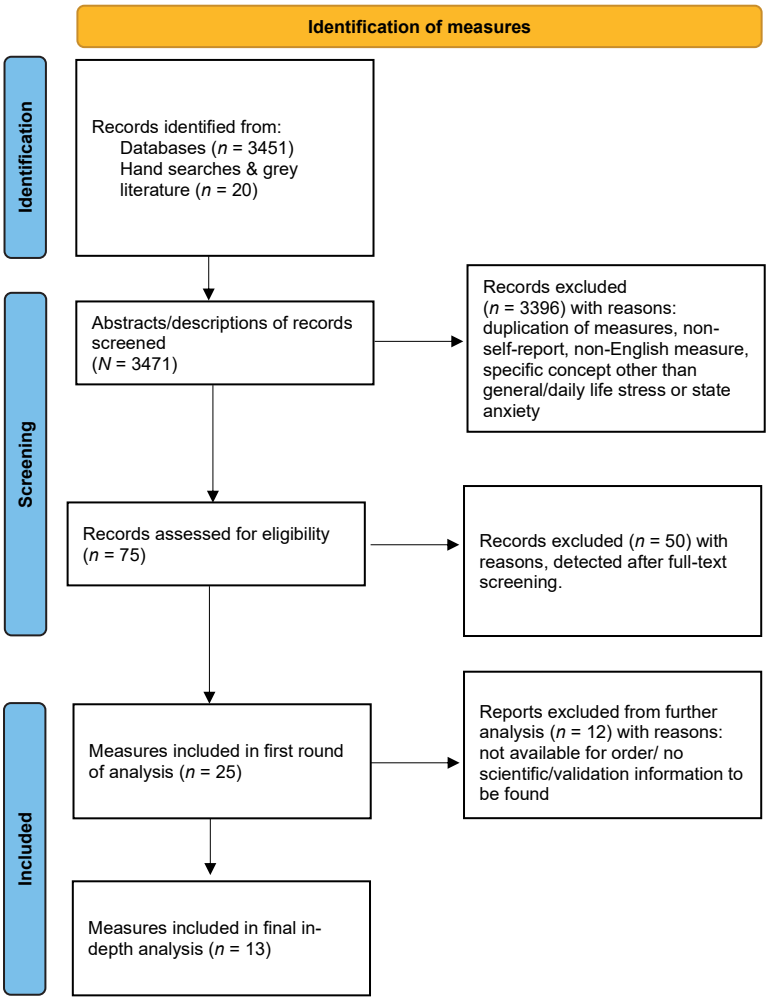
To provide insights into the characteristics and quality of the included self-report stress measures, criteria were formulated to describe their properties. Instrument characteristics relating to the criteria were found in the actual self-report stress measure itself, the user manual, validation studies, and other publications about the self-report measure in peer-reviewed and grey literature. The criteria applied to (1) the psychometric quality of the measure, (2) the assessment procedure of the self-report stress measure and, (3) the suitability for adults with MID. A further definition of the assessment criteria is presented below. Outcomes that relate to each criterion are presented in Table 1 in the Results section for all instruments.

2.2.1 Psychometric Quality

Reliability and validity are considered the main measurement properties of outcome measures used in clinical practice and research (Frost et al., 2007).

Reliability. A reliable measure is one that measures a construct consistently across time, individuals, and situations. When defining the psychometric quality of measures, three indicators of reliability are generally considered: test-retest reliability (stability over time), internal consistency (coherence of items with the concepts under study), and interrater reliability (equivalence across different researchers or assessors; Salmonds, 2008). Assessing test-retest reliability is typically done by computing Pearson's *r.* A Pearson's r of .70 or above indicates acceptable alternate-forms reliability (Chiang et al., 2015). For internal consistency, Cronbach's α is most often reported. An α greater than or equal to .70 is generally considered adequate, and a value of α greater than or equal to .80 is generally considered an indicator of good internal consistency (Allen et al., 2010; Chiang et al., 2015). Interrater reliability concerns the extent to which the different observers are consistent in their judgements. Interrater reliability is often reported as Cronbach's α. For each included self-report measure, we reported the published internal consistency coefficients (Cronbach's α). Manuals were investigated for clear instructions regarding the interpretation of test scores to support objectivity (Moosbrugger & Kelava, 2012).

Figure 1
Flow Diagram of the Selection Process



Validity. The term validity refers to the property of an instrument to measure exactly what it proposes. The main criteria and statistical tests for the assessment of validity are used to determine the content, criterion and construct validity of a measure (Frost et al., 2007). Content validity is evaluated to determine whether the instrument items were generated in accordance with relevant theory. To determine the content validity of the self-report measures, it is important that the self-report stress measure contains a clear description of the measuring construct; all terms related to the target group and outcome measure(s) have to be operationalized. We reported whether the self-report stress measure operationalized the key terms appropriately, such as a description of the characteristics of the type of stress

measured, and whether the distinction between stress exposition and stress reaction was described (Chiang et al., 2015; Harkness & Monroe, 2016). Moreover, to provide more insights in the validity of the included self-report measures, we also refer to independent validation research and / or assessments by test commissions. Criterion validity refers to the extent to which the measure agrees with an external standard measure. In the case of stress measurement, the outcomes of psychological self-report measures can be, for instance, compared to physiological measures related to stress responses.

Another relevant form of validity concerns *construct validity,* which refers to the extent to which scores on a measure correlate with the results of a different test. *Concurrent validity* is a form of construct validity that determines if the measure correlates highly with an established or widely used test already considered valid (the 'gold standard'). If there is a high correlation, this gives a good indication that the test measures what is intended. Alternatively, measures that should *not* be related, should demonstrate low correlations, therefore providing evidence for *discriminant validity* of the measure.

2.2.2. Assessment Procedure

In addition to its psychometric robustness, the suitability for a stress measure for practical and research purposes can be defined by a number of practical and procedural attributes of the instrument. These include the length of the assessment (determined by the number of the items and procedure), the presentation format (paper/pencil, digital, oral), the role of the assessor (group, guided or individual assessment), and the intended population. These attributes define the context and organizational prerequisites for administration and whether it should be stipulated in the manual.

2.3 Suitability for Adults with MID

2.3.1 Review of the Literature

One of the main purposes of this review concerned investigating the suitability of the measure for people with MID. After analyzing each stress self-report measure, we performed a literature search to see if any scientific evidence could be found on the use of the self-report measure in populations that included people with MID. The search was performed in Google Scholar. The following search string was used to guide the search: "learning disabilit*" OR "developmental" OR "mental retard*" OR "intellectual dis* AND [self-report measure]. If a reference was made regarding the suitability of the particular self-report measure in people with intellectual disabilities, learning disabilities, or developmental problems, we reported this.

2.3.2 Expert Consultation

As mentioned, adaptations to standard self-report instruments are generally needed to make them suitable for people with MID. As yet, no comprehensive guidance on how to make these adaptations is available (Kooijmans et al., submitted for publication). To be able to

provide more information, we consulted experts in the field of MID research and clinical practice. We used purposive sampling to select internationally renowned researchers in the field of intellectual disability research. The sampling frame was devised from a previously conducted systematic review (Kooijmans et al., submitted for publication). This sample was expanded by probing the authors' network and asking colleagues in the field of ID research to nominate researchers and clinicians they deemed experts on the topic. We then invited 40 experts from the United States, Europe and Australia to complete an online survey. Of these, 13 experts (33%) from four European countries completed the survey. Participants were academic and clinical staff from the United Kingdom, the Netherlands, Belgium and Germany with considerable experience in working and conducting research with people with (M)ID. See Table 1 for an overview of the characteristics of the participating experts.

Table 1
Demographic Characteristics of Experts Consulted

Total N = 13	*n* (%*)
Country of residence	
The Netherlands	6 (46%)
United Kingdom	5 (38%)
Germany	1 (8%)
Belgium	1 (8%)
Current employment	
Academic setting	9 (69%)
Clinical setting	2 (15%)
Joint academic / clinical	2 (15%)
Years of experience working with people with MID	
1-5	2 (15%)
6-10	4 (31%)
11-20	3 (23%)
20 +	4 (31%)

*percentages not adding up to 100% due to rounding differences

In the survey, the experts were asked to reply on open-ended questions on the subject of how to attune self-report measures to the needs and abilities of people with MID. They were asked to forward suggestions that address the content of self-report stress measures, such as language, response options and supportive media, and procedural issues, such as assessment procedures, questionnaire structure and instructions. Thematic analysis was applied to synthesize the results into general recommendations. The expert consultation on self-report stress measures was carried out within the context of a larger research project on self-report instruments for persons with (M)ID. For a more detailed description of the methodology applied, we refer to Kooijmans et al. (submitted for publication). As part of the assessment of the suitability of the included self-report stress measures for people with

MID, we compared the recommendations from the survey with the published information of the self-report stress measures.

3. Results

A total of 13 stress-related self-report measures met the final inclusion criteria. Nine of these explicitly focus on stress as an outcome and four on state anxiety as an outcome. Of the included self-report stress measures, the Glasgow Anxiety Scale for Intellectual Disabilities (GAS-ID), the Life Inventory (LI), and the Self-Rating Anxiety Scale for Intellectual Disabilities (SAS-ID) were specifically developed for assessing stress in adults with (mild) intellectual disabilities. First, we share our findings of the analysis of the self-report stress measures included purely from the perspective of the literature. We then discuss the findings of experts consulted, and present the integration of both types of data in Table 3.

3.1 Included Self-Report Stress Measures

The characteristics of each individual instrument are described for each of the three criteria: psychometric quality, assessment procedure, and suitability for people with MID based on the consultation of experts and the scientific literature. The findings are summarized in Table 2 and described in more detail below for each instrument (in alphabetical order).

3.1.1 Beck Anxiety Inventory (BAI)

The original publication of the BAI dates back to 1988 (Beck et al.) and it is still widely used today. It measures (state) anxiety symptoms and their level of intensity over the past week. It includes 21 items that target both somatic and more cognitive symptoms of state anxiety, for which respondents rate the intensity on a 4-point rating scale ranging from 'not all' to 'severely'. The total score is rated as minimal, mild, moderate or severe (state) anxiety.

Psychometric Quality. The BAI was found to have high internal consistency (average α coefficients across studies = 0.91; Bardhoshi et al., 2016) and adequate test-retest reliability (test–retest reliability = 0.65; Bhardoshi et al., 2016). It demonstrated both convergent validity with related measures of anxiety (other self-report instruments, diaries, clinical ratings; correlation coefficients ranging from 0.24 to 0.81; Bhardoshi et al., 2016) and moderate discriminant validity with other types of psychopathology (e.g. nonsignificant correlations with a measure of OCD symptomatology; Williams et al., 2013; moderate correlations with the Beck Depression Inventory; average *r* of 0.59 across studies; Bardhoshi et al., 2016). Both exploratory and confirmatory factor analytic studies generally support a two-factor structure in clinical populations. One factor represents cognitive symptoms of anxiety and the other represents somatic symptoms (Wilson et al. 1999).

Assessment Procedures. The BAI can be self-reported or interviewer-administered. Self-report generally takes a maximum of 10 minutes to complete. It can be administered in paper-and-pencil or interview format, but it is also available online.

Suitability for Adults with MID. The factor structure and other psychometric properties of the BAI were examined in a sample of people with MID (*N* =108; Mean IQ 67.1; Lindsay & Skene, 2007). To ensure that most people in the sample were able to meaningfully complete the BAI, some adaptations were made. The terminology of some of the items was simplified and the four-point response scale was presented in the form of four bar graph histograms of differing sizes. All questions were read aloud to all respondents by the assessor. On the basis of the analyses in their study, Lindsay and Skene (2007) asserted that people with MID appear to use the BAI reliably and consistently, and that the factors emerging from the sample were similar to those from mainstream populations.

3.1.2 Depression Anxiety Stress Scales (DASS)

The DASS (Lovibond & Lovibond, 1995a) measures three emotional states: depression, anxiety and stress. Three subscale scores for each of the emotional states are obtained that can be compared to norms and clinical cut-offs. For the purpose of this review, the properties of the Stress subscale were considered.

Psychometric Quality. High internal consistency coefficients are reported for each of the subscales of the 42-item and the 21-item versions (Cronbach's α of 0.90 to 0.95 for DASS-Stress; Parkitny & McAuley, 2010). Good evidence has been found for the construct validity through factor analyses (Crawford & Henry, 2003; Lovibond & Lovibond, 1995a) and convergent validity for the anxiety subscales of both the long and short versions of the DASS (correlation between DASS and BAI $r = 0.81$; Lovibond & Lovibond, 1995a), but the properties of the Stress subscale have been evaluated less extensively. Research in clinical populations has demonstrated responsiveness to treatment effects in, among others, psychiatric patients (Lovibond & Lovibond, 1995a; Ng, 2007) and persons with autistic spectrum disorders (Park et al., 2020).

Assessment Procedures. According to the manual (Lovibond & Lovibond, 1995b), completion takes 10 to 20 minutes for the 42-item version that comprises all three subscales. The shorter 21-item version of the DASS (DASS-21) takes 5 to 10 minutes to complete. A respondent indicates to what extent the statements applied to their lives over the past week on a 4-point scale. The DASS can be administered by paper-and-pencil or computer. The paper-and-pencil questionnaires and scoring forms are available at no cost from the developers' website. No specific training is needed to administer and score the DASS. Numerous officially endorsed translations of the DASS are available in many languages.

Suitability for Adults with MID. No empirical studies involving people with MID were found. Generally, people with MID were excluded from psychometric studies. The developers state that the DASS should not be presumed valid for some subpopulations,

including '[persons with] ...low literacy...' (Psychology Foundation of Australia [PFA], 2021). This effectively precludes many people with MID from using the DASS.

3.1.3 Derogatis Stress Profile (DSP)

The DSP is a self-report inventory rooted in interactional stress theory (Derogatis, 1987). Assessment of the DSP results in a detailed profile that identifies stressors on an environmental, personality, and emotional level, in interaction with each other. Cumulative scores provide a quantitative overall summary estimate (global stress score) of the respondent's current stress level.

Psychometric Quality. Strong support for the internal consistency (Cronbach's α > 0.80 for all 'stress domains'), reliability (test-retest coefficients > 0.72 for subscales and total scores) and validity of the DSP (by means of factor analyses) is provided in a small clinical sample and a larger non-clinical sample (Derogatis, 1987). A study on the correlation between several associated stress measures, including physiological correlates, yielded some support for the convergent and construct validity of the DSP (Dobkin et al., 1991).

***Assessment Procedures*.** Respondents are asked to rate 77 statements on a 5-point scale ranging from 'not-at-all true of me' to 'extremely true of me'. According to the information provided on the developer's website, 'the scale takes approximately 12 to 13 minutes to complete under normal conditions, although some individuals may require a few minutes longer.' (Derogatis Testing, 2021).

***Suitability for Adults with MID*.** No empirical studies addressing the suitability of the DSP for people with MID were found. The number of items and the complexity of the measure suggest that assessment may be a challenge for most people with MID (Bell et al., 2018; Hartley & McLean, 2006).

3.1.4 Glasgow Anxiety Scale for People with an Intellectual Disability (GAS-ID)

The GAS-ID (Mindham & Espie, 2003) was specifically developed for people with (M)ID to provide a reliable measure of state anxiety. It targets cognitive and emotional symptoms of state anxiety in the past week, as well as physiological symptoms that are assessed in the here and now.

Psychometric Quality. The GAS-ID showed sufficient methodological quality and excellent reliability (Cronbach's α = 0.96; test-retest r = 0.95) and validity results (ρ correlation coefficient of 0.75 with the BAI; ρ = 0.52 with pulse rate) as reported by the developers themselves (Mindham & Espie, 2003). However, only one external validation study was found (Hermans et al., 2013); the authors concluded that the GAS-ID can be regarded as a reliable self-report measure. High Cronbach's α's (>0.80) and test-retest ICC (0.89) were reported, and the GAS-ID showed satisfactory correlations with related measures (correlation with the HADS-A of r = 0.61).

***Assessment Procedures*.** No manual is available for the GAS-ID. The assessment time is reported to be 5–10 minutes (Mindham & Espie, 2003). The questionnaire is administered

as a structured interview. Respondents are asked to rate how often they experienced 27 expressions of fears, worries and physiological symptoms in the past week on a 3-point answer scale (from 'never' to 'always'). Furthermore, respondents are asked whether they experience any physiological symptoms associated with state anxiety in the here and now. Clinical cut-off scores are proposed by Mindham and Espie (2003), but they state that more research is needed.

Suitability for Adults with MID. The GAS-ID is designed specifically to be administered to people with MID. In the process of development, several alternative versions were tested for optimum suitability for people with MID. The resulting measure is perceived by the authors as being suitable for use with those people with MID who demonstrate sufficient ability to communicate verbally in day-to-day interactions (Mindham & Espie, 2003). The GAS-ID is frequently used in research on stress and anxiety with people with MID (e.g. Hartley & MacLean, 2008), is referenced as a preferred diagnostic tool in clinical guidelines for people with MID (e.g. Davis et al., 2008), and is mentioned in several textbooks on diagnostics and treatment of people with (M)ID (e.g. Stavrakaki & Lunsky, 2007).

3.1.5 Index of Clinical Stress (ICS)

The ICS (Abell, 1991) is a self-report questionnaire for individuals older than 12 years. It measures the degree or magnitude of clients' perceptions of personal stress, which is defined by a '... perceived imbalance between the demands of daily living and a person's ability to respond.' The ICS is part of the Walmyr Assessment Scales (WAS), a compendium of more than 25 short-form measurement scales designed for use in assessing the severity or magnitude of a variety of personal and social problems (Walmyr Publishing, 2021).

Psychometric Quality. Psychometric evaluation studies were conducted by the developer or researchers affiliated to the WAS (Abell, 1991; Hudson et al., 1995). High Cronbach's α's of .96 (Abell, 1991) and .90 (Hudson et al., 1995) were reported. Evidence for convergent validity was demonstrated by means of significant correlations with associated constructs (mean *r* =.48) and nonsignificant correlations with discriminant factors (mean *r* =.08).

Assessment procedures. The respondent is required to respond to the 25 items on the test form by selecting one response from a 7-point scale ranging from 'none of the time' to 'all of the time'. The respondent is expected to fill in the questionnaire unassisted. The WAS manual details no administration times, but is reported to be 'rapid'. The ICS is available in paper-and-pencil form and can be administered digitally through the publisher's own digital administration application.

Suitability for Adults with MID. The manual states that those completing the questionnaire must be literate and have no severe cognitive impairment. Readability statistics for the measure are given. The Flesch-Kincaid Grade Level of four suggests that a fourth level reading grade is required to complete the form autonomously. As the majority of people with MID are unable to read beyond grade three level (Conners, 2003), autonomous completion

of the ICS may be challenging for many. However, the ICS was developed for individuals from the age of 12 years upwards, meaning that the level of understanding may be appropriate for some people with MID.

3.1.6 Lifestress Inventory (LI)

The LI (Fogarty et al., 1997) is a self-report questionnaire designed to measure frequency and impact of stressors in daily life. It was developed specifically for people with MID as an update of the Subjective Stress Scale (SSS) that is no longer available.

***Psychometric Quality*.** In three studies, none of which were conducted by independent authors, the psychometric quality was found to be sufficient (Bramston et al., 1999; Fogarty et al., 1997; Lunsky & Bramston, 2006). For internal consistency, Lunsky and Bramston (2006) found Cronbach's α to equal .80. In the same study, some evidence was presented for the convergent validity of the LI, by showing significant correlations with related measures (r =.64 to .78). Modest correlations were presented between self-report and informant measures (r =.34 to .70). According to Fogarty et al. (1997), confirmatory factor analysis indicated three underlying factors that impact the experience of stress in daily life. These factors were labeled General Worry, Negative Interpersonal Relations, and Coping.

Assessment procedures. According to the scoring instructions / manual provided by the authors (Bramston & Fogarty, n.d.), the 30 items of the LI are intended to be read aloud. A series of buckets from empty to full can be used as a visual representation for the response options to facilitate understanding. Other possibilities to ensure that an item is understood correctly include repeating or re-wording a question, as well as asking the respondent to elaborate on their answer to make sure they interpreted the question correctly. As an extra response option, "0" indicates that an item/event was not experienced by the respondent; this option helps establish a frequency score. The other response options–from 1 ("no stress") to 4 ("a great deal of stress")–indicate the impact of single stressors. Assessment is preferably completed by an experienced psychologist.

***Suitability for Adults with MID*.** The LI has been specifically developed for people with MID and research into validation has been, as quoted above, carried out with people with MID. Notably, the LI was developed by means of focus groups with people with MID and staff members, and was designed to be easily understood and completed by people with MID (Scott & Havercamp, 2018).

3.1.7 Perceived Stress Scale (PSS)

The PSS (Cohen et al., 1983) has become one of the most widely used psychological instruments to measure the degree to which situations in people's lives are appraised as stressful. Cohen et al. (1983) define psychological stress as the extent to which a person perceives that demands exceed his/her ability to cope.

***Psychometric Quality*.** Although scores on the 14-item PSS exhibit good reliability estimates across the literature, four of the items tend to perform poorly when evaluated

using exploratory factor analysis (Cohen & Williamson, 1988; Lee, 2012). As a result, the PSS is commonly implemented using the 10-item form. In the review of Lee (2012) on the psychometric qualities of the PSS, it is shown that all included studies (N = 19) reported α coefficients of >.70. The test-retest reliability of the PSS-10 was assessed in four studies, and met the criterion of >.70 in all cases. The PSS correlated significantly and predictably with a range of other measures of stress and pathology (correlations typically in the 0.30–0.70 range), such as the Job Responsibilities Scale, HADS and STAI. Additionally, higher PSS scores have been shown to be associated with higher levels of cortisol; a biological indicator of stress (van Eck & Nicolson, 1994).

***Assessment Procedures*.** The PSS is available in a 14 and 10-item form and the average completion time is 5-10 minutes. Items are designed to tap how unpredictable, uncontrollable, and overloaded respondents generally find their lives. The scale also includes a number of direct queries about current levels of experienced stress.

***Suitability for Adults with MID*.** The PSS is designed for use in community samples for those with at least a junior high school education. Although there is no information available on the use of the PSS in adults with MID, some research has been carried out with younger college students with disabilities, such as learning disabilities, ADHD, and autism spectrum disorders (Janusis & Weyandt, 2010). The students with disabilities tended to score higher on the PSS, but the differences did not approach significance. No separate norms for people with disabilities were constructed on the basis of this study.

3.1.8 Perceived Stress Questionnaire (PSQ)

The PSQ (Levenstein et al., 1993) measures the experience or perception of stress by the individual during stressful situations, and is considered valid in the context of a transactional model of stress (Kocalevent et al., 2007). The PSQ was developed for use within the field of clinical psychosomatic research (Levenstein et al., 1993, 2000). There are two forms of the PSQ: the "general" (the last two years) and the "recent" (during the last four weeks) form.

Psychometric Quality. The original authors developed the instrument in English and Italian and validated it among 230 subjects (Levenstein et al., 1993). Internal consistency of the original English version (measured by Cronbach's α) ranges from 0.80 to 0.86 (Kocalevent et al., 2007; Levenstein et al., 1993), and research on test-retest reliability (Pearson correlation coefficients r between 0.80 and 0.86; Levenstein et al., 1993; Sanz-Carillo et al., 2002). The PSQ shows positive associations with compatible self-report measures such as Cohen's (1983) Perceived Stress Scale (PSS; r = 0.73; Levenstein et al., 1993). Notably, there are some indications that PSQ scores seem to differ between populations of patients and healthy individuals, and that scores seem to be sensitive to change after treatment (Fliege et al., 2005).

Assessment procedures. The PSQ has 30 items. Response options and items of both the PSQ-General (past 1 to 2 years) and the PSQ-recent (past month) are identical. Respondents are asked to estimate how often they deal with stress-related experiences on a 4-point Likert

scale. While no extensive manual has been published, free scoring instructions are available to researchers. The administration time is expected to be 5 minutes. Translations along with validation studies are available in Swedish (Rönnlund et al., 2015), Norwegian (Østerås et al., 2018), Spanish, Chinese and German. The instrument is available at no cost under a Creative Commons license.

Suitability for Adults with MID. No information on the suitability for people with MID has been found in previous empirical studies. The PSQ was originally intended for adults, but has also been successfully validated for adolescents aged 15-16 years (Østerås et al., 2018). Mutz and Müller (2016) used the PSQ to assess 14-year-old German upper secondary school pupils, without commenting on the application of the instrument to the target group. The adolescent research projects indicate that research about the usefulness of the instrument for (some) people with MID can be recommended.

3.1.9 Psychological Stress Measure (PSM-9)

The PSM was first published in 1988 (Lemyre & Tessier, 1988) and updated in 2003. The PSM-9 is an abridged nine-item version of the original 49-item assessment of self-reported state stress. Respondents are asked to rate stress symptoms they experienced in the past three to four days on an 8-point Likert scale (from 'not at all' to 'extremely'). The result is a single-factor indicator of perceived stress.

***Psychometric Quality*.** The authors report a wide range of reliability (Cronbach's α's > 0.90; test-retest *r*'s 0.68–0.80) and validity coefficients for the 49-item version in a series of publications by the developers of the instrument (Lemyre & Tessier, 1988; Lemyre et al., 1990; Lemyre & Tessier, 2003; Lemyre et al., 2009). The psychometric properties of the short PSM-9 version are reported to be 'the same as the original version' (Lemyre & Tessier, 2003), but only a Cronbach's α of 0.89 is reported for the PSM-9. No external validation studies have been published.

Assessment procedures. The PSM-9 appears to be a short single-page paper-and-pencil questionnaire. No scoring instructions could be retrieved. Only a French version of the manual was published (Lemyre et al., 1990), but it could not be retrieved by the reviewers.

***Suitability for Adults with MID*.** No evidence was found in previous empirical studies that the PSM-9 would be suitable for people with MID.

3.1.10 Self-Rating Anxiety Scale for Intellectual Disabilities (SAS-ID)

The SAS-ID is an adaptation of the Zung Self-Rating Anxiety Scale for persons with ID by Lindsay and Michie (1988). The SAS is a 20-item self-report assessment instrument for measuring state anxiety. Respondents are asked to indicate to what extent a series of statements apply to themselves within a period of one or two weeks prior to assessment. A total score reflects a general level of state anxiety as experienced by the respondent.

Psychometric Quality. Several researchers have assessed the psychometric quality of the SAS-ID (Lindsay et al., 1994; Masi et al., 2002; Ramirez & Lukenbill, 2008). Psychometric

evaluation was conducted by independent researchers and those affiliated to the original developers. Internal consistency coefficients (Cronbach's α) averaged a satisfactory 0.80. Convergent validity was established by finding significant correlations between the SAS-ID and related self-report instruments and diagnostic interviews (correlation coefficients ranging from 0.33 to 0.73).

Assessment procedures. The SAS-ID is a 20-item scale with a yes–no response format. It takes 5-10 minutes to complete. The SAS-ID is presented to respondents orally on an individual basis. Assessors are instructed to rephrase or reword the questions if the respondents appear to lack understanding.

***Suitability for Adults with MID*.** The SAS-ID is an adaptation of the original SAS intended for use in the general population. Adaptations are made to ensure that most people with MID are able to meaningfully complete the assessment with assistance. Adaptations made to the original are the use of a yes-no response format instead of a 4-point Likert-type scale and the rewording of items perceived to be difficult. The SAS-ID has occasionally been used in research involving people with MID (e.g. Carraro & Gobbi, 2012) and is mentioned in textbooks on diagnostics and treatment of persons with (M)ID (e.g. Hatton & Taylor, 2013; Vargas-Vargas et al., 2019).

3.1.11 State-Trait Anxiety Inventory, State Version (STAI-S)

The state version of the STAI (Spielberger, 1983) is one of the most long-standing and commonly used clinical self-rating scales to measure state-anxiety, which is defined as a temporal cross section in a person's emotional stream of life, consisting of subjective feelings of stress, tension, apprehension, nervousness, worry, and activation of the autonomic nervous system (Cattell & Scheier, 1961; Spielberger, 1983). In research, the 20-item STAI subscale is often used to measure state-anxiety before and after an intervention or task. Translated forms of the STAI are now available in more than 60 languages (Spielberger & Reheiser, 2009).

***Psychometric Quality*.** Many psychometric evaluation studies have been published which show that the STAI-S provides excellent psychometric properties: the internal consistency measured using Cronbach's α coefficient ranges from good to excellent (i.e. > 0.70) across several populations (e.g., Creamer et al., 1995; Fonseca Pedrero et al, 2012; Ortuno-Sierra et al., 2016; Spielberger, 1983). Noteworthy, α coefficients are typically higher for the STAI-S when state anxiety is assessed under conditions of psychological stress (Spielberger, 1983; Spielberger & Reheiser, 2009).

Assessment procedures. The STAI-S is a 20-item self-rating inventory which may be given either individually or to groups. The scale is composed of short verbal statements that participants have to rate using a 4-point Likert scale according to the subjective experienced intensity of each described feeling (1 = not at all, 4 = very much so). It is clear that the questionnaire's ease of administration, as well as the simple and straightforward scoring procedure have led many researchers to use this specific instrument (Rossi & Pourtois, 2012).

Suitability for Adults with MID. Although no studies have been published on the applicability of the STAI-S in persons with (M)ID, a STAI child-version (STAI-C) has been developed (Spielberger, 1973), especially constructed for 9 to 12-year old children. The STAI-C manual states that the scale may also be used with older children/adolescents who are below average in ability. In future research, the appropriateness of the STAI-C version for use in people with MID should be investigated.

3.1.12 Stress Arousal Checklist (SACL)

The SACL (Mackay et al., 1978) is a list of mood adjectives intended to measure stress experience as well as arousal. The authors refer back to work by Thayer (1967) and his factor analysis of the Activation-Deactivation Adjective List (AD-ACL). The two-dimensional structure of stress and arousal is explained as follows: "The stress dimension refers to the perceived favorability of the external environment, while arousal refers to ongoing autonomic and somatic activity" (Cox & Mackay, 1985).

***Psychometric Quality*.** In an independent factor analysis, the two-factor structure found by the original authors has been replicated (McCormick et al., 1985). This study also supports the two-dimensional model of stress and arousal operationalized in the SACL. Reliability was found to be relatively high in several studies (> 0.70), especially for the stress scale, while α coefficients showed more variance for the arousal scale (Watts et al., 1983). Evidence for the construct validity of the SACL was found in factor analyses (Fischer & Donatelli, 1987; Fischer et al., 1988; King et al., 1983). However, Hinton et al. (1991) stated that in their view, the stress scale of the SACL does not measure stress as defined by the authors and "is virtually identical to the state version of the STAI".

***Assessment Procedures*.** There does not seem to be a published manual, but the authors provide scoring instructions and note that "scoring keys are easily made" (Cox & Mackay, 1978, p.284). The 30-item list consists of positive and negative adjectives, for each of which the symbols "++", "+", "?" or "-" can be circled by respondents. Responses can be summed up separately for the 'stress' and 'arousal' subscales (Cox & Mackay, 1978, p. 284).

***Suitability for Adults with MID*.** No evidence was found in empirical studies that the SACL would be suitable for people with MID.

3.1.13 Stress Overload Scale (SOS)

The SOS (Amirkhan, 2012) is designed to measure "stress overload", a state described in stress theories as occurring when demands overwhelm resources. Respondents are asked to answer 30 questions and reflect on the occurrence of stress-related feelings and cognitions in the past week. Total scale scores and scores on two subscales–Personal Vulnerability and Event Load–are calculated. A short 10-item version (the SOS-S) is also available (Amirkhan, 2018).

***Psychometric Quality*.** All psychometric evaluation studies were conducted by the developers (Amirkhan, 2012; Amirkhan et al., 2015; Amirkhan, 2018). They report an excellent internal consistency of the SOS (with Cronbach's α's > 0.94 for both subscales and the measure

Table 2
Included Self-Report Stress Measures

Title (author/s, publication date)	**Outcome**	**Target group**	**Psychometric quality:** a) independent validation research available? b) number of available validating studies (approximately) c) internal validity (Cronbach's α)*	**Design**: a) number of items b) response options c) duration of administration	**Published information on applicability with people with MID available?**
Beck Anxiety Inventory (BAI), Beck et al. (1988).	State anxiety	Adults	a) yes b) >10 c) average across studies = 0.91	a) 21 items b) 4-point Likert scale c) 10 minutes max.	Yes Adaptation for MID by Lindsey & Skene (2006; 2009)
Depression Anxiety Scales (DASS) (Lovibond & Lovibond, 1995)	Stress + state anxiety	Adults	a) yes b) >20 c) α of 0.84 to 0.92 for DASS--Anxiety, and 0.90 to 0.95 for DASS-Stress	a) 21 short form, 42 regular form b) 4-point Likert scale c) 5-10 minutes short form, 10-20 minutes long form (all 3 subscales)	No Parkitny & McAuley (2010): "certain patient groups (eg ... the developmentally delayed...) may have difficulty understanding the questionnaire items or responding to them in an unbiased manner.
Derogatis Stress Profile (DSP) (Derogatis, 1980)	Stress	Adults	a) yes (only 1 study found) b) 2 c) α between 0.83 and 0.88 for different domains	a) 77 items b) 5-point Likert scale + VAS (0-100) for subjective stress experience c) 12-15 minutes	No
Glasgow Anxiety Scale (GAS-ID) (Mindham & Espie, 2003)	State anxiety	"People with an intellectual disability" (age/level of ID not specified)	a) yes (only 1!) b) 2 c) > 0.80	a) 27 b) 3-point Likert scale of frequency c) 5-10 minutes	Yes
Index of Clinical Stress (ICS) (Abell, 1991)	Stress	Adults & youths age 12+; Reading level > grade 4	a) no b) 2 (developer + affiliated researchers) c) 0.96 (Abel, 1991)/0.90 (Hudson,1997)	a) 25 b) 7-point Likert scale of frequency c) not specified, but stated as "rapid"'.	Yes Manual: "Persons who are only mildly impaired might be able to complete the WAS scales with considerable accuracy. The major things to watch for are the literacy skills, cognitive development, and ability to integrate affective responses with the item content and meaning of each of the scales." Flesch reading ease: 89 (6th grade level); Gunning Fog Index: 6 (sixth grade level); Flesch-Kincaid Grade Level: 4.

Title (author/s, publication date)	**Outcome**	**Target group**	**Psychometric quality:** a) independent validation research available? b) number of available validating studies (approximately) c) internal validity (Cronbach's α)*	**Design**: a) number of items b) response options c) duration of administration	**Published information on applicability with people with MID available?**
Lifestress Inventory (LI) (Bramston & Fogarty, 1997)	Stress	Age not specified; "suitable for administration to a wide range of people, including the mildly intellectually handicapped"	a) no b) 3 c) 0.80	a) 30 b) 4-point Likert scale + visual aid showing a series of buckets empty through to full can be used to improve understanding of the Likert-type options c) not specified	Yes
Psychological Stress Measure (PSM-9) (Lemyre & Tessier, 1988)	Stress	Adults	a) no b) 4 c) 0.89	a) 9 b) 8-point Likert scale c) not specified	No
Perceived Stress Questionnaire (PSQ) (Levenstein et al., 1993)	Stress	Adults	a) yes b) 6 c) ranging from 0.90 to 0.93	a) 30 regular, 20 short form b) 4-point Likert scale c) 5 minutes	No
Perceived Stress Scale (PSS) (Cohen et al., 1983)	Stress	Adults ("community samples with at least a junior high education"; "accessible to any subpopulation")	a) yes b) 19 or more c) above 0.70 across studies	a) 10 b) 5-point Likert scale c) 5-10 minutes	No
Stress Arousal Checklist (SACL) (Cox & Mackay, 1978)	Stress	Not specified	a) yes b) 6 or more c) for stress scale ranging from 0.81 to 0.86, lower for arousal scale	a) 30 b) 4-point Likert scale c) not specified	No

Title (author/s, publication date)	**Outcome**	**Target group**	**Psychometric quality:** a) independent validation research available? b) number of available validating studies (approximately) c) internal validity (Cronbach's α)*	**Design**: a) number of items b) response options c) duration of administration	**Published information on applicability with people with MID available?**
Self-Rating Anxiety Scale for adults with Intellectual Disabilities (SAS-ID). (Lindsay & Michie,1988) Adaptation of SAS (Zung, 1971)	State anxiety	People with an intellectual disability (age/level of ID not specified)	a) yes b) 3 or more c) average of 0.80 across studies	a) 20 b) yes-no answer format. c) 5-10 minutes	Yes Adaptations from the original instrument include yes–no response format, rewording of the items, and addition of supplementary items.
SOS Stress Overload Scale (Amirkhan, 2012)	Stress	Adults	a) no b) 3 c) 0.94	a) 30 or 10-item short form b) 5-point Likert scale c) not specified	No
STAI State-Trait Anxiety Inventory (Spielberger, 1981)	State anxiety	Adults (used in ID research)	a) yes b) lots of studies in many different languages c) ranges from good to excellent across several populations	a) 20 items for the State scale (Y1 form) b) 4-point Likert scale . c) approx. 10 minutes for "less educated or emotionally disturbed persons".	Yes: Manual specifies some instructions for the assessor in the case of guided assessment. Norms are based on a sample of 'working persons', which generally will include relatively few persons with MID. The STAI is frequently used in research with persons with MID in unaltered form.

Note. References and additional psychometric results are provided in detailed descriptions of the single measures in the result section.

as a whole). Test-retest coefficients averaged 0.75 over a one week period. Convergent validity has been demonstrated in significant correlations with other measures of stress (e.g. correlation coefficient *r* of 0.45 with the PSS-10) and criterion validity has been shown in the SOS's ability to predict illness following a stressful event. Psychometric properties for the original and short versions are all but identical.

***Assessment procedures*.** Participants are asked to rate feelings and cognitions related to life stress on a 5-point Likert scale (from 'not at all' to 'a lot'). No information on the duration of the assessment of the original or short forms has been published and no manual is available. Scoring instructions are attached to the form.

***Suitability for Adults with MID*.** The development and validation of the SOS made use of community samples. Some attention was paid to make sure that '... Only items that were consistently understood across [a] wide socioeconomic and ethnic spectrum were chosen for the SOS' (Amirkhan, 2012). However, its comprehensibility and general usefulness for people with MID has not yet been demonstrated.

3.2 Results of the Expert Consultation

The experts were asked to reply to open-ended questions on the subject of how to attune self-report measures to the needs and abilities of people with MID. They unequivocally indicated that the factors that improve appropriate use by people with MID *in general* also apply to the self-reported measurement of stress. Thematic analysis of the answers revealed six general recommendations relevant to the measurement of stress in people with MID.

The first recommendation was to use concrete and easy-to-understand vocabulary, simple grammar, and short sentences. The next was to use relatively short time frames for the retrieval of information. Assessors should not ask to retrieve information over longer periods than one week, as time processing abilities are generally impaired. A third recommendation relates to the use of Likert scales. When designing self-report measures for people with MID, the number of response options in Likert scales should be limited to three for people with moderate ID to MID and five to people with MID to borderline intellectual functioning. Fourth, an 'I don't know' option should be included in both forced-response and open-ended questions to prevent invalid answers from those who do not understand the question. A fifth recommendation was to use visualizations to support the meaning of questions and responses, although how exactly these should be configured was not specified. In regard to the assessment procedures, a sixth recommendation was to use pre-scripted alternative wording if the respondent seems unable to understand the question. Standardization ensures comparability of scores across assessments. The extent to which these factors were reflected in the self-report measures' design and assessment procedures differed across the included instruments. An overview of the suitability of each self-report stress measure for people with MID, according to the experts, is presented in Table 3.

Table 3
Findings of Experts Consultation compared to Self-Report Stress Measures found

	use of easy-to-understand language	max. 1-week time frame	max. 5 answer options*	"i don't know" answer option	use of visual support	scripted alternative wording
BAI	✓ (adaptation by Lindsay & Skene, 2007)	✓	✓	X	✓ (adaptation by Lindsay & Skene, 2007)	X
DASS	X	✓	✓	X	X	X
DSP	X		✓	X	X	X
GAS-ID	✓	✓	✓	X	X	X
ICS	✓	✓	X	X	X	X
LI	✓	/	✓	X	✓	X
PSM-9	/	✓	X	X	X	X
PSQ	/	X	✓	X	X	X
PSS	/	X	✓	X	X	X
SACL	/	✓	✓	X	X	X
SAS-ID	✓	✓	✓	X	X	X
SOS	/	✓	✓	X	X	X
STAI-S	/	✓	✓	X	X	X

***** This can both refer to a Likert-scale or yes/no answer option.
Note. ✓ = yes, according to literature; X = no, according to literature; / = not clear, according to literature

4. Discussion

The need to measure the degree of stress as accurately as possible in people with MID is reflected in both the literature reviewed and the information of the consulted experts. This can be seen as a response to the fact that people with MID are much more vulnerable to stress (Hatton & Emerson, 2004; Scott & Havercamp, 2014). Moreover, persistent stress experiences in people with MID may lead to more impaired information processing (Heyman & Hauser-Cram, 2015) which will adversely affect coping skills even more. Our study not only provides the first overview and analysis of self-report stress measures, but also provides more insights in how the self-report stress measures can be accurately applied in people with MID. In total, 13 self-report stress measures were found. Our findings show that three self-report stress measures appeared to be specifically designed for use with adults with (M)ID, that five did not appear to be suitable for populations other than the normally gifted population, and that based on the literature, five could potentially be appropriate.

4.1 Main Findings

Three of the 13 self-report stress measures address the needs and possibilities of adults with MID. The Lifestress Inventory (LI) was specifically designed for the MID population. Two others, the Glasgow Anxiety Scale for people with Intellectual Disability (GAS-ID) and the Self-rating Anxiety Scale for adults with Intellectual Disabilities (SAS-ID) generally reported that they could be used in people with ID, however their user manuals did not specify the exact intelligence range. As the items concerned mainly refer to insights, feelings and experiences from daily life, participants must be able to reproduce this in at least spoken language, suggesting that they are targeted towards adults with MID instead of the total ID population. Generally, these three self-report stress measures have in common that they use items that require a response on *simple Likert scales*, which could possibly be combined with *visual representations* of answer alternatives. This is in line with findings reported in previous studies as well as the expert consultations in our study, which show agreement that responses requiring a simple Likert rating scale or only yes/no can lead to appropriate responses from individuals with MID (Hartley & MacLean 2006; Heal & Sigelman 1995; Ramirez 2005). For those individuals in the lower range of MID, pictorial representations of response alternatives could increase the likelihood of gaining appropriate responses (Hartley & MacLean, 2006), which was echoed by the experts consulted, suggesting that visualizations to support worded response alternatives would be helpful.

Our findings also show that some of the other stress self-report measures seem to be more or less applicable to adults with MID. First, some evidence was provided in previous validity studies on populations in which participants with intellectual, learning, or developmental disabilities were also included. This applies specifically to the BAI (see Lindsay & Skene, 2007), the DASS (see PFA, 2021), and the PSS (see Janusis & Weyandt, 2010). Second, other self-report stress measures stated that they could also be used in younger aged populations, which may possibly refer to suitability for people with MID. This applies to the STAI-child version (9-12 years), the ICS (from 12 years), and the PSQ (from 14 years). Hurley (2008) suggests that the use of instruments designed for children may offer a useful basis for adaptation, because the measures contain concrete levels of vocabulary and simple sentence structures. This process has also been used by many other researchers (e.g. Guerin et al., 2009; Marshall & Willoughby-Booth, 2007). However, since these stress self-report measures have not been validated specifically for the adult MID population, we recommend thoroughly screening the measurement construct and assessment procedure before using them in clinical practice or in future research (Kooijmans et al., manuscript in preparation).

The findings from the expert consultations show the importance of adding an extra answer alternative such as “I don’t know” to prevent people with MID who do not understand the question from filling in a random answer. However, none of the self-report stress measures, even those specifically developed for people with MID, included this option. The Lifestress Inventory (LI) added the answer alternative “actually not experienced”, but this refers to the fact that the participant did not experience any stress at all. In addition, none

of the self-report measures included "alternative wording" to the questions and/or answer alternatives, as recommended by the expert panel. On the other hand, helping factors such as allowing assessment assistance (SAS-ID) or having someone else read the items (LI and GAS-ID) were not mentioned by any of the experts. Finally, response visualizations seem to be missing from both the GAS-ID and SAS-ID. This is remarkable, as this is considered one of the most important factors with regard to suitability for people with MID, both in the literature and by the experts consulted (e.g. Hartley & MacLean, 2006; Scott & Havercamp, 2018). Of the three self-report stress measures for people with MID, the LI appears to be most consistent with the findings of the experts. However, our findings show that in addition to consulting experts, screening the assessment procedures of existing self-report measures specifically adapted or designed for people with (M)ID is a worthwhile exercise.

4.2 The Added Value of Self-Reported Information

Although proxy reports are commonly used in MID, self-report measures prove to be more accurate and more sensitive, even in the MID population (Moss et al., 1996; Scott & Havercamp, 2018). Therefore, they are best for complementary use. Stimulating the use of self-reported information alongside proxy reports ensures that the voice of adults with MID is better heard in decision-making processes. However, studies comparing proxy and self-reported data have shown that data often vary when assessing subjective concepts, such as stress (Scott & Havercamp, 2018). An explanation for this difference may be that proxy reporters more often focus on the perceived disabilities as being particularly stressful, which is not reported by the participants with MID themselves (Lunsky & Bramston, 2006). Regarding the Lifestress Inventory (LI), relatively low correlations were found between caregiver reports and self-reports of stress. In addition, other studies show that people with MID tend to report more stress than their caregivers (Bramston et al., 1999; Lunsky & Bramston, 2006). However, three of the top five stressors reported by participants and proxies turned out to be the same.

The importance of self-reported information of people with MID, such as assessing subjective stress experiences, is also reflected in the increased recognition in our society that people with (M)ID are full citizens with the same rights as non-disabled persons, meaning that participation and social inclusion should dominate all organized activities (e.g., Devi, 2014; Giesbers et al., 2019). In other words, including the opinions, feelings, and thoughts of people with MID by using self-report measures, fits the call for knowledge democratization, as citizens increasingly demand their say in policies and research affecting them (Anderson, 2017; Dedding et al., 2020). This is important, because self-determination can be seen as an essential dimension of quality of life and is linked to many positive outcomes for people with (M)ID) (Frielink et al., 2018; Schalock & Verdugo, 2002; Wehmeyer, 2007). Therefore, both the findings of this review and the empirical evidence show that increasing our knowledge of self-report stress measures for people with MID is a highly recommended addition and in line with the contemporary opinion that the voice of people with MID should be included in matters that concern them.

4.3 Measuring the Concept of Stress

The way the concept of stress was operationalized by the self-report measures varied according to the theoretical underpinnings and constructs. Different paradigms or stress theories were used, such as the interactional stress model (e.g. the DSP or the PSS), theories on stress as a transitory anxiety state (e.g. the STAI and the BAI), and the tripartite model of anxiety and depression that describes stress as a common symptom for both (e.g. the DASS). Moreover, some of the self-report measures do not seem to have origins in a certain stress theory or model, but were developed empirically, involving expert consensus on the manifestation of stress in clinical practice (e.g. the GAS-ID). Others are based on the manifestation of stress symptoms described in classification systems of psychiatric disorders (e.g. the SAS-ID). In addition, a distinction can also be seen between self-report stress measures that focus mainly on stress as an experienced psychological and physiological state (e.g. the BAI, the SACL and the STAI) and those that focus on the experience of stress in the context of situations that actually or hypothetically cause stress, such as job related stress or stressful social situations (e.g., the LI, the PSS and the PSQ). To ensure that the concepts being studied are consistent with the design and intended use of the self-report measure, we recommend paying attention to how the concept of stress is theoretically framed when deciding to use a self-report stress measure (Cook et al., 2006).

4.4 Implications for Clinical Practice

There is a strong tendency in clinical practice to move away from attributing the symptoms of psychopathology solely to the cognitive deficits of people with MID; this is known as diagnostic overshadowing (Hagopian & Jennett, 2008; Reiss et al., 1982). Clinicians are becoming increasingly sensitive to the fact that people with MID can also suffer from symptoms of psychopathology. Since the degree of stress is now recognized as a significant factor in the development of severe psychopathology, especially in people with MID, it has become more important to correctly observe/assess stress-related states in clinical practice (Scott & Havercamp, 2014). This review therefore provides a practical basis for determining whether and which self-report stress measures are suitable for people with MID within their own clinical context.

To provide some guidance for clinical practice, we have formulated several recommendations based on our findings. First, with this review, we want to draw attention to the concept of stress and the importance for clinical practice to consider the degree of (daily or present) stress as a *crucial factor* in the quality of life and course of further psychological treatment in people with MID. In our view, stress assessment should be included as a regular part of the diagnostic phase of clients with MID when consulting clinical practice. Second, as mentioned, we strongly advise clinical practice to always strive to obtain self-reported information in addition to proxy-reports when it comes to medical, psychological, and service decisions involving people with MID. Third, we particularly recommend using the three self-report stress measures specifically designed for adults with (M)ID. These

self-report measures are characterized by simple Likert rating scales and/or items requiring yes/no responses. Specifically, the use of simpler wording, fewer response options, and the ability to provide supportive visualization are the main differences with the self-report stress measures developed for the non-ID population. Another significant difference is that self-report measures developed for (M)ID often allow the respondent to be assisted during the assessment (SAS-ID) and that the items can be read aloud by someone else (LI and GAS-ID).

While there is general consensus that it is necessary to timely assess stress in people with MID, we are also aware that this requires experiential knowledge of clinical professionals working with the MID population. The challenge for clinical practice is to prevent that difficult-to-understand behavior of people with MID too quickly leads to a psychiatric classification, which often has far-reaching consequences (Didden et al., 2016). On the other hand, psychological problems still have to be recognized timely. This requires continuous in-depth behavioral observations and careful consideration by clinical professionals, as people with MID, certainly in combination with additional behavioral/psychological problems, often are unable to clearly request help (ten Wolde et al., 2006). Decisions made should therefore be adequately aligned with personal and environmental circumstances, as well as with the level of cognitive functioning (Nouwens et al., 2020). Determining and applying suitable self-report measures for clients with MID could contribute to this purpose. Moreover, as indicated earlier, the use of self-report measures is also a way of letting the client's voice speak, and thereby enhances feelings of autonomy, initiative and freedom of choice. In this study, we have attempted to provide a first guide with regard to the use of self-report stress measures.

4.5 Limitations of the Present Study

There are some limitations of the present study that should be noted. First, because we strictly followed our inclusion criteria, we may have excluded some self-report measures which could be also suitable for assessing stress in people with MID (see Table A4). For example, they may have not yet been applied in (clinical) outcome studies published in peer-reviewed scientific journals. Another reason for exclusion was that measures were unavailable in the English language; it is possible that suitable measures exist in other languages.

Second, for the appraisal of the psychometric properties of each measure, we had to rely on the parameters reported by authors in their publications. Nearly all studies report Cronbach's alpha as the main indicator of reliability. Recent advances in psychometric research suggest that this may be a flawed indicator of the internal stability or reliability of a measure. It is stated that other indicators, such as *omega*, are more robust, and that reliability research should be preceded by Factor Analysis (Crutzen & Peters, 2017).

Third, we would have liked to share more specific information from the expert consultations. However, due to the use of an online survey, there was no opportunity to ask further questions. Therefore, for future research, we recommend adding a more interactive form of data collection when consulting experts on similar questions, such as a multidisciplinary focus group method. Another limitation concerning the expert consultations

is that the results reflect the participating experts' professional opinion. Although their clinical and research expertise are highly valued, the experts were not asked to substantiate their statements with references to empirical literature. Therefore, the suggestions by the experts must be valued as tentative and supplementary to the evidence from empirical studies. Finally, the experts' findings were only compared with the published information in the user manuals of the self-report measures, i.e. only with the information already described. An option for follow-up research would be to use a more detailed screening list and to screen the individual instruments with different researchers in the field of MID blinded from each other. This would ensure more accurate statements about the use of existing self-report stress measures in people with MID.

4.6 Implications for Future Research

Our study provides an overview of existing self-report stress measures, but can only offer limited guidance on the suitability of the self-report measures for people with MID. Despite many relevant arguments for the use of self-report measures in intellectual disability research, there are few validated self-report measures available, with even fewer for sensitive topics like stressful experiences (Ali et al., 2008; Ruddick & Oliver, 2005). Information on the suitability of a self-report measure for certain subgroups within the general population such as persons with cognitive impairments, limited verbal abilities, or clinical populations, is generally found in the manual or published peer-reviewed validation research. However, in many cases, self-report measures do not have detailed manuals, the manuals are unavailable, or they do not even exist. We therefore strongly advise future researchers to always publish clear user manuals and/or assessment procedures of self-report measures, even if they seem to be simple and easy to use. In addition, for those self-report measures not specifically designed for people with (M)ID, there is no published research on the use in the MID population. Moreover, norm data from validity studies are often based on research that excluded people with MID *a priori* based on their level of IQ. The relevance/suitability of many of the self-report stress measures found for people with MID therefore still remains unclear. More research is needed on the 'performance' of a measurement instrument in populations including people with MID. Therefore, we recommend that future validation studies of self-report measures always include a subpopulation composed of respondents with MID.

As noted earlier, stress is operationalized by many different theoretical constructs in the self-report stress measures analyzed. This raises the question of whether this could affect the measured results. On the other hand, research also shows that the operationalization of apparently different concepts, such as "stress" and "state-anxiety", essentially measure the same items and therefore can be regarded as the same type of outcome (Hook et al., 2008; de Witte, Pinho et al., 2020; de Witte, Spruit, et al., 2020). This has led to these concepts being used interchangeably in literature when it comes to outcome studies (Bradt & Dileo, 2014; de Witte, Spruit, et al., 2020; Wetsch et al., 2009). Nevertheless, we think it essential to provide a theoretical framework underpinning the measurement concepts involved. Not

only will this offer the necessary background information for future users, like clinicians, but it also increases the content validity of the self-report measure (Higgins & Straub, 2006; Lynn, 1986).

In order to validly and reliably assess stress-related outcomes in people with MID, attempts should be made to make the self-report stress measures more 'MID-inclusive'. However, it is still not entirely clear which specific instrument components or adaptations are required for this purpose. The recent study by Kooijmans et al. (in press) shows that there are still many gaps to fill on this topic. Findings show, for example, that researchers and clinicians assume questions should be read aloud by the assessor in order to assist people with MID. However, there is reason to believe that this may introduce various forms of bias in the results, arising from complex interviewer-interviewee dynamics (Finlay & Antaki, 2012). More research on the impact of assistance on the outcome of self-report measures is needed to decide whether this is an acceptable practice.

Lastly, the literature shows that Likert scales with three to five answer alternatives can be reliably used in research with people with MID (Fang et al., 2011). However, in the field of stress research, more nuanced response formats may be needed to capture the subtle differences in perceived stress over time. Visual Analogue Scales (VAS), for example, may offer an interesting alternative for this and have potential for assessing stress levels in people with MID. The Subjective Units of Distress Scales (SUDS) developed by Wolpe (1969) is an example of this. Notably, Mevissen et al. (2016) show promising results when using the SUDS in the treatment of trauma-related symptoms of people with MID. As many VAS scales differ in form, more research is advised on how to optimally attune these VAS scale formats to the needs of people with MID.

In conclusion, many adults with MID frequently experience stress in daily life and this has a major impact on their wellbeing. This emphasizes the importance of assessing stress levels as part of their support needs assessment. Research suggests that self-report measures are more accurate and sensitive compared to proxy measures. However, this scoping review found that there are few self-report stress measures suitable for this purpose.

This underlines the need for continuing efforts to develop high quality and "MID-sensitive" self-report stress measures.

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CHAPTER 6
6

Development of a Music Therapy Micro-Intervention for Stress Reduction

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Abstract

The negative impact of stress is a serious risk factor for the onset and progression of a wide range of physical illnesses and emotional problems. In the literature, we have seen an increasing examination of music therapy interventions for stress reduction over the past decade, yet music therapy interventions for stress reduction have not been systematically developed and described. Moreover, there is a growing need to develop *micro-interventions*, which are defined as short parts of sessions in which the music therapist uses specific therapeutic techniques to work on a client's goals. Therefore, we developed a music therapy micro-intervention for stress reduction based on the both empirical and practice-based knowledge, in which two developmental phases could be distinguished. In the first phase, the micro-intervention was described based on the integration of findings from empirical studies (N = 52) focused on the effects of music therapy on stress reduction, and the main findings from a previously held focus group study focused on the perspectives of music therapists. In the second phase, the Delphi technique was applied to collect feedback on the micro-intervention described, and to arrive at a group opinion by surveying a panel of 16 music therapy experts. This procedure resulted in an improved description of the music therapy micro-intervention for stress reduction, including a receptive and an active intervention variant. Implications for clinical practice and recommendations for future research are discussed.

Keywords: Music Therapy, Stress Reduction, Arousal, Micro-Intervention, Delphi Method

1. Introduction

The negative impact of stress can be a serious risk factor for the onset and progression of a wide range of physical and emotional problems (American Psychological Association [APA], 2017; Australian Psychological Society [APS], 2015). It is well known that music can provide relaxation and calmness, which ensures that music therapy interventions are increasingly used to reduce stress and enhance the well-being of clients across a variety of clinical populations (Agres et al., 2021; Bainbridge et al., 2021; de Witte, Spruit, et al., 2020; Juslin & Västfjäll, 2008). Several reviews show positive effects of music therapy interventions on stress reduction (e.g. Bradt et al., 2016; Landis-Shack et al., 2017; Martin et al., 2018; Pelletier, 2004; de Witte, da Silva Pinho, et al., 2020; de Witte, Spruit, et al., 2020). However, no specific music therapy intervention for stress reduction has yet been systematically described or protocolized. From a scientific point of view, clear intervention descriptions are needed to further investigate what is effective in music therapy interventions (Hoffmann et al., 2014).

1.1 The Impact of Stress

In daily life, almost everyone experiences stress from time to time. In the short term, stress can lead to reduced concentration and difficulty learning new information (The American Institute of Stress, n.d.). Long-term stress can lead to psychopathology such as anxiety disorders, depression, addictions and burnout (Akin & Iskender, 2011; Pittman & Kridli, 2011; Wang et al., 2019), as well as to health issues, such as high blood pressure, cardiovascular disease, insomnia and an increase or decrease in weight (Bally et al., 2003; Keech et al., 2020; Pittman & Kridli, 2011). To cope with stressors, millions of people around the world use tranquilizing medications, which are associated with numerous contraindications and negative side effects (e.g., Bandelow et al., 2015; Olfson et al., 2015; Puetz et al., 2015). It is therefore important to develop and examine promising non-pharmacological interventions for the prevention and management of stress, such as experiential approaches which focus on the "here and now" while guided by a therapist through stress responses and real-time emotional regulation. Through safely structured active experiences, stress inducing situations can be co-navigated, and stress reducing strategies can be developed and/or practiced (de Witte, 2017; de Witte, da Silva Pinho, et al., 2020).

1.2 Music Therapy for Stress Reduction

Music therapists are specifically trained to use the unique qualities of music, also known as musical components, (e.g., melody, rhythm, tempo, dynamics, pitch) in the therapeutic relationship to work on the patient's treatment goals (Bruscia, 1987; Wheeler, 2015; de Witte, da Silva Pinho, et al., 2020). During music therapy sessions, music therapists attune to the patient by adjusting the way of music-making as an *immediate response* to the client's needs (Aalbers et al., 2019; Magee, 2019). This can be related to the term "synchronization," meaning that the music therapist and the patient interact simultaneously and are regulated through

time, yielding a similar expression in movement, matching pulse, rhythm, dynamics and/or melody (Aalbers et al, 2019; Bruscia, 1987; Schumacher & Calvet, 2008; de Witte, da Silva Pinho, et al., 2020). For example, the music therapist may influence patients' perceived stress during musical improvisation by synchronizing with the patient's music-making, subsequently changing the musical expression by playing slower and less loudly (de Witte, da Silva Pinho, et al., 2020). This specific form of patient-therapist attunement is commonly used in music therapy practice and refers to the so-called Iso Principle (e.g., Altshuler, 1948; Heiderscheit & Madson, 2015). The literature shows that the tempo and loudness are important for the experienced intensity of the music (Gabrielsson & Lindström, 2010), and music with a slow steady rhythm may provide stress reduction by altering inherent body rhythms, such as heart rate (Thaut & Hoemberg, 2014; Thaut et al., 1999). Thus, the stress reducing effect of music therapy interventions can be explained by music itself as well as the continuous attunement of music by the music therapist to the individual needs of a patient.

There has been a rapid increase of research on the effects of music therapy on stress reduction. Results of a recent meta-analytic review (de Witte, da Silva Pinho, et al., 2020), including 47 quantitative controlled studies, showed an overall medium-to-large effect of music therapy on stress-related outcomes (d = .723, [.51-.94]). This is in line with previous reviews and meta-analyses, which show positive effects of music interventions on the reduction of stress or state-anxiety (Bradt & Dileo, 2014; Bradt, Dileo, & Potvin, 2013; Bradt, Dileo, & Shim, 2013; Bradt et al., 2011; Carr et al., 2013; Gold et al., 2009; Kamioka et al., 2014; de Witte, Spruit, et al., 2020).

1.3 The Need for Music Therapy Micro-Interventions

In music therapy literature, the term "intervention" may refer to both a specified therapeutic action and a process of intervening characterized by a structured and coherent collection of therapeutic actions (Aalbers et al., 2019; de Witte, Lindelauf, et al., 2020). Music therapy interventions may thus vary from one single technique or action within a single music therapy session to therapy programs or protocols consisting of multiple therapy sessions. In the last decade, there has been a growing recognition that one-size-fits-all approaches to intervention may be suboptimal for the patient and healthcare system alike (Gauthier et al., 2017; Rush et al., 2004). Moreover, it is assumed that intervention effects are variable across patients both in magnitude and time (Cuijpers et al., 2012; Kessler et al., 2017). This argues for the need to develop more flexible and more widely applicable interventions in accordance with the patient's needs, such as micro-interventions. A music therapy micro-intervention can be regarded as a short part of a session in which the music therapist uses specific therapeutic techniques or steps to work on specific patient's goals (Hakvoort, 2020; Hakvoort & van der Eng, 2020). Despite the fact that micro-interventions are short-lived, they have been systematically described and follow a step-by-step approach based on both recent theoretical models as well as the latest scientific evidence.

The development of music therapy micro-interventions is important to music therapy practice, on the one hand because the way of intervening in micro-interventions is strongly linked to core components of music therapy, on the other hand because describing interventions helps to further develop the profession. As the level of a clients' perceived stress can differ from session to session, it is important that music therapists can respond directly to their clients' stress levels, at the time it is needed in the specific context of that moment. This fits well with the specific way of patient-therapist attunement widely used in music therapy and which can be seen as one of the main characteristics of music therapy. Therefore, short-term therapeutic interventions that align easily with the existing structure of the session or clients' musical preferences are particularly suitable. Describing micro-interventions may also stimulate transferability of valuable clinical practices which in turn may strengthen thinking about the relationship between clinical practice, theory, and research (Aigen, 1999; Smeijsters & Vink, 2006; Stige, 2015).

1.4 Purpose of the Present Study

In the literature, we have seen an increasing examination of music therapy for stress reduction in the last decade (de Witte, da Silva Pinho, et al., 2020; de Witte, Spruit, et al., 2020). In addition, there is a growing need for music therapists to be more explicit about their' tacit knowledge in order to create more transferability in the way they work on a client's stress relief (see also: de Witte, Lindelauf, et al., 2020). Without these descriptions, music therapists face difficulties in reliably implementing interventions in their clinical practices and researchers can experience difficulties replicating studies (Hoffmann et al., 2014). By developing a micro-intervention in this context, we are in line with the recent developments in healthcare that emphasize the importance of short-term and flexible therapeutic interventions in general. In addition, the development of a micro-intervention is an important first step towards achieving more insight into which specific therapeutic factors lead to change, which is becoming increasingly important in the field of music therapy research (de Witte et al., accepted manuscript).

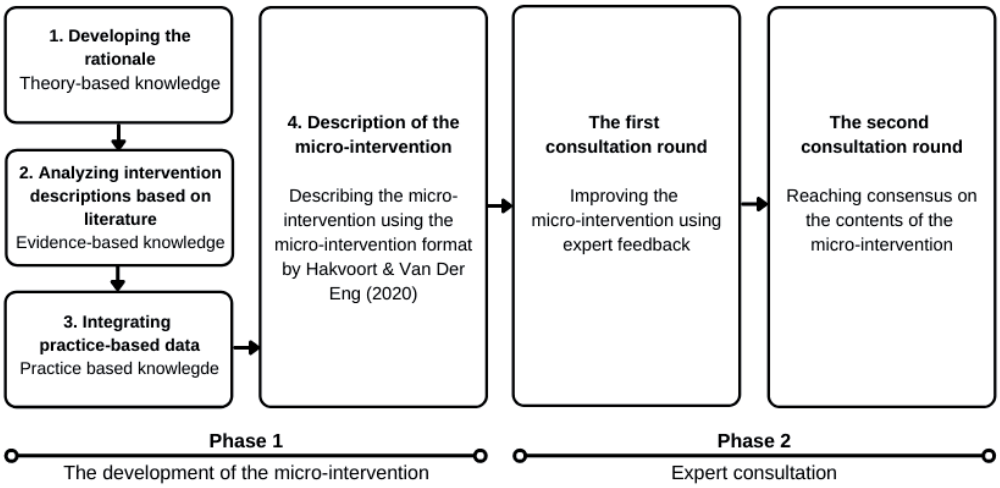
In order to provide a comprehensive analysis of music therapists' stress-reducing interventions, it is necessary to integrate the available practice-based knowledge. Published trials often demonstrate a lack of transparency in reporting detailed information on the content of the music therapy interventions (Aalbers et al., 2019; Robb et al., 2011). This is also evident in the recent meta-analysis by de Witte, da Silva Pinho, et al. (2020) in which the included studies mainly examined receptive (music listening) interventions, whereas in daily practice music therapists prefer to use active (music making) interventions to reduce their clients' stress (de Witte, Lindelauf, et al., 2020). In addition, developing a music therapy micro-intervention through an iterative process aimed at integrating theory-based, evidence-based, and practice-based knowledge is consistent with how other creative arts therapy interventions have been successfully developed (e.g., Aalbers et al., 2019; Bellemans et al., 2018; Haeyen et al., 2017). The main purpose of the present study is therefore to provide a

detailed description of a music therapy micro-intervention for stress reduction, which can be used directly by music therapists as well as provide a clear basis for future research.

2. Method

In this study, two developmental phases can be distinguished, namely, (a) describing the micro-intervention by analyzing and integrating the perspectives from both literature and clinical practice, and (b) consulting experts in the field of music therapy to reach consensus on the content and application of the micro-intervention developed. See Figure 1 for the procedural diagram of the method of the current study.

Figure 1
Procedural Diagram of the Method



2.1 Phase 1: the Development of the Micro-Intervention

We used a recently developed format by Hakvoort and Van Der Eng (2020) to describe the micro-intervention as this was particularly designed for describing music therapy micro-interventions. The use of this format ensures a comprehensive and detailed description and encourages a grounded scientific rationale. The format consists of several sections that must be described, such as specification of the target group, treatment domains, function of music, requisites, therapeutic attitude, the scientific / theoretical foundation, and a stepwise description of the micro-intervention.

2.1.1 Describing the rationale

To describe a theoretical rationale for the use of the music therapy micro-intervention, we needed both to clearly understand the problem of stress as well as a framework for how music therapy leads to stress reduction. To make both the origins and consequences of stress more concrete, we searched for literature in common online databases (Pubmed, PsycINFO, Web of Science, Wiley Online Library, ScienceDirect, and Google Scholar). To provide a theoretical framework on the relationship between music and stress, we mainly used the rationales of two recent meta-analytic reviews on the effects of music interventions and music therapy on stress-related outcomes (de Witte, da Silva Pinho, et al., 2020; de Witte, Spruit, et al., 2020). Both studies can therefore be regarded as providing key input to describing the scientific rationale for the micro-intervention. In addition, the introductory sections of the empirical studies on the effects of music interventions on stress included in the analysis of this study, were screened for additional theoretical background information.

2.1.2 Analyzing intervention descriptions based on literature

The following step involved analyzing the *intervention descriptions* of 52 empirical studies examining the effects of music therapy interventions on stress-related outcomes to create a solid basis for the content of the micro-intervention. The majority of the studies (n = 47), correspond to those included in the recently performed meta-analysis by de Witte, da Silva Pinho, et al. (2020). The primary aim of this earlier study was to demonstrate the overall effect of music therapy interventions on stress-related outcomes, in which a detailed analysis of the interventions examined was not taken into account. In this study, we therefore provide an in-depth analysis of particularly the *content* of the music therapy interventions examined. Five studies were initially excluded in the meta-analysis due to lack of quantitative data, however, we included them for the purpose of our study. The included studies concerned both clinical controlled trials (CCT) and randomized controlled trials (RCT) conducted in medical and mental health care settings, examining the effects of music therapy interventions on physiological and/or psychological stress-related outcomes. Only those studies in which a trained and qualified music therapist offered the intervention were selected. See de Witte, da Silva Pinho, et al. (2020) for more information about the applied search strategy and selection criteria; an overview of the characteristics of the 52 studies included in our study can be found in Table A5 (Appendix 5, page 263).

We then analyzed the extracted intervention descriptions using the coding principles of qualitative content analysis, which is frequently applied to answer questions such as what, why, and how, whereby the common patterns in the data were deduced using a consistent set of codes to organize text into identified categories of similar meanings (Cho & Lee, 2014; Moretti et al., 2011). To first gain more insights into how the initial data related to the particular sections of the format for the micro-interventions described by Hakvoort and van der Eng (2020), the open codes were grouped into “interventions and methods”, “non-musical interventions”, “preconditions”, “instruments and genres”, and “treatment goals”. Open codes

either identical or very similar to each other were then grouped, such as “patient chooses song” , “patient selects songs” and “patient chooses music”. If a code could not be merged with others, we left it separate. This axial coding step led to the categorization of codes based on their overarching similarities to property levels (Corbin & Strauss, 2008).

2.1.3 Integrating practice-based data

After the analysis of intervention descriptions, the next step was the examination of practice-based knowledge, as outcome studies do not always reflect clinical practice in all its facets. For this purpose, we used an existing dataset of a previous qualitative study. The aim of this particular study was to gain insights into how music therapists reduce their clients’ stress, especially in people with mild intellectual disabilities. It consisted of three focus groups held in three different countries in which 13 music therapists participated (see De Witte, Lindelauf, et al., 2020). The data from this study was transcribed and open coded by topic. The topic “interventions used within the music” proved particularly relevant for purposes of the present study. The open codes were extracted and then added to the initial categories that emerged from the analysis of the intervention descriptions from the literature.

2.1.4 Analysis of combined data

Due to the differing amounts of data, the categories consisting of of either at least 4 codes from intervention descriptions from the empirical literature or at least 2 codes from the practice-based data were included as an *intervention component*. The categories formed by practice-based codes only were counted twice compared to those from the literature. Selective coding was then applied to create an integrated model in which those categories of intervention components could be linked to each other to interpret the steps of the micro-intervention (Charmaz, 2003). To minimize possible bias on the part of the researcher who analyzed the data, the entire process of data analysis was continuously monitored by two co-authors (SH and MDW) and decisions were made in consensus to ensure that the confirmability criteria were met.

2.1.5 Description of the micro-intervention

Based on the analyses of the empirical literature and the qualitative data, the micro-intervention was described following Hakvoort and Van der Eng (2020), consisting of a theoretical rationale, intervention goals, type of setting, treatment phase in which it can be applied, and contraindications. In order to further shape the specific intervention content, we used the integrated model of intervention components that emerged from data analysis. To establish consensus on the summarized narratives, we organized a final member check of a subgroup of the research team (MdW, AK, SvH).

2.2 Phase 2: Expert Consultation

In the second phase, the Delphi technique was applied in order to arrive at a group opinion by surveying a panel of experts, and to reach consensus through an iterative process of collecting feedback (Linstone & Turoff, 1975; McMillan et al., 2016; Skulmoski et al., 2007). The Delphi consensus procedure was applied in two different iterations.

2.2.1 Participants

Participants were sixteen music therapists and researchers in the field of music therapy. All had at least eight years of working experience as a music therapist. Six of them had already participated in one of the focus group interviews from our recent qualitative study (de Witte, Lindelauf, et al., 2020). Ten participants were recruited through the international network of music therapists and researchers associated with KenVaK – a research center for arts therapies (www.kenvak.nl). Selected participants were located in four different countries, namely Belgium, Germany, the Netherlands and the United States. All participants gave informed consent.

2.2.2 The first consultation round

An online questionnaire (available in Dutch and English) along with the described micro-intervention was sent to the experts. The questionnaire focused on all sections of the described micro-intervention and, for each topic, the level of agreement could be indicated with a four -point Likert scale. In addition, the participants could add comments in each section, e.g. on reason of disagreements, new suggestions, and other feedback. In addition, the questionnaire included questions on the participants' professional background to gain more insights into their individual perspectives. The questionnaire can be requested from the first author. The amount of agreement was calculated for each section. All suggestions and comments were listed in a file and analyzed by content. Then, every suggested change from the original micro-intervention description was discussed (MdW, AK, SvH) and decisions were made in consensus with each other. This procedure resulted in a renewed and improved description.

2.2.3 The second consultation round

For the second consultation round, the adapted version of the micro-intervention was sent again to the participants with a brief summary of the processed feedback and suggestions. They were asked to respond within two weeks if they disagreed with aspects of the new version of the micro-intervention.

3. Results

3.1 The First Phase: the Development of the Micro-Intervention

3.1.1 Describing the rationale

***Identifying the problem of stress*.** In the short term, it is known that the negative impact of stress can lead to reduced concentration and difficulties when learning and memorizing new information (Schwabe & Wolf, 2010). As a result, working on treatment goals when the client is experiencing stress will be less effective and inefficient (de Witte, Lindelauf, et al., 2020). It is therefore important to first reduce stress and tension so that the client is able to focus on the initial treatment goals. An overview of related health consequences of both long term and short term stress are described in the introduction of this study. A leaner narrative of this information is described in the format of the micro-intervention at "Specific domain that is targeted or treated" (see Table A6).

***Clarifying the stress-reducing effect of music interventions*.** Both music listening (receptive) and music making/singing (active) have been associated with a reduction of *physiological arousal* which increases during stress; this is visible in a reduction of cortisol levels or decrease in heart rate and blood pressure (e.g. Hodges, 2011; Koelsch et al., 2016; Kreutz et al., 2012; Linnemann et al., 2015; Nilsson, 2009). In addition, a large body of neuroimaging studies show that music can influence stress-related emotional states by modulating activity in brain structures, such as the *amygdala*, that are known to be involved in emotional processes (e.g. Blood & Zatorre, 2001; Hodges, 2011; Koelsch, 2015; Levitin, 2009; Moore, 2013; Zatorre, 2015). An increased dopamine activity in the mesolimbic reward brain system has been shown to be associated with feelings of happiness in response to listening to favorite/own-preferences music (e.g., Blood & Zatorre, 2001; Salimpoor et al., 2011; Salimpoor et al., 2013; Zatorre, 2015).

Plausible explanations for the positive effects of music interventions on stress, can also be sought in psychological and behavioral oriented scientific theories. Listening to pleasant music may have a positive influence on *emotional valence*, which can be explained by the degree of attraction that an individual feels towards a specific object or event (Jäncke, 2008; Juslin & Västfjäll, 2008). Music experienced as pleasant increases the intensity of emotional valence (the felt happiness), which has a stress-reducing effect (Jiang et al., 2016; Rohner & Miller, 1980; Sandstrom & Russo, 2010; Witvliet & Vrana, 2007). Listening to music can also provide direct *distraction* from stressful feelings or thoughts (Bernatzky et al., 2011; Chanda & Levitin, 2013). Research that shows the benefits of music to distract people from aversive states is supported by short-term music interventions for acute stress reduction (Fancourt, et al., 2014; Linnemann et al., 2015; de Witte, Spruit, et al., 2020). Lastly, music listening or music making together with others is also related to stress relief (Juslin et al., 2008). This can be explained by the fact that people synchronize with each other during music activities which evokes feelings of togetherness and social cohesion during the music experience (Boer & Abubakar, 2014; Linnemann et al., 2016). This in turn may be explained

by the release of the neurotransmitters endorphin and oxytocin (e.g., Dunbar et al., 2012; Freeman, 2000; Tarr et al., 2014; Weinstein et al., 2016), which are positively associated with the defensive response to stress (e.g. Amir et al., 1980; Dief et al., 2018). A short narrative of these findings is presented in the format of the micro-intervention at "Function of music during the intervention" (see Table A6).

3.1.2 Analysis of the interventions from literature

Some of the included studies only offered limited descriptions of the examined intervention, whereas others offered detailed and rich intervention descriptions or even intervention protocols. However, each intervention description led to one or more open codes. Intervention descriptions showed both receptive interventions (*n* = 22), such as listening to live or pre-recorded music, as well as active interventions (*n* = 10), such as improvisation, playing existing songs, and songwriting. A combination of both receptive and active interventions was found in 20 of the intervention descriptions. In addition, in 20 of the studies, a complementary intervention/technique was offered along with music therapy, such as breathing exercises, muscle relaxation, and mindfulness exercises. In 38 intervention descriptions, the specific use of music (musical instruments or singing) was reported. Singing was mentioned in most studies (*n* = 28), followed by percussion instruments (*n* = 21), guitar (*n* = 15), and piano (*n* = 10). After the open coding step, categories were formed through axial coding by similar codes being grouped.

3.1.3 Integration of the practice-based data

Data-analysis of the focus groups indicated that the participating music therapists mainly use many active interventions (14 interventions) and few receptive interventions (one mentioned) for stress reduction. The active interventions included musical improvisation, playing existing music, songwriting, recording own music, and singing mantras or preferred songs. In addition, data showed that the following therapeutic techniques are most often used to reduce a client's stress: synchronization, pacing, structuring, increasing and decreasing dynamics and tempo, repeating themes, simple musical structures, and using familiar instruments and songs. The first coding step resulted in 33 categories and remaining single codes.

3.1.4 Analysis of the combined data

The final analysis resulted in a total of 14 categories of intervention components, which we present in order of the total number of codes counted: *music based on preferences* (10), *patient chooses song* (7), *expressing emotions* (5), *recording music* (4), *patient chooses intervention* (4), *therapist chooses intervention* (4), *verbal processing of emotions* (4), *using familiar songs* (4), *music based on emotional state* (4), *accelerating tempo* (4), *keeping appropriate physical distance* (4), *simplicity in harmony* (4), *slowing down tempo* (4), and *lower register* (4). These intervention components formed input for the further design of the micro-intervention.

3.1.5 Description of the micro-intervention

The next step was to describe the music therapy micro-intervention in detail. Information on the theoretical background of the problem of stress and the rationale for using music interventions to lower people's stress levels were added in the format for music therapy micro-interventions (Hakvoort & van der Eng, 2019). Then, intervention goal, the target population and field, possible contra-indications, requisites, and specification of the setting, were supplemented. In order to remain as close as possible to the results of the data analysis, we described both an *active* and a *receptive* variant of the music therapy micro-intervention. This allows music therapists to choose the variant that best suits the client's needs and possibilities at that moment.

Because the micro-intervention is specifically designed to directly reduce the client's stress, i.e., in the music therapy session itself, the intervention goal was formulated as follows: "reducing tension and stress directly in the music therapy session". We consider the micro-intervention as transdiagnostic and therefore it does not only relate to the treatment of one specific condition or disorder. However, as research shows that some client populations are more vulnerable to stress, such as people with mild intellectual disabilities or those with impaired cognitive functions (e.g., Emerson, 2003; Scott & Havercamp, 2014), we expect that the music therapy micro-intervention might be particularly suitable for these client groups. Precisely because of this broad applicability and the fact that we developed two variants (receptive and active), there were no contra-indications. However, clients with severe autism, severe intellectual disabilities, or clients suffering from acute psychosis are expected to have difficulty participating because of their reduced ability to be in contact with the therapist. The main prerequisites for applying the micro-intervention include a sound-isolated room (especially in clinical settings), chairs for the client(s) and music therapist, access to a sufficient selection of musical instruments (active variant), and sheet music of the client's preferred music (receptive variant). Furthermore, the micro-intervention can be applied both individually and in groups. See Appendix 6 for more details of the abovementioned content of the micro-intervention.

All 14 intervention components were included in one of the micro-intervention variants (see Figure 2 and 3). However, analysis showed that the intervention components "*patient chooses the intervention*" and "*therapist chooses the intervention*" appeared to contradict each other. If the client chooses the intervention, often used to appeal to client autonomy, it means that the therapist is not able to decide to use the micro-intervention. As music therapists use the micro-intervention precisely when it is needed to lower their client's stress levels, it is not possible to have the client choose the intervention themselves. However, to encourage client autonomy in another way, instead of letting them choose the intervention, we decided to offer them the choice of the song in the receptive variant and that they could take the lead role in the active variant by building up the music tempo and dynamics.

Figure 2
The Active Variant of the Micro-Intervention

Active variant:

1. The music therapist chooses to use the micro-intervention when the client indicates feeling stressed or tense, or when the therapist observes stress in the client [6]. The music therapist explains the micro-intervention and its purpose in clear language. The music therapist coordinates physical proximity in such a way that both the music therapist and the client have sufficient space, that there is a comfortable distance between them and that eye contact can be made [11].
2. The client and music therapist both choose an instrument [5] by which large dynamic variations can be made, such as a djembé, drum kit, piano or guitar. If this proves difficult, the therapist can suggest a number of instruments to choose from. If the music therapist chooses to use a harmonic instrument, the harmonies should be simple [12] and preferably played in the lower register [14].
3. The music tempo and dynamics are adjusted to the current stress level of the client [9]. Before proceeding with the micro-intervention, the music therapist checks whether the intensity of the music matches the client's current level of stress. If this is not the case, the music therapist adjusts the intensity based on observations and–when possible–instructions from the client.
4. If the music matches the client's stress level, the volume and music tempo will be increased evenly, such as when releasing/discharging [3, 10]. The client has a leading role in building up the music tempo and dynamics, and also determines when the intensity has reached a peak and can be reduced again [5]. The music therapist actively guides the client in this part of the intervention and helps to evenly bring the tempo back to 60-80 bpm and the dynamics to silence [13].
5. Afterwards, the music therapist and client discuss how the client experienced any tension during the intervention and how the stress has changed after the intervention [7].
6. The improvisation can be recorded and listened to [4] to reflect better, to indicate moments of tension and relaxation more easily, or to compare the intensity and course of the intervention in different sessions.

1. ~~Music based on preferences~~, 2. ~~Client chooses song~~, 3. Expressing emotion, 4. Recording music, 5. Client chooses intervention*, 6. Therapist chooses intervention, 7. Verbal processing of emotions, 8. ~~Using familiar song~~, 9. Music based on emotional state, 10. Accelerating tempo, 11. Keeping appropriate physical distance, 12. Simplicity in harmony, 13. Slowing down tempo, 14. Lower tonal register.

Techniques that have been crossed out do not apply in this variant.

Figure 3
The Receptive Variant of the Micro-Intervention

Receptive variant:

1. The music therapist chooses to use the micro-intervention when the client indicates feeling stressed or tense, or when the music therapist observes stress in the client [6]. The music therapist explains the micro-intervention and its purpose briefly. The music therapist coordinates physical proximity in such a way that both the music therapist and the client have sufficient space, that there is a comfortable distance between them and that it is easy to make eye contact [11].
2. The client chooses a song [1, 2, 5, 8]. If this proves difficult the music therapist can suggest a few songs or let the client choose from a song book. The music therapist plays the (if necessary simplified) chords [12] of this song on guitar or piano, preferably in the lower register [14]. The client is invited to listen.
3. The music tempo and dynamics are adjusted to the client's current stress level [9]. Before proceeding with the micro-intervention, the music therapist checks whether the intensity of the music matches the client's current level of stress. If this is not the case, the music therapist adjusts the intensity based on observations and–when possible–instructions from the client.
4. The music therapist can then choose to further increase the intensity of the music [10] and then decrease it, or to let the intensity be equal to the client's stress level and then gradually decrease the tempo to 60-80 bpm and the dynamics to silence [13].
5. Afterwards, the music therapist and client discuss how the client experienced any tension during the intervention and how the stress level has changed after the intervention [7].

1. Music based on preferences, 2. Client chooses song, 3. ~~Expressing emotion~~, 4. ~~Recording music~~, 5. Client chooses intervention*, 6. Therapist chooses intervention, 7. Verbal processing of emotions, 8. Using familiar song, 9. Music based on emotional state, 10. Accelerating tempo, 11. Keeping appropriate physical distance, 12. Simplicity in harmony, 13. Slowing down tempo, 14. Lower tonal register.

Techniques that have been crossed out do not apply in this variant.

3.2 The Second Phase: Expert Consultation

3.2.1 The first consultation round

Analysis of the experts' feedback led to significant changes in the description of the micro-intervention. Based on their suggestions, we added information, i.e. "voice" as one of the main instruments, allowing the client to experience the present stress before reducing it, specifying the therapeutic role and attitude with four functional domains, and the supporting role of the group when the micro-intervention is offered to just one of the group members. A note was added on contra-indications regarding clients suffering from trauma or anxiety disorders and on the function of the concepts "synchronizing" and "containing" which are

related to stress reduction. An overview of the feedback given can be requested from the first author.

3.2.2 The second consultation round

The second round of consultation resulted in consensus among all experts, meaning that no further changes had to be made. The final description of the micro-intervention can be found in Appendix 6 (page 270).

4. Discussion

In our study, we systematically developed a music therapy micro-intervention aimed at stress reduction based on findings from theoretical and empirical studies as well as practice-based knowledge. The micro-intervention was developed for and evaluated by music therapists for use during the music therapy session when it is necessary to lower clients' stress levels. Although the micro-intervention does not relate only to the treatment of one specific condition or disorder and can be considered as broadly applicable, the literature indicates that some client populations may benefit more due to their higher vulnerability to stress, such as people with mild intellectual disabilities (MID) or those with impaired cognitive functions (e.g., Emerson, 2003; Scott & Havercamp, 2014). For them, an experiential approach may be more appropriate than the cognitive approach (Didden et al., 2016; de Witte, Lindelauf, et al., 2020). To our knowledge, this is the first study in which a music therapy micro-intervention has been systematically developed with the aim of direct stress reduction in the music therapy session.

4.1 Strengths and Limitations of the Present Study

The way the micro-intervention was developed has several strengths. First, the systematic and comprehensive approach, which relied on data both from empirical studies as well as from clinical practice, resulted in a well-described micro-intervention. This approach has many similarities to the "Intervention Mapping" approach, a systematic method for the development, implementation, and evaluation of health interventions by constructing programs grounded both in theory and on empirical data (Bartholomew et al., 2011). However, Intervention Mapping was originally designed to create larger or longer-term intervention and treatment programs (Eldridge et al., 2016), and therefore does not fully align with the concept of micro-interventions, which are short-term interventions and can even be used as stand-alone techniques in existing treatment programs (de Witte, Lindelauf, et al., 2020). Second, the inclusion of data derived from controlled outcomes studies (RCTs and CCTs: N = 52) offered a scientifically robust foundation for the core elements of the micro-intervention. This is relevant so that the basic claims made in the present study are clear (Aalbers et al., 2019; Crooke et al., 2016). Moreover, the included outcome studies were derived from a

recently conducted meta-analytic review (de Witte, da Silva Pinho, et al., 2020) in which the inclusion criteria exactly matched the aims of this study. It can also be argued that this study strengthens the overall scientific basis of music therapy for stress reduction, as the previous meta-analysis looked primarily at effects using quantitative analyses, while in this study we qualitatively analyzed the content of the intervention, thus answering the *how* music therapeutic interventions can lead to stress reduction. Third, in the second phase of this study, the micro-intervention was submitted for consultation to music therapy experts from different countries in order to reach consensus in a collaborative process. Thus, thanks to this expert evaluation, the micro-intervention does not rely solely on pre-existing data. This strengthens its generalizability and makes it more plausible that the micro-intervention can be implemented easily in clinical practice.

Some limitations need to be noted. Through the years, several theoretical models have been developed to provide insights into the influence of music on stress. One of the most widely used models of the last decade involves models rooted in biological and neurological theories, so we also used these models to provide theoretical explanations of the relationship between stress and music. These models formed the basis of two earlier meta-analytical reviews of music interventions for stress reduction (de Witte, da Silva Pinho, et al., 2020; de Witte, Spruit, et al., 2020). However, we are aware that the general construct of stress integrates many scientific fields, in which both environmental, psychological, and biological/physical factors are interrelated within a comprehensive framework (Aldwin, 2007; Cohen et al., 2007). In this sense, the strength of exclusively including intervention information from controlled outcome studies can be seen as a limitation; information on the content of interventions can also be obtained from less robust designs, such as case studies or one group designs. However, the importance of analyzing intervention content that demonstrates positive effects was paramount in our study. Related to the previous, the data from this large number of outcome studies mainly showed descriptions of receptive interventions, while the practice-based data almost exclusively showed active interventions. This may indicate a gap between what is applied in clinical practice and what is investigated in robust research designs (de Witte, Lindelauf, et al., 2020). However, it may also be related to the context of a specific target group, such as clients with MID, who were central in the practice-based data. Because we wanted to stay as close as possible to the initial data, this led to the development of two different variants of the micro-intervention: the active and the receptive variant (see Figure 2 and 3).

4.2 Recommendations for Future Research

Clear intervention descriptions are needed to further investigate what is effective in music therapy interventions (Hoffmann et al., 2014). Future research should focus on whether the developed music therapy micro-intervention for stress reduction does lead to stress reduction during the session. However, methods that can measure the direct effects of the micro-intervention on stress-related outcomes will be needed. Previous reviews of stress measures

show that many researchers emphasize the importance of measuring stress outcomes related to both physiological arousal as well as to people's subjective experiences (Scott & Havercamp, 2018; de Witte, Kooijmans, et al., submitted for publication; de Witte, Spruit, et al., 2020).

As it is still unclear *how* and *why* music therapy interventions lead to certain outcomes such as stress reduction, more research on therapeutic factors[4] is needed to further develop music therapy micro-interventions. In our micro-intervention, *music tempo* can be seen as one of the most important elements, and therefore we expect it to be an important therapeutic factor leading to stress relief. This is in line with previous research showing that music tempo can be considered one of the most significant moderators of music-related arousal and relaxation effects (e.g., Bringman et al., 2009; de Witte, Spruit, et al., 2020). We therefore recommend that future research includes a secondary research question that focuses on therapeutic factors, such as the tempo of the music, in order to increase knowledge not only regarding efficacy, but also regarding what contributes to these effects.

Micro-interventions also allow researchers to conduct a *micro-analysis* of specific parts of the music therapy session (Lee, 2000; Wosch & Wigram, 2007). The most important questions are: "what exactly happened and why?". Through micro-analysis, therapy processes can be better understood or clarified, for example by analyzing the musical activity, social interaction, or nonmusical behavior of a short segment of a session (Wosch & Wigram, 2007). Micro-interventions are therefore highly suitable for pinpointing specific therapeutic factors that cannot be examined when testing over a larger period of time (de Witte, Orkibi, et al., 2021).

4.3 Implications for Clinical Practice

Micro-interventions lend themselves well to music therapy practice because of their flexible character and the way in which the therapist can respond to the client's needs in the moment itself. By offering a musical frame, any musical expression produced by the client can be musically encouraged and responded to in a musical dialogue (e.g., Aigen, 2005; MacDonald et al., 2013; Nordoff & Robbins, 1966). Most components of our developed micro-intervention are strongly related to certain therapeutic factors of music therapy, namely "musical dialogue" and "shared musical experiences". Other important therapeutic factors concern the structuring nature of music, such as tempo. A recent review shows that it is precisely these therapeutic factors that are often associated with positive change in music therapy (De Witte, Orkibi, et al., 2021). In addition, the content of the micro-intervention is also in line with Bruscia's principles (1987), who developed 64 musical improvisation techniques based on using the unique qualities of music to establish or influence the musical dialogue with the client; these still form the basis of global music therapy education. However, the concept of the micro-intervention provides more insights into (a) the needs and abilities of certain client populations that the intervention is focused on, (b) particular outcomes, (c)

4 Therapeutic factors are those factors identified by empirical studies that lead to therapeutic change and are associated with particular outcomes (Elliot, 2010; Kazdin, 2009).

specific characteristics of the intervention, and (d) the underlying theoretical models that explain the relationship between music (therapy) and the targeted outcome. By describing this information, which is mainly subconsciously understood and applied by music therapists, it may stimulate them to strengthen the transferability of their clinical work and may provide more insights into the relationship between clinical practice, theory, and research (Aigen, 1999; Smeijsters & Vink, 2006; Stige, 2015).

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CHAPTER 7

7

General Discussion

1. Brief Introduction

The goal of this dissertation is to contribute to the body of scientific knowledge on music interventions for stress reduction. All over the world, music is a part of people's daily lives, bringing joy, motivation, and positive distraction, and most importantly, providing relaxation in stressful times. Indeed, the calming and stress reducing effects are its most widely studied effects of music (Chanda & Levitin, 2013; Juslin & Västfjäll, 2008; Koelsch, 2015). The first aim in this dissertation was to examine the overall effects of different types of music interventions on stress-related outcomes by reviewing the empirical literature, and to assess whether the overall effects were influenced by outcome, study, sample and intervention characteristics. The second aim was to provide more insights into how to accurately assess subjective stress experiences, as well as evaluate their applicability in a target group known for high levels of stress, in particular people with MID. The third aim was to systematically collect practice-based knowledge on how music therapists can reduce stress in their clients during music therapy sessions. The final goal was to develop and evaluate a music therapy micro-intervention for stress reduction integrating theory, evidence, and practice-based knowledge.

In this final chapter, first a summary of all results is presented. Second, the overall contribution of the findings to the empirical knowledge is discussed. Third, implications for clinical practice and directions for future research are provided. Fourth, the strengths and limitations of the studies included in this dissertation are discussed, and finally the overall conclusion of this dissertation will be presented.

2. Summary of the Studies

In the first study, a systematic review and two multilevel meta-analyses on the effects of *music interventions* on stress-related outcomes, an overall significant effect on stress reduction in both physiological (d = .380; 61 RCTs) and psychological (d = .545; 79 RCTs) outcomes was shown. We defined music interventions as purposeful musical exercises or methods in which music listening, music making, or singing is central, offered by a music therapist (in the context of music therapy), by other healthcare professionals or without any support (music medicine). Significant stronger moderating effects were found in studies in which physiological arousal was measured by heart rate, compared to those measuring blood pressure or stress-related hormone levels. None of the other moderator variables of intervention effects proved significant. However, the results did show some noteworthy trends of intervention characteristics. Larger effect sizes were found in studies where the music tempo of 60-80 bpm was offered, and where only one session was offered compared to multiple sessions. Moreover, the findings showed that when music therapy was offered it had at least the same effect (d = .423; 7 RCTs) on physiological stress-related outcomes as in those studies in which "music medicine" was offered (d = .379; 54 RCTs), both with no

significant difference. This can partly be explained by the lack of statistical power due to the difference in the number of studies. Overall, this meta-analytic review provides high-level evidence that music interventions can be effective in reducing stress, and it provides justification for the increasing use of music interventions for stress reduction in both medical and mental health care practice.

The second study involved a systematic review and meta-analysis specifically examining the effects of music therapy on stress reduction. Music therapy was defined as the clinical and evidence-informed use of music interventions to accomplish individualized goals within a therapeutic relationship, offered by a trained and qualified music therapist. Overall, we found a significant medium-to-large overall effect of music therapy on stress-related outcomes (d = .723; 47 RCTs/CCTs). Significant stronger moderating effects were found in studies conducted in non-Western countries compared to Western countries, studies using quasi-experimental CCTs compared to RCTs, and studies with a waiting list control condition compared to those with CAU or another stress-reducing intervention. As in the first meta-analysis, the selected music therapy variables did not moderate the overall effects of music therapy on stress reduction, but the large effects of music with a tempo of 60-90 bpm (d = .900), group music therapy (d = .927), and the impact of more sessions of music therapy (d = .894), are worth noting. This meta-analytic review provides evidence that music interventions, in the context of music therapy carried out by a qualified music therapist, are effective in reducing stress in both medical settings and mental healthcare practice.

In the third study, we conducted three focus group interviews with music therapists from different countries working with patients with MID to gain more insight into *how* music therapists exactly lower the stress levels of their patients with MID during music therapy. The main findings indicated that three therapy goals for stress relief can be distinguished, namely, “synchronizing”, which can be seen as a sub goal because it often precedes working on the other goals of “tension release” or “direct relaxation”. Little to no receptive (e.g., music listening) interventions for stress reduction were used by the music therapists; they preferred to use active interventions, which were mainly based on musical improvisation. Furthermore, results show that the tempo and the dynamics of the music are considered the most important musical components for reducing stress in adults with MID. Implementing a solid and predictable musical structure during applied interventions was identified as an important factor leading to stress reduction in adults with MID.

In the fourth study we presented a scoping review about the validity and reliability of self-report stress measures, and furthermore their suitability for people with MID. Since people with MID are amongst those who are very vulnerable to stress, timely recognizing their levels of stress is important. Results showed a total of 13 self-report stress measures that met the inclusion criteria, of which three were developed specifically for assessing stress in adults with MID: the GAS-ID, LI, and SAS-ID. The findings showed that in contrast to self-report stress measures developed for the non-ID population, self-report measures developed for people with (M)ID often allow the respondent to be assisted during the assessment (SAS-ID),

and that the items can be read aloud by someone else (LI and GAS-ID). Moreover, results show that the use of simpler wording, fewer response options, and the ability to provide supportive visualization may lead to more appropriate responses of individuals with MID. We noted a lack of clearly written user manuals, as well as a lack of validation studies that include a subpopulation composed of respondents with MID or cognitive impairments.

The final study concerned the development of a music therapy micro-intervention for stress reduction through an iterative process based on the integration of theoretical and empirical studies, and practice-based knowledge. A music therapy micro-intervention can be defined as a short part of a session in which the music therapist uses specific therapeutic techniques or steps to work on specific patient's goals. We distinguished between two developmental phases, namely, (a) describing the micro-intervention by analyzing and integrating the perspectives from both empirical studies and clinical practice, and (b) consulting experts in the field of music therapy to reach consensus on the content and application of the micro-intervention. In the first phase, a qualitative analysis of the intervention descriptions of 52 controlled outcome studies on music therapy for stress reduction was performed, which eventually led to 14 overarching categories of intervention components. Then practice-based data from the previously held focus group study was integrated, which finally resulted in a detailed step-by-step description of both a receptive and active variant of the music therapy micro-intervention. In the second phase of this project the Delphi technique was applied in order to collect feedback on the micro-intervention developed, and to arrive at a group opinion by surveying a panel of 16 music therapists and researchers. This procedure resulted in an improved description of the music therapy micro-intervention for stress reduction.

In the following section, the findings of the studies will be discussed in the light of both science and clinical practice. Several issues related to the effectiveness of music interventions on stress reduction and the possible influence of the both study- and intervention variables on the effects found will be discussed. The importance of accurately measuring the subjective experience of stress, especially in vulnerable patient populations like those with MID, is the final topic to be addressed in the discussion of the findings of this dissertation.

3. Discussion of the Findings

3.1 Overall effects of Music Interventions on Stress-Related Outcomes

This dissertation contributes significantly to the body of scientific knowledge on music interventions for stress reduction. In the first meta-analytic review, music interventions were shown to have significant overall effects on physiological and psychological stress-related outcomes. This is consistent with findings from other reviews and/or meta-analyses on the effects of music interventions on stress-related outcomes (Bradt et al., 2016; Bradt & Dileo, 2014; Bradt, Dileo, & Potvin, 2013; Bradt, Dileo, & Shim, 2013; Gillen et al., 2008; Kim et al., 2015; Pelletier, 2004; Rudin et al., 2007). The second meta-analytic review on the effects

of *music therapy* interventions showed an even larger significant overall effect on stress reduction. Here again, our findings are in line with previous reviews and/or meta-analyses on the effects of music therapy on stress-related outcomes (Bradt et al., 2016; Bradt & Dileo, 2014; Bradt, Dileo, & Potvin, 2013; Bradt, Dileo, & Shim, 2013; Carr et al., 2013; Gold et al., 2009; Kamioka et al., 2014). However, most of these previous reviews and meta-analyses focused on one particular clinical setting or patient population, whilst both meta-analytic reviews in this dissertation have shown the relevance of music interventions for stress reduction in various kinds of medical- and mental healthcare settings. In both meta-analyses, no moderating effects were found for the type of setting in which the music intervention was applied, which strengthens the assumption that the positive effects of music interventions on stress reduction are independent of context or setting. This is consistent with neurological evidence for the positive effects of music interventions on the stress response with respect to arousal regulation, with no indications that the influence of music interventions on the stress response varies across contexts (Koelsch, 2015; Koelsch et al., 2016; Linnemann et al., 2015; Pfaff et al., 2007; Thaut & Hoemberg, 2014).

3.2 Possible Moderating Effects of Intervention Variables

Contrary to expectations raised by previous research, the findings of both meta-analytic reviews showed no significant moderating effects of intervention variables. Although the absence of statistically significant moderator effects can be partially attributed to the lack of statistical power in some of the categories of intervention variables, it remains difficult to explain which of the intervention characteristics do actually contribute positively to the overall effect sizes. With respect to the meta-analysis on music therapy interventions (Chapter 3), this lack of significant moderator effects of intervention variables may also be explained by the wide diversity of the music therapeutic approaches/methods used in clinical practice, and that interventions are often tailored to clients' needs at that particular moment (e.g., Agres et al., 2021; Magee, 2019; Wheeler, 2015), as was argued in Chapter 4. This in turn is reflected by the fact that most of the studies included did not report on the use of a therapy protocol or a step-by-step description of the applied intervention(s). Thus, despite the growing number of systematic reviews and meta-analyses on music interventions, reflecting the increase in individual outcome studies, little is still known about the specific factors that contribute to these positive effects. The findings of a recent review emphasize that research focusing specifically on therapeutic factors or mechanisms of change is only just beginning to emerge in the field of music therapy (de Witte et al., accepted manuscript).

Despite the fact that *music tempo* did not significantly moderate the overall effect of music interventions on stress reduction, findings of both meta-analyses showed larger effect sizes for music with 60–90 bpm, which is associated with a slow music tempo, compared to music with another or unspecified tempo. A slow music tempo is widely considered to be one of the most crucial moderators of relaxation effects (Bernardi et al., 2005; Bringman et al., 2009; Chlan, 2000; Hilz et al., 2014; Nomura et al., 2013). It is known that music tempo

and changes in tempo can influence different physiological and neurological responses, for example arousal, which is strongly related to the degree of perceived stress (Roth, 2014; Thaut et al., 1999).

This implies that an increase in music tempo leads to increased activation of the both nervous system, increased muscle tension and heart rate, whereas a decrease in tempo can lead to muscle relaxation and a lowered heart rate, resulting in more relaxation and thus stress reduction (e.g., Bernardi et al., 2006; Bringman et al., 2009; Juslin & Västfjäll, 2008; Nomura et al., 2013). However, this has mostly been demonstrated in experimental studies with healthy subjects instead of intervention studies including patients with stress-related complaints (Elliot et al., 2011; Ooishi et al., 2017). In addition, although music tempo is considered the most crucial component of music when it comes to relaxation effects, the literature also shows that it often involves complex interactions between various musical components, such as rhythmic regularity, slow tempo, and simple and consonant harmonies (Gabrielsson & Lindstrom, 2001; Ooishi et al., 2017). It therefore remains difficult to demonstrate causality between music tempo and stress reduction.

In clinical practice, music tempo is also considered one of the most important ingredients of stress-reducing interventions (Bruscia, 1987; Davis et al., 2008); this was supported by the music therapists contributing to our focus group study (Chapter 4). Despite both the existing evidence and the collected experiences from clinical practice, there were no significant moderator effects of music tempo on stress reduction found in both of the meta-analytic studies. From the perspective of music therapy practice, where *active* interventions are mainly applied, a plausible explanation is that the exact music tempo during these interventions often varies or is difficult to determine, and therefore it is almost never reported in outcome studies. This in turn may be related to the fact that music therapists are specifically trained to attune to the patient by adjusting the way of music-making as an immediate response to the patient's needs (e.g., Aalbers et al., 2019; Magee, 2019). Using one and the same music tempo is therefore not feasible or even not desirable. Our findings reflect this by suggesting that synchronizing with the patients' music making and then facilitating changes in tempo is an effective way of achieving stress reduction.

Furthermore, despite the overlapping findings of both meta-analytic reviews, the notable differences need to be discussed further, such as the *direct* influence of music interventions on stress reduction versus longer term effects, the influence of the specific *study design* of the included studies, and the use of *receptive* versus *active* music interventions for stress reduction.

3.3 The Direct Stress-Reducing Effect of Music Interventions

The first meta-analytic review on *music interventions* showed similar or even larger effects when only one session was offered compared to multiple sessions. This may be partly explained by the fact that the included studies primarily concerned music listening interventions for stress reduction in a medical context, such as "music medicine", which were

offered only once before/during/after a medical procedure. Moreover, in medical settings, the physiological measures used to assess the direct effects of music interventions on stress reduction are commonly part of standard medical treatment, such as measures of heart rate and blood pressure (e.g., Gupta & Gupta, 2015; Lee et al., 2017). This could explain that in this first meta-analysis the frequency and /or number of sessions did not positively influence the overall effect of music interventions, indicating that just one session can lead to the desired stress relief. On the other hand, it is consistent with previous research, which found that music in general has an immediate positive effect on stress reduction (e.g., Koelsch, 2015; Zatorre, 2015), making single session effects likely.

However, the second meta-analytic review on *music therapy* showed that the overall effect of music therapy on stress-related outcomes actually increases with the use of multiple sessions. This is consistent with the previous meta-analytic review by Gold et al. (2009), who demonstrated more substantial benefits in psychiatric patients who took a longer course of music therapy or attended more frequent sessions. A more recent review on the effectiveness of music interventions on depression also showed that more than one session yielded larger effects (Leubner & Hinterberger, 2017). However, these reviews did not examine stress-related outcomes. As in the first meta-analysis, only studies conducted in a medical setting measured one-session effects of music therapy on stress-related outcomes. In contrast to the first meta-analysis, physiological stress-related outcomes (n = 9), which are strongly related to assessing the direct effects of music on stress levels, were rarely included compared to psychological stress-related outcomes (n = 45). This may be explained by the fact that music therapy studies are more often conducted from a psychotherapeutic point of view instead of listening to prerecorded music for stress relief before, during or after medical procedures. This is also reflected by the fact that 12 of the included music therapy studies were conducted in mental healthcare settings in which psychiatric patients with stress-related complaints were treated, whereas none of these patients /settings were included in the first meta-analysis. In music therapy research, including physiological measurements is difficult due to practical and ethical considerations. Such measurements may in fact (unintendedly) induce stress in patients.

In summary, although in both meta-analyses differences were shown with regard to the direct effect of music interventions on stress reduction, these differences may be attributed to study variables, such as the type of setting (medical- or mental health care) and the type of music intervention (music therapy or music medicine). With respect to the differences between the studies included in the meta-analyses, both direct effects as well as effects over multiple sessions can be expected with regard to music interventions for stress reduction. It is precisely the direct stress-relieving nature of music interventions that fits so well with the needs and possibilities of vulnerable client populations, such as people with MID, that led us to develop a music therapy micro-intervention for the direct reduction of stress during the music therapy session.

3.4 The Effect Moderating Impact of the Study Design

Another difference between the findings in the two meta-analytic reviews concerns the methodology of the selected studies. Three issues related to study design are particularly important to discuss. First, the meta-analytic review on music interventions only included studies using an RCT design in contrast to the meta-analytic review on music therapy interventions in which also (non-randomized) controlled clinical trials (CCTs) were included. This may have partly contributed to the larger effect size on stress-related outcomes of the meta-analytic review on *music therapy* (d = .723) compared to the meta-analytic review on *music interventions* (d = .380 for physiological outcomes and d = .545 for psychological outcomes).

Since selection bias in non-randomized study designs can lead to overestimations of treatment effects (e.g., Page et al., 2018; Valentine & Thompson, 2013), it is crucial to mark study design as an effect-moderating variable if both RCT and CCT design are included in meta-analyses, especially with respect to intervention studies. However, although RCTs are still considered the primary source for evidence on the efficacy and safety of clinical interventions, a meta synthesis by Vinkers et al. (2020) in which more than 170,000 RCTs were analyzed showed that many RCTs had severe methodological flaws, and their results were often biased. The authors also conclude that the likelihood of bias in RCTs has generally decreased in recent years, which may be the result of increased knowledge on research methodology, improved education, trial registrations, and more stringent reporting guidelines and journal requirements (Sundell & Åhsberg, 2018; Vinkers et al., 2020).

The second aspect of study design that can impact the overall effects in meta-analyses concerns the type of control condition. In the meta-analysis of music therapy (Chapter 3), studies comparing music therapy with waiting list control groups showed significantly larger effects than studies with care as usual (CAU) or other stress-reducing intervention control conditions. This is in line with findings from previous research that indicate that trials with waiting list controls may overestimate treatment effects (Cunningham et al., 2013; Hart et al., 2008). However, effectively withholding interventions from control group participants can be difficult to implement in clinical practice due to ethical concerns (Barkauskas et al., 2005; Mohr et al., 2009). The ethical benefits of waiting list designs are therefore important to consider, because they allow interventions to be provided to research participants seeking help instead of intentionally delaying interventions because of research interests (Mohr et al., 2009).

The third study design variable that may overestimate overall effect sizes in meta-analyses concerns the degree of blinding procedures applied. There is empirical evidence of pronounced bias due to lack of patient blinding, specifically with respect to trials including patient-reported outcomes (Hróbjartsson et al., 2014). Moreover, blinding procedures are a particularly important factor when assessing study quality of controlled trials included in systematic reviews and meta-analyses (Effective Public Health Practice Project [EPHPP], 2009; Jüni et al., 2001). Despite the fact that study quality did not significantly moderate

the overall effect of music therapy on stress-related outcomes, low quality studies yielded larger effects compared to those with a moderate or strong study quality. The blinding of participants seems to be especially difficult in music therapy studies, unless two types of music therapy interventions are compared, such as receptive music therapy versus active music therapy (Bradt, Dileo, & Shim, 2013). Notwithstanding, there are alternative blinding procedures more appropriate to music therapy research, such as procedures in which only the researcher is blinded to the allocation of the intervention or in which participants are blinded to the purpose of the study (Bradt, Dileo, & Shim, 2013; Day & Altman, 2000; Magee et al., 2017).

Thus, although blinding and randomization procedures, and the use of control conditions, are the principal methods for ensuring internal validity of intervention studies, previous studies have shown that when these procedures are not rigorously followed, it actually leads to biased results. However, according to Sundell and Åhsberg's (2018) review on methodological quality in controlled trials, the content of published articles do not always provide a true reflection of the study quality, but may instead reflect the quality of reporting as necessitated by publication requirements. In addition, Magee et al. (2017) report poor reporting quality on blinding of participants and outcome assessors in their Cochrane review on effects of music interventions and strongly advise researchers to improve this concerning future publications. Therefore, we want to emphasize that in music therapy research it is crucial to accurately report the methodological procedures as well as the arguments on which the study design was chosen, so that a good examination of effect-moderating study-related variables can be made.

3.5 Receptive and Active Music Interventions

A notable difference between the findings in both the meta-analytic reviews (evidence-based data) and the findings in chapter 4 (practice-based data) in this dissertation concerns the use of *receptive* versus *active* music interventions. The studies included in both meta-analyses primarily involved receptive music interventions, whereas music therapists in clinical practice primarily use active music interventions to reduce their clients' stress levels. This may indicate that despite the growing body of evidence on the positive effects of receptive music interventions, music therapists in clinical practice prefer to involve the client actively in the music-making process to reduce stress instead of offering interventions in which the client only "receives" the music through listening to live or prerecorded music. This is in line with the literature showing that interventions in which patient(s) and therapist actively improvise on musical instruments are highly common in music therapy practice (e.g., Bruscia, 1987; Gold et al., 2009; Pavlicevic, 2000; Wigram, 2004). On the other hand, music therapists are also familiar with receptive interventions for relaxation, especially in the context of dementia care, rehabilitation, or palliative settings (e.g., Magee, 2019; Tsoi et al., 2018; Warth et al., 2015). However, these are often receptive interventions in which the music therapist performs live

music for the patient and can thus attune directly, by means of music-making, to a patient's needs.

Nevertheless, we believe it is important to provide plausible explanations for this gap between the findings of evidence-based literature and experiences in clinical practice. One of the main explanations may be that receptive interventions in the context of "music medicine", based on listening to prerecorded music, align well with RCT designs because randomization and blinding procedures can usually be applied. In contrast, most effectiveness studies of music therapy are quasi-experimental, as in daily practice it is often difficult to meet the requirements for randomization and blinding procedures due to practical and ethical concerns (Bradt, 2012; Bradt, Dileo, & Shim, 2013; Magee et al., 2017). More specifically, participants included in research on receptive music interventions in a medical setting visit the hospital for medical treatment, therefore randomization and allocation procedures related to the music intervention do not impact their access to initial medical treatment. In addition, these types of receptive music interventions often use prerecorded music administered through headphones provided by medical personnel, making it easier for both the patient and the caregiver to be blinded to the intervention. This may have contributed to the striking increase in RCTs on the effects of receptive music interventions on stress reduction in medical settings.

The focus group study presented in Chapter 4 covered music therapy interventions in the context of mental healthcare. While the effects of receptive interventions in medical contexts are mainly related to the general influence of music on the stress response, the effects of music therapy interventions applied in mental healthcare settings may be explained by means of the therapeutic relationship. Through patient-therapist attunement by the active use of music, music therapists individualize their interventions to meet the patients' needs (Agres et al., 2021; Bradt & Dileo, 2014; Wheeler, 2015). Moreover, the focus group study only included music therapists working with patients with MID, who are known to benefit from an action- and experiential approach to intervention (Cuijpers et al., 2007; Didden et al., 2016; de Witte et al., 2017). This may also explain the fact that the participating music therapists were more likely to apply active music interventions to reduce their clients' stress.

3.6 Measuring the Subjective Experience of Stress

In the various studies in this dissertation, measuring the negative impact of stress is considered an important issue. Indeed, in biomedical literature, stress is not necessarily considered to be a negative outcome. Increased physical stress levels are also associated with positive outcomes such as personal growth, increased feelings of self-esteem and self-efficacy, or enhanced performance (Aldwin, 2007; Cohen et al., 1997). Stress is therefore a complex concept to measure in intervention studies where stress is usually studied primarily as a health-related risk factor. Furthermore, research shows that even when referring to the negative impact of stress on health, the perception of stress is related to a wide range of individual factors involving differences in perceptual processes and cognition (Kopp et al., 2010; Monroe, 2008).

Outcome studies show that the degree of stress can be measured from three different perspectives: (a) the environmental perspective, which focuses on specific stressors or life events; (b) the biological perspective, which assesses the activation of physiological arousal by means of biomarkers; and (c) the psychological perspective, which assesses subjective stress responses involving emotional states (Cohen et al., 1997; Kopp et al., 2010). As primary human responses to stress involve both physiological and psychological outcomes, it is necessary to take both types of outcomes into account in order to validly and reliably examine the direct effects of music interventions on stress.

In the last decade, much research has been presented on how to measure stress-related physiological arousal, leading to various physiological measurement methods involving biomarkers such as heart rate (HR), blood pressure, heart rate variability (HRV), or hormone levels (e.g., Chandola et al., 2010; Föhr et al., 2017; Marrocco et al., 2017). In psychology literature, there is consensus on the aspect that psychological measurement instruments should be adapted to be used in special to patient groups, such as people with MID with regard to language use and assessment procedures (Kooijmans et al., submitted for publication). Thus we need to answer the question of how exactly this can be done and what standards can be used. While the scoping review of Chapter 5 provides insight into the characteristics of existing self-report stress measures, and the extent to which they are applicable to people with MID, it also demonstrates the difficulty of comparing those self-report stress measures and providing clear guidelines. This could be explained by the many different theoretical underpinnings and constructs of the concept of stress used across the self-report measures found.

Despite the increase of empirical knowledge on both the origins and negative consequences of stress, as well as on the assessment of stress, the lack of consensus on the meaning and operationalization of stress among the various instruments, is also seen in two previous reviews on stress measurements (Kim et al., 2018; Thayer et al., 2012). Thayer et al. (2012) suggest that this may be the result of the absence of a comprehensive model to examine how humans in general function in and adapt to constantly changing environments. However, since stress is understood as being produced by a person-environment transaction (Aldwin, 2007; Riley & Park, 2015), and moreover can be considered a transdiagnostic factor, it may not be feasible or even be desirable to pursue a single theoretical framework for measuring stress-related outcomes. Notwithstanding, comparing the findings of outcome studies or adequately generalizing them to clinical practice is thus a complex matter in which one should carefully consider both the characteristics of measurement instruments and the patient population and clinical setting involved. However, since the negative impact of stress is recognized as a global problem, and has been subject to numerous empirical studies in recent decades (American Psychological Association [APA], 2017; World Health Organization [WHO], 2010), it is strongly recommended that future research takes into account how stress-related outcomes can best be measured with respect to the specific context.

Thus, although the scoping review of Chapter 5 does not yet provide a firm answer to the question of how the psychological experience of stress (especially in people with MID) can be measured as accurately as possible, it does contribute to the increasing scientific knowledge on self-report stress measurements, which can be used in more rigorous study designs for future research into the effects of music interventions on stress reduction.

3.7 Vulnerable Client Populations

Another topic we addressed in this dissertation concerned music therapy interventions for patient populations known to be especially vulnerable to stress, such as people with MID (Hatton & Emerson, 2004; Schuengel & Janssen, 2006; WHO, 2010). This group was subject of both the scoping review presented in Chapter 5 and the focus group study presented in Chapter 4. Although the music therapy micro-intervention for stress reduction presented in Chapter 6 is suitable for use in many different target groups, we believe it is particularly suitable for adults with MID as this approach allows them to engage more easily in stress reducing activities. The paradox we encountered, however, is that although people with MID may especially benefit from experiential stress-reduction interventions that offer direct stress reduction (Hooper et al., 2008; de Witte et al., 2017), it remains challenging to accurately assess the experiences of these individuals because of their cognitive limitations (Schalock et al., 2010), which is supported by both our findings in Chapter 5 and the literature showing that there is still a great need for high-quality self-report measures of stress for adults (Scott & Havercamp, 2018; Kooijmans et al., submitted for publication).

Although our scoping review provided insights into the existing self-report stress measures and showed its applicability for adults with MID, it remains a first overview with recommendations for future research. For example, we recommend developing evidence-based guidelines for (a) the direct use of existing self-report stress measures in people with MID and furthermore (b) adapting or tailoring these self-report measures to better align with the needs of adults with cognitive limitations in general. Since it is particularly important to be able to validly and reliably assess the effects of music interventions on stress, especially in populations particularly vulnerable to stress, we recommend that future studies not only focus on effectiveness, but also on how self-report stress measures can be applied. On the one hand, the negative impact of stress is best captured by combining data on physiological (biomarkers) and psychological (self-reported information) stress-related outcomes. On the other hand, encouraging the use of self-reported information ensures that the voice of people with MID is better heard in decision-making processes, as society increasingly recognizes that people with MID are full citizens with the same rights as non-disabled people (e.g. Devi, 2014; Giesbers et al., 2019).

4. Strengths and Limitations

Given the widespread use of music interventions for relaxation and/or stress relief and the accompanying rapid increase in effectiveness studies, it is remarkable how little empirical evidence is available regarding the specific characteristics of music interventions that may influence stress-related outcomes. In this dissertation, we were therefore not only focused on the effects of music interventions on stress reduction, but we also aimed to gain insights into why and how music interventions can lead to stress reduction. A strength of this dissertation is that the included studies provide clear insights into the differences in effects and applicability of the two main domains of music interventions: music therapy and music medicine, which are still often considered one and the same in the scientific literature (Agres, et al., 2021; Bradt et al., 2016). Chapters 2 and 3 therefore not only increase the body of scientific knowledge on the effectiveness of music interventions in general, but also show the differences between both types of music interventions, with important implications for future research and clinical practice.

As previously addressed in the general discussion, this dissertation provides new empirical knowledge about the impact of research methodology with respect to the effects found in both types of music interventions, which in turn is strongly connected to the type of setting in which studies are conducted (mental healthcare versus medical healthcare settings) and the way they are administered (receptive versus active interventions). In addition, Chapter 4 shows that the music interventions considered effective by music therapists in clinical practice are less likely to emerge in effectiveness studies. By demonstrating this potential gap between research findings and what is observed in clinical practice, we provide new directions for future research on both the effects of music interventions on stress-related outcomes and possible therapeutic factors.

It is increasingly recognized that intervention effects vary from patient to patient, both in magnitude and in time course (Cuijpers et al., 2012; Kessler et al., 2017), which strongly argues for the development of interventions that are more flexible and more widely applicable in accordance with the patient's needs, such as micro-interventions. The development of a music therapy micro-intervention for stress reduction (Chapter 6) can therefore be considered as a strength, as it is consistent with this growing recognition that a one-size-fits-all approach to interventions may be suboptimal for both the patient and healthcare system (Gauthier et al., 2017; Rush et al., 2004). Moreover, the development of the micro-intervention is based on both knowledge from efficacy studies on music interventions for stress reduction and the perspectives of clinical practice. As a result, the micro-intervention content is consistent with the available evidence on the subject and grounded in the reality of clinical practice, which can be considered a strength. By processing the findings of the studies in this dissertation, we have been able to do justice to both the high-level evidence that emerged from both meta-analytic reviews, and to the knowledge derived from clinical practice through expert consultation.

A related aspect, which may be seen as a shortcoming, is that, besides the music therapy experts, we did not involve future users /clients of the micro-intervention in the development process. Thalen et al. (2021) suggest that clinical initiatives such as therapeutic interventions should be systematically evaluated and improved based on the knowledge and experiences of its current users. It is believed that these participatory approaches will not only enhance clinical practice but also strengthen the common research methodology (Bach et al., 2017).

Another aspect that may be considered a possible limitation is that as the meta-analytic review regarding music therapy (Chapter 3) showed no significant difference in effect size between studies conducted in non-Western and Western countries, we did not consult music therapists from non-Western countries in the two studies (Chapters 4 and 6). However, this was not an intentional consideration, but the result of the existing international network on which we relied. A closer look at the data reveals that the non-Western countries included in the meta-analysis show substantial heterogeneity in terms of culture and topographic region, and that a large proportion of the Western countries also include high percentages of immigrants, making it difficult to equate country and culture (Morales & Ladhari, 2011). Nevertheless, it is important to consider potential cultural differences when it comes to the experience of stress and how this may involve outcome measures (e.g., Lonner, 2007; Tweed et al., 2004) and the way music interventions can be most effectively applied.

5. Directions for Future Research

In addition to the recommendations for future research already mentioned, some recommendations are brought to special attention here as a result of the above discussion of the main topics, complemented by the strengths and limitations of this dissertation. First, more research is needed on the differences and similarities of receptive interventions versus active interventions to increase knowledge about what type of music intervention works for whom at what specific moment. Although this is what was recommended in the conclusion of a previous Cochrane review on music interventions (Vink et al., 2013), only a few studies have directly compared the two types of music interventions. These studies hypothesized better outcomes for active music interventions. However, the results showed no significant differences in effect between both types of interventions (Aalbers et al., 2019; Atiwannapat et al. 2016; Bradt et al., 2015). In this dissertation, we clearly demonstrate the potential gap between what effectiveness studies focus on and what music therapists actually apply in clinical practice. We therefore strongly recommend that this be taken into account in future research, despite the methodological difficulties.

Second, although findings of both meta-analyses of Chapter 2 and 3 have shown larger effect sizes for some intervention characteristics, such as a music tempo of 60-90 bpm, no significant moderating effects of these intervention characteristics were found. However, the

focus group interviews with music therapists in Chapter 4 revealed a number of *therapeutic factors*[5], which align with the findings of the moderator analyses of the meta-analyses. Specifically the tempo of the music was considered by the music therapists as one the most important intervention characteristics to reduce stress. The second recommendation, therefore, involves setting up more robust quantitative research in clinical practice to identify therapeutic factors associated with the effects of music interventions on stress reduction. Indeed, naturalistic study designs are an indispensable part of the process of generating and testing hypotheses about therapeutic factors of clinical interventions (e.g., Hayes, 2013; Kazdin, 2009). In this context, it is important to distinguish between "common factors", which are not related to one specific form of therapy but can be seen as general and overarching factors, and "specific factors," which are exclusively related to one specific therapeutic intervention (de Witte et al., accepted manuscript). With respect to music interventions for stress reduction, identifying specific factors may provide more certainty as to which intervention characteristics can influence effects. Nevertheless, we must also consider the possibility that common factors, which are thus not specifically related to music, substantially contribute to achieving the desired effect, as has been argued by a number of psychotherapy researchers (Grawe, 1997; Lambert, 2013). Therefore, we recommend including both types of therapeutic factors in future research, also because common and specific factors are likely to be significantly correlated (de Felice et al., 2019).

The third recommendation is that future research that targets music interventions for people with MID should be conducted in a participatory approach, where researchers strive for maximal inclusion of people with MID. This would involve increasing participants' engagement with regard to the intervention, but also building a strong collaboration between co-researchers with and without MID (McDonald et al., 2013; O'Brien et al, 2013).

The final direction for future research involves the further investigation of music therapy micro-intervention for stress reduction of Chapter 6. Since the micro-intervention is theoretically based and firmly grounded in both evidence-based and practice-based knowledge, it is timely to examine both the direct effects within a single session, and the effects after multiple sessions in which the micro-intervention was applied. Given the contrary findings of some of the studies of this dissertation on the use of receptive and active music interventions for stress reduction, it is highly recommended that future effectiveness studies also compare these two types of music interventions. Moreover, the developed music therapy micro-intervention already includes a receptive and an active intervention variant. In addition, examining music therapy micro-interventions also responds to the call for more research on therapeutic factors, because micro-intervention testing allows for micro-analyses on very brief segments of the therapeutic process. Micro-interventions are therefore highly suitable for pinpointing elements of the intervention that cannot be examined when testing over a larger period of time, such as the direct influence of music tempo on stress reduction. When

5 A well-specified change factor that is theorized to produce therapeutic benefits in a specific therapeutic approach (Kazdin, 2009).

examining the effects of the micro-intervention, it is important to include both physiological (stress-related biomarkers) and psychological (self-report measures) stress-related outcomes, and to conduct a proper analysis of which measurement instrument is best suited to the context in which it occurs.

6. Final Conclusion

The overall conclusion is that music interventions in the form of both music listening interventions and in the context of music therapy delivered by trained therapists can greatly benefit patients in medical and mental health care settings. Music therapy studies generally showed larger effects, which can be explained by the personalized, tailored approach of the music therapist specifically trained to attune musically to the patient's needs. However, these effects may also be partly related to some methodological weaknesses or the type of setting.

Nevertheless, more research into both the effects and applicability of music therapy interventions is needed, including (a) well operationalized concepts of stress, (b) study designs consistent with RCT standards, and (c) valid and reliable measurement methods. Research needs to be conducted in patient populations proven more vulnerable to stress such as those with cognitive impairments, because a more experiential approach to interventions is precisely what best matches their needs and abilities. The inclusion of a secondary research question aimed at identifying effect-moderating variables or therapeutic factors is an absolute necessity so that we can empirically explain the effects both in the scientific literature and in clinical practice.

According to the findings of this dissertation, music interventions should be implemented more structurally in both medical and mental healthcare settings. First, because of the high demand for non-pharmacological therapeutic interventions for stress reduction because of the increasing awareness worldwide of the negative side effects of tranquilizing medications, which are still often considered as one of the first treatment options. Second, because it is precisely the action-oriented and experiential approach to music interventions that is particularly well-suited to the needs of growing patient populations that are especially vulnerable to stress, and for whom a cognitive approach alone does not effectively address stress-related complaints.

Finally, this dissertation demonstrates the added value of a trained and qualified music therapist offering the music interventions for stress reduction. Despite the difficulty of examining these music therapy interventions, given the fact that they are often individually tailored and that music therapists are specifically trained to musically attune from moment to moment to respond to their patient's needs, more robust research that focuses on both efficacy and the hypothesized therapeutic factors is crucial to the further implementation of music therapy interventions in healthcare settings, particularly when it comes to stress reduction.

7. References

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CHAPTER 8
8

Summary

Summary

The goal of this dissertation is to contribute to the body of scientific knowledge on music interventions for stress reduction. To cope with the negative impact of stress, millions of people around the world use tranquilizing medication, which in turn is associated with numerous contraindications and negative side effects. Therefore, there is an urgent need to develop and examine innovative and non-pharmacological interventions for stress reduction, especially for patient populations known to be more vulnerable to stress and for whom there is still a scarcity of appropriate psychological interventions. People with mild intellectual disabilities (MID) are considered such a population, and currently account for more than 40% of psychiatric inpatients in the Netherlands.

Music interventions, including receptive interventions (the patient listens to the music) and active interventions (the patient is actively involved in making music), can be considered such non-pharmacological intervention. Music interventions can be offered by a qualified music therapist, in the context of music therapy, but can also be self-administered or offered by other (healthcare) professionals. Since the stress-reducing qualities of music have been associated with a broad range of positive outcomes, music interventions are increasingly being used to reduce stress in both medical and mental healthcare settings. Moreover, the experiential and action-oriented approach of music interventions is particularly well suited to the abilities and needs of people with cognitive impairments, such as those with MID. This dissertation therefore aims to increase scientific knowledge on (a) the effects of different types of music interventions on stress-related outcomes, (b) how and why music interventions may specifically lead to stress-reducing effects, and (c) how stress can be accurately assessed, specifically in people who are more vulnerable to stress, such as those with MID.

The first study in this dissertation (Chapter 2) examined the empirical literature on the effects of music interventions on physiological and psychological stress-related outcomes. To summarize the growing body of empirical research, two multilevel meta-analyses of 104 RCTs, containing 327 effect sizes and 9,617 participants, were performed to assess the strength of the effects of music interventions. By conducting two separate multilevel meta-analyses, the current study aimed to assess the strength of the effect of music interventions on both physiological and psychological stress-related outcomes. Furthermore, the study aimed to examine which outcome, study, sample or intervention characteristics moderated the strength of the effect on physiological and psychological stress-related outcomes. Overall, we found a significant small-to-medium effect of music interventions on physiological stress-related outcomes (d = .380; 61 RCTs) and a medium effect of music interventions on psychological stress-related outcomes (d = .545; 79 RCTs), indicating that music intervention groups benefited more than the comparison groups. Moderator analyses showed that the type of outcome assessment moderated the effects of music interventions on stress-related outcomes. Larger effects were found on heart rate, compared to blood pressure and hormone levels. None of the possible moderators of intervention effects proved to be significant, but

results showed some noteworthy trends of the intervention characteristics. Larger effect sizes were found in studies with a music tempo of 60-80 bpm and in studies offering only one single session compared to more than one session. Results showed no moderating effect for the settings in which the music intervention was conducted, which might indicate that the effects of music interventions do not depend on the type of setting. In conclusion, this study provides high level evidence that music interventions can be effective in reducing stress and provides justification for the increasing use of music interventions in both medical and mental healthcare practice.

The second study (Chapter 3) shows a meta-analytic review specifically examining the effects of music therapy interventions. To summarize the growing body of empirical research on music therapy, a multilevel meta-analysis, containing 47 studies, 76 effect sizes and 2.747 participants, was performed to assess the strength of the effects of music therapy on both physiological and psychological stress-related outcomes, and to test potential moderators of the intervention effects. Overall, we found a significant medium-to-large overall effect of music therapy on stress-related outcomes (d = .723; 47 RCTs/CCTs). Larger effects were found for clinical controlled trials (CCT) compared to randomized controlled trials (RCT), waiting list controls instead of care as usual (CAU) or other stress-reducing interventions, and for studies conducted in Non-Western countries compared to Western countries. The selected intervention characteristics did not have a statistically significant impact on the effectiveness of music therapy. However, some substantial differences in effect sizes were found in moderator analyses that did not reach the conventional level of significance due to lack of statistical power, mostly caused by an unequal distribution of studies (and effect sizes) among moderator categories. These involved larger effects in studies that offered a larger number of sessions, the use of music with a tempo of 60-90 bpm, and the provision of music therapy in a group setting. In conclusion, this study provides evidence that music therapy can be effective in reducing stress and provides justifications for the increasing use of music therapy carried out by a qualified music therapist in both mental health care practice and medical settings.

The third study (Chapter 4) provides more insight into the practice-based knowledge on *how* music therapists lower stress levels of their patients with MID during music therapy sessions. Three focus group interviews were conducted with music therapists working with adults with MID (N = 13) from different countries and clinical institutions in Europe. Results show an overview of the most-used interventions for stress reduction, specified by therapeutic goals, intervention techniques, the use of musical instruments, and related therapeutic change factors. Findings show that two ways of reaching stress reduction could be distinguished, namely, “tension release” or “direct relaxation”, which can also be regarded as two different therapy goals. The goal of “synchronizing” can be seen as a subgoal because it often precedes working on the other two goals. In addition, results indicate that music therapists used little to no receptive (e.g., music listening) interventions for stress reduction, but preferred to use active interventions, which were mainly based on musical

improvisation. The tempo and dynamics of the music are considered as the most important musical components to reduce stress.

The fourth study (Chapter 5) presents a scoping review to provide more insight into the validity and reliability of self-report stress measures and their suitability for people with MID. Findings from literature were supplemented with information from an expert consultation. Although the increased use of physiological measures to assess people's stress levels has added substantial value to stress research, it is no substitute for the importance of self-report measures, as literature shows that physiological and psychological stress-related outcomes are not necessarily directly related. Therefore, we felt it was timely to review existing self-report stress measures and thereby increase the knowledge of stress measurement in general. A total of 13 self-report stress measures met the final inclusion criteria, of which three were developed specifically for assessing stress in adults with MID: the Glasgow Anxiety Scale for people with Intellectual Disability (GAS-ID), the Lifestress Inventory (LI), and the Self-rating Anxiety Scale for adults with Intellectual Disabilities (SAS-ID). Five of the self-report stress measures found did not appear to be suitable for populations other than the normally gifted population, and that based on the literature, five of them could potentially be appropriate (see Chapter 5 for more details). The experts provided the following important recommendations, which may provide certain guidance to both clinical practice and future research: (a) using concrete and easy-to-understand vocabulary, simple grammar, and short sentences, (b) using short time frames for the retrieval of information (< one week), (c) response options in Likert scales should be limited to three, (d) an 'I don't know' answer option should be included, (e) using visualizations to support the meaning of questions and responses, and (f) using pre-scripted alternative wording if the respondent seems unable to understand the question. The findings of this study underlines the need for continuing efforts to develop high quality and "MID-sensitive" self-report stress measures. In addition, this study contributes to the increasing scientific knowledge on self-report stress measures, which can be used in more rigorous study designs for future research into the effects of music interventions on stress reduction.

The aim of the last study in this dissertation (Chapter 6) was to develop a music therapy micro-intervention for stress reduction, based and firmly grounded in both evidence-based knowledge and practice-based knowledge. A music therapy micro-intervention can be defined as a short part of a session in which the music therapist uses specific therapeutic techniques or steps to work on specific patient's goals. Despite the fact that micro-interventions are short-lived, they have been systematically described and do follow a step-by-step approach based on both recent theoretical models as well as the latest scientific evidence. In the first phase, a qualitative analysis of the intervention descriptions of 52 controlled outcome studies on music therapy for stress reduction was performed, which eventually led to 14 overarching categories of intervention components. Then practice-based data from the previously held focus group study was integrated, which finally resulted in a detailed step-by-step description of both a receptive and active variant of the music therapy micro-intervention. The Delphi

technique was applied in order to collect feedback on the micro-intervention developed, and to arrive at a group opinion by surveying a panel of 16 music therapists and researchers. This procedure resulted in an improved description of the music therapy micro-intervention for stress reduction.

Overall, there can be concluded that music interventions in the form of both music listening interventions and in the context of music therapy delivered by trained therapists can greatly benefit patients in medical and mental health care settings. Music therapy studies generally showed higher effects, which can be explained by the personalized, tailored approach of the music therapist specifically trained to attune musically to the patient's needs. However, these effects may also be partly related to some methodological weaknesses or the type of setting. More research is needed, especially in patient populations proven to be more vulnerable to stress as those with cognitive impairments. We believe that further research of music therapy micro-intervention for stress reduction from Chapter 6 lends itself well to this. Since the micro-intervention is theoretically based and firmly grounded in both evidence-based knowledge and practice-based knowledge, it is timely to examine both the direct effects within a single session, and the effects after multiple sessions in which the micro-intervention was applied. Furthermore, well operationalized concepts of stress, study designs consistent with RCT standards, valid and reliable measurement methods, and the inclusion of a secondary research question aimed at identifying therapeutic factors are strongly recommended in future studies.

APPENDICES

Chapter Appendices

Samenvatting (Dutch summary)

Curriculum Vitae

Dankwoord (Acknowledgements)

List of Publications included in this Dissertation

Chapter Appendices

Appendix 1 (Chapter 2)

Table A1
Characteristics of Included Studies of the Meta-Analyses

Authors	Year	N	Impact factor	Study quality	Type of measures	Type of outcome(s)	Physiological measure	Type setting	Intervention
Aba et al.	2017	186	1.379	Strong	psych	Anxiety	-	Med-proc	MA
Allen et al.	2000	40	4.580	Strong	phys	Stress	BP, HR	Surgery	MA
Ames et al.	2017	41	-	Strong	psych	Anxiety	-	Surgery	MA
Angioli et al.	2014	372	1.283	Moderate	phys, psych	Stress, Anxiety	BP, HR	Med-proc	MA
Bally et al.	2003	107	1.326	Strong	psych	Anxiety	-	Med-proc	MA
Bauer et al.	2010	80	2.050	Strong	psych	Stress	-	Med-proc	MT
Beck et al.	2015	20	0.800	Weak	phys, psych	Stress	Horm	Non-med	MT
Bekiroglu et al.	2013	60	1.935	Moderate	phys	Stress	BP	Med-proc	MA
Bringman et al.	2009	326	2.322	Moderate	phys, psych	Stress, Anxiety	BP, HR	Surgery	MA
Buffum et al.	2006	170	0.524	Weak	phys, psych	Stress, Anxiety	BP, HR	Surgery	MA
Bulfone et al.	2009	60	0.659	Moderate	psych	Anxiety	-	Med-proc	MA
Camara et al.	2008	203	1.170	Weak	phys	Stress	BP, HR	Surgery	MA
Cassileth et al.	2003	62	6.072	Moderate	psych	Anxiety	-	Med-proc	MT
Chan et al.	2006	43	1.197	Weak	phys	Stress	BP, HR	Med-proc	MA
Chen et al.	2012	73	1.754	Strong	psych	Anxiety	-	Med-proc	MA
Cutshall et al.	2011	100	1.243	Weak	phys, psych	Stress	BP, HR	Surgery	MA
Chang et al.	2015	76	1.426	Weak	phys	Stress	BP, HR	Med-proc	MA
Chang Yu, Chen & Chen	2015	296	1.545	Strong	psych	Stress	-	Med-proc	MA
Chang et al.	2008	236	1.255	Strong	psych	Stress	-	Med-proc	MA
De la Torre-Luque et al.	2017a	21	1.296	Strong	phys, psych	Stress, Anxiety	HR	Non-med	MA
De la Torre-Luque et al.	2017b	58	1.394	Strong	phys, psych	Stress, Anxiety	HR	Non-med	MA
DeMarco et al.	2012	26	-	Moderate	phys, psych	Stress, Anxiety	BP, HR	Surgery	MA
Di Nasso et al.	2016	100	2.807	Moderate	phys	Stress	BP, HR	Med-proc	MA
Doğan & Şenturan	2012	200	-	Strong	psych	Anxiety	-	Med-proc	MA
Doro et al.	2016	100	2.698	Moderate	psych	Anxiety	-	Med-proc	MT
Drzymalski et al.	2017	99	3.140	Moderate	phys	Stress	BP, HR	Med-proc	MA
El-Hassan et al.	2009	180	5.727	Weak	psych	Anxiety	-	Med-proc	MA
Elliott	1994	38	1.657	Strong	phys, psych	Stress, Anxiety	BP, HR	Med-proc	MA
Ghetti	2013	23	1.185	Moderate	phys, psych	Stress, Anxiety	BP, HR	Med-proc	MT
Ghezeljeh et al.	2017	92	0.801	Strong	psych	Anxiety	-	Med-proc	MA
Gomez-Urquiza et al.	2016	120	1.998	Moderate	phys, psych	Stress, Anxiety	BP, HR	Surgery	MA
Graversen & Sommer	2013	75	2.232	Weak	phys	Stress	Horm	Surgery	MA
Groener et al.	2015	35	1.555	Moderate	psych	Stress	-	Med-proc	MA
Gupta & Gupta	2015	60	1.900	Strong	phys, psych	Stress, Anxiety	BP, HR	Non-med	MA
Guzetta	1989	53	1.657	Strong	phys	Stress	HR	Med-proc	MA
Hamel	2001	101	1.326	Strong	phys, psych	Stress, Anxiety	BP, HR	Med-proc	MA

Authors	Year	N	Impact factor	Study quality	Type of measures	Type of outcome(s)	Physiological measure	Type setting	Intervention
Hammer	1996	16	1.000	Strong	phys, psych	Anxiety	-	Non-med	MT
Hamidi & Ozturk		100	2.270	Strong	phys, psych	Stress, Anxiety	BP, HR	Med-proc	MA
Han et al.	2017	137	1.255	Strong	phys	Stress, Anxiety	BP, HR	Surgery	MA
Horne-Thomson & Grocke	2010	25	2.023	Moderate	psych	Stress	HR	Non-med	MT
Hook et al.	2008	110	-	Strong	psych	Anxiety	-	Surgery	MA
Hsu et al.	2008	70	2.056	Moderate	psych	Anxiety	-	Med-proc	MA
Hayes et al.	2016	198	0.671	Weak	psych	Anxiety	-	Med-proc	MA
Jeppesen et al.	2003	143	-	Moderate	phys	Anxiety	-	Surgery	MA
Jiménez-Jiménez et al.	2016	40	0.211	Strong	psych	Stress	BP, HR, Horm	Surgery	MA
Johnson et al.	2013	119	0.662	Strong	phys	Anxiety	-	Surgery	MA
Kar et al.	2012	34	-	Moderate	psych	Stress	Horm	Surgery	MA
Ko et al.	2015	138	1.252	Weak	phys, psych	Anxiety	-	Med-proc	MA
Kunikullaya et al.	2017	88	0.922	Strong	phys	Stress, Anxiety	BP, HR, Horm	Med-proc	MA
Kushnir et al.	2015	60	1.264	Strong	phys, psych	Stress	BP, HR	Surgery	MA
Lai & Li	2012	54	1.741	Strong	phys, psych	Stress	BP, HR, Horm	Non-med	MA
Lai et al.	2011	44	1.255	Strong	phys	Stress, Anxiety	BP, HR	Med-proc	MA
Lai et al.	2008	60	1.814	Strong	phys	Stress	BP, HR	Non-med	MA
Latha et al.	2013	80	-	Moderate	phys	Stress	BP, HR	Non-med	MA
Leardi et al.	2014	60	5.596	Moderate	phys, psych	Stress	Horm	Med-proc	MA
Lee et al.	2007	64	1.585	Moderate	phys, psych	Stress	BP	Non-med	MA
Lee et al.	2016	140	1.427	Strong	phys, psych	Stress, Anxiety	HR, HRV	Surgery	MA
Lee, Lai, Sung et al.	2012	85	2.344	Strong	phys, psych	Stress, Anxiety	BP, HR	Med-proc	MA
Lee, Lee, Hsu et al.	2017	85	1.549	Strong	phys, psych	Stress, Anxiety	BP, HR	Surgery	MA
Lee, Wu, Lee et al.	2017	100	2.013	Weak	psych	Stress, Anxiety	BP, HR	Surgery	MA
Lin et al.	2017	98	1.384	Strong	phys, psych	Anxiety	-	Med-proc	MA
Lin et al.	2011	88	1.604	Moderate	psych	Stress	BP, HR	Med-proc	MA
Mahdipour et al.	2012	118	2.253	Moderate	psych	Anxiety	-	Med-proc	MA
López-Cepero Andrada	2004	150	-	Moderate	psych	Stress, Anxiety	-	Surgery	MA
Mandel et al.	2012	68	0.800	Weak	phys, psych	Stress, Anxiety	BP	Surgery	MT
Menegazzi et al.	2007	38	4.580	Weak	phys	Stress	BP, HR	Med-proc	MA
Ming et al.	1991	197	3.640	Strong	psych	Anxiety	-	Med-proc	MA
Miyata et al.	2016	42	-	Strong	phys, psych	Stress, Anxiety	HR	Med-proc	MA
Ni et al.	2016	172	1.384	Weak	phys, psych	Anxiety	BP, HR	Surgery	MA
Nilsson	2011	58	1.332	Strong	phys	Stress	BP, HR, Horm	Surgery	MA
Nilsson et al.	2009	75	3.634	Strong	phys	Stress, Anxiety	BP, HR, Horm	Surgery	MA

Authors	Year	N	Impact factor	Study quality	Type of measures	Type of outcome(s)	Physiological measure	Type setting	Intervention
O'Callaghan et al.	2005	97	1.182	Moderate	psych	Anxiety	-	Med-proc	MA
Padam et al.	2012	132	0.810	Strong	phys, psych	Stress, Anxiety	BP, HR	Med-proc	MA
Pothoulaki et al.	2017	60	1.748	Weak	psych	Anxiety	-	Med-proc	MA
Radstaak et al.	2008	123	3.473	Strong	psych	Stress	-	Non-med	MA
Romito et al.	2014	62	0.769	Strong	psych	Stress, Anxiety	-	Med-proc	MT
	2013						BP, HR		
Ripley et al.	2014	70	0.824	Moderate	phys	Stress	-	Med-proc	MA
Salehi et al.	2016	166	0.972	Strong	psych	Anxiety	-	Med-proc	MA
Sanal & Gorsev	2014	70	0.647	Weak	psych	Anxiety	-	Non-med	MA
Sendelbach et al.	2006	86	2.172	Moderate	phys, psych	Stress, Anxiety	BP, HR	Surgery	MA
Shabanloei et al.	2010	100	0.240	Moderate	psych	Anxiety	-	Med-proc	MA
Shin & Kim	2011	233	0.849	Moderate	psych	Stress, Anxiety	-	Med-proc	MA
Simavli et al.	2014	141	3.570	Moderate	psych	Anxiety	-	Med-proc	MA
Smith	2008	80	-	Weak	psych	Anxiety	-	Non-med	MT
Solomon & Ridgeway	2016	66	3.560	Moderate	psych	Anxiety	-	Med-proc	MA
Soo et al.	2016	80	2.993	Strong	psych	Anxiety	-	Med-proc	MA
Stein et al.	2010	56	0.622	Weak	psych	Stress, Anxiety	-	Surgery	MA
Su et al.	2013	28	1.917	Moderate	phys	Stress	BP, HR, Horm	Surgery	MA
Tabrizi et al.	2012	90	1.292	Weak	phys	Stress	BP, HR	Surgery	MA
Tan et al.	2015	100	1.158	Moderate	phys	Stress	-	Med-proc	MA
Thoma et al.	2015	92	2.130	Weak	psych	Anxiety	-	Med-proc	MA
Toker & Kömürcü	2017	70	2.013	Moderate	psych	Anxiety	BP, HR	Med-proc	MA
Triller et al.	2006	200	2.651	Moderate	phys	Stress	Horm	Med-proc	MA
Uedo et al.	2004	29	0.930	Moderate	phys	Stress	-	Med-proc	MA
Vachiramon et al.	2013	100	1.936	Strong	psych	Anxiety	-	Surgery	MA
Voss et al.	2004	61	5.836	Strong	psych	Stress, Anxiety	BP, HR	Surgery	MA
Wang et al.	2015	60	1.395	Strong	phys, psych	Stress, Anxiety	HRV	Surgery	MT
Warth et al.	2016	84	2.649	Strong	phys, psych	Stress	BP, HR	Med-proc	MT
White	1992	40	0.766	Strong	phys, psych	Anxiety	BP, HR	Surgery	MA
Wiwatwongwana et al.	2016	91	2.275	Strong	phys, psych	Stress, Anxiety	BP, HR	Surgery	MA
Yeo et al.	2008	70	1.710	Weak	phys	Stress	BP, HR, Horm	Med-proc	MA
Zengin et al.	2013	100	1.935	Weak	phys	Stress	-	Med-proc	MA
Zhang et al.	2014	124	0.680	Strong	psych	Anxiety		Med-proc	MA

Note: psych = psychological stress-related outcomes; phys = physiological stress-related outcomes; BP = blood pressure; HR = heart rate; Horm = hormone levels; HRV = heart rate variability; MA = music activity; MT = music therapy.

Table A2
Characteristics of included studies of the Meta-Analysis.

Author(s)	Year	N	Study Quality	Study Design	Type of Measures	Type of Outcome(s)	Physiological Measure(s)	Type of Setting
Aldridge et al.	2005	20	Weak	RCT	Psych	Anxiety	-	Medical HC
Argstatter et al.	2006	56	Moderate	RCT	Phys, psych	Stress,Anxiety	BP, HR	Medical HC
Bradt et al.	2014	62	Moderate	RCT	Psych	Stress	-	Medical HC
Browning	2001	20	Weak	RCT	Psych	Stress	-	Medical HC
Burns et al.	2008	19	Moderate	RCT	Psych	Anxiety	-	Medical HC
Cassileth et al.	2003	60	Moderate	RCT	Psych	Stress	-	Medical HC
Chang et al.	2018	49	Moderate	CCT	Psych	Anxiety	-	Mental HC
Chen et al.	2015	194	Strong	RCT	Psych	Anxiety	-	Mental HC
Choi et al.	2008	26	Moderate	CCT	Psych	Anxiety	-	Mental HC
Clark et al.	2006	63	Moderate	CCT	Psych	Stress	-	Medical HC
Crawford et al.	2013	38	Weak	RCT	Psych	Stress	-	Medical HC
Domingo et al.	2015	68	Weak	CCT	Psych	Anxiety	-	Medical HC
Dóro et al.	2016	100	Moderate	RCT	Psych	Anxiety	-	Medical HC
Erkkila et al.	2011	67	Strong	RCT	Psych	Anxiety	-	Mental HC
Ezegbe et al.	2018	280	Weak	CCT	Psych	Stress		Mental HC
Ferrer	2007	50	Weak	RCT	Phys, psych	Stress,Anxiety	BP	Medical HC
Gallagher et al.	2018	163	Weak	RCT	Psych	Stress	-	Medical HC
Ghetti	2013	22	Moderate	RCT	Phys, psych	Stress	BP, HR	Medical HC
Gold et al.	2013	79	Weak	RCT	Psych	Stress	-	Mental HC
Haack & Silverman	2017	19	Moderate	RCT	Psych	Anxiety	-	Medical HC
Hammer	1996	16	Strong	RCT	Psych	Anxiety	-	Mental HC
Hanser et al.	2006	42	Moderate	RCT	Psych	Anxiety	-	Medical HC
Hernandez-Ruiz	2005	28	Weak	CCT	Psych	Anxiety	-	Mental HC
Horne-Thompson & Grocke	2008	25	Moderate	RCT	Phys, psych	Stress,Anxiety	HR	Medical HC
Kim et al.	2011	18	Moderate	RCT	Psych	Stress	-	Medical HC
Mandel et al.	2007	68	Weak	RCT	Phys, psych	Stress,Anxiety	BP	Medical HC
McKinney et al.	1997	36	Moderate	RCT	Psych	Stress	-	Mental HC
Ming Wu	2002	24	Strong	CCT	Psych	Anxiety	-	Mental HC
Mohammadi	2011	19	Weak	RCT	Psych	Stress	-	Mental HC

Author(s)	Year	N	Study Quality	Study Design	Type of Measures	Type of Outcome(s)	Physiological Measure(s)	Type of Setting
Mondanaro et al.	2017	60	Strong	RCT	Psych	Anxiety	-	Mental HC
Morgan et al.	2011	49	Strong	CCT	Psych	Stress	-	Medical HC
Palmer et al.	2015	133	Strong	RCT	Psych	Anxiety	-	Medical HC
Pitts et al.	2015	10	Weak	RCT	Psych	Stress	-	Mental HC
Raglio et al.	2017	38	Moderate	RCT	Psych	Anxiety	-	Medical HC
Raglio et al.	2016	30	Moderate	RCT	Psych	Anxiety	-	Medical HC
Ribeiro et al.	2019	21	Strong	RCT	Phys	Stress,Anxiety	HR	Medical HC
Romito et al.	2013	62	Strong	RCT	Psych	Stress	-	Medical HC
Rosenow & Silverman	2014	18	Weak	RCT	Psych	Stress	-	Medical HC
Rossetti et al.	2017	78	Strong	RCT	Psych	Stress,Anxiety	-	Medical HC
Schmid & Aldridge	2004	20	Weak	CCT	Psych	Anxiety	-	Medical HC
Selle et al.	2017	36	Weak	RCT	Psych	Stress	-	Medical HC
Smith	2008	80	Weak	RCT	Psych	Anxiety	-	Mental HC
Teckenberg-Jansson et al.	2018	101	Strong	RCT	Psych	Stress	-	Medical HC
Verstegen & Silverman	2018	13	Moderate	RCT	Psych	Stress	-	Medical HC
Volpe et al	2018	106	Moderate	RCT	Psych	Stress	-	Mental HC
Warth et al.	2015	84	Strong	RCT	Phys, psych	Stress	HRV	Medical HC
Yates & Silverman	2015	22	Moderate	RCT	Psych	Stress	-	Medical HC

Note: psych = psychological stress-related outcomes; phys = physiological stress-related outcomes; Anxiety = state anxiety; HR = heart rate; HRV = heart rate variability; BP = blood pressure; medical HC = medical healthcare; mental HC = mental healthcare.

Table A3
Music Therapy Intervention Index

This index provides a more detailed description of the music therapy interventions mentioned by the music therapists of this study. Some interventions were only briefly mentioned, in which case we provided additional literature to explain the interventions.

Music therapy intervention	Description by the music therapists	Additional literature
Free improvisation	Free improvisation is mentioned across all three focus groups and described as a form of musical improvisation where no rules, restrictions or guidelines are imposed. The first step is to synchronize with the patient within the free improvisation. After reaching synchronization the music therapist can choose to change certain musical components to stimulate stress release or relaxation.	Improvisation and its various therapeutic uses and forms have been thoroughly described by Tony Wigram (2014) and Kenneth Bruscia (1987). One method worth mentioning is the Free Improvisation Therapy by Juliette Alvin (1975, 1976, 1978).
Freestyling	This intervention allows the patient to release tension by directly expressing their anger, fear and/or stress in music. The music therapist facilitates and guides this process. The patient chooses the type of beat, the instruments, etc. "*This can be very difficult for patients with an intellectual disability … when they are relaxed, they are capable of verbalizing their story in a great way … but when they just have had bad news for example, a whole bucketload of frustration tends to come out*".	Freestyle singing and rapping is used in music therapy settings to promote well-being through the expression and modulation of emotions, both positive and negative (Hadley & Yancy, 2012; Uhlig, Dimitriadis, Hakvoort, & Scherder, 2017).
6/8-meter improvisation	The music therapist and patient(s) play a continuous 6/8 rhythm at a pulse of approximately 80 bpm. Variations in tempo and dynamics are limited as much as possible. This is sustained for approximately 10 minutes. This intervention can be experienced as a very meditative and relaxing exercise. "*But at the same time, you turn inwards and come into contact with your emotions, some of which you might not want to be confronted with*".	Wigram (2004) describes the use of pulsed improvisation. Juslin, Liljeström, Västfjäll and Lundqvist (2010) describe the effect of rhythm and tempo on the emotional experience of music listening.
Interplay on piano	This intervention can be regarded as the beginning of an improvisation with the purpose to synchronize with the patient. The patient is asked to play their natural/intrinsic tempo on a single piano key. The music therapist accompanies their patient on the same piano, and the patient can be asked to choose between a major or minor key. "*This allows me to get a feel for the patient's state of mind at that moment*".	Wigram (2014) describes how to execute a one-note improvisation and the significance of pulse in musical improvisation. Juslin, Liljeström, Västfjäll and Lundqvist (2010) describe the process of *rhythmic entrainment*, where emotion is induced when the external rhythm of the music interacts with an internal body rhythm.
Expressing feelings in music	By using the musical components in a specific way, the patient tries to match the improvised music with their own feelings. This could be a way to express any negative emotions they are experiencing.	Symbolizing is an improvisational technique described by Bruscia (1987). It is the process of expressing personal feelings and experiences in music, to explore and modulate said feelings.

Music therapy intervention	Description by the music therapists	Additional literature
Taking turns	-	Taking turns (or turn-taking) refers to musical interactions in which the music therapist and patient take turns producing musical sounds. This reciprocal form of musical interaction depends on the fact that the patient can focus his/her social attention on his/her musical partner (Holck, 2002).
Mantra singing	Starting with just the voice, the music therapist sings mantras for or with the patient. The mantras can consist of just sounds, but a name can also be used as a mantra. "*Many patients have never sung their own name, but it's the most intrinsic thing you can do*". Looking to the pictures of loved ones while singing their names, can be a variation on this intervention. If needed, instrumental accompaniment van be added too.	The use of (improvised) vocal expressions to experience and express repressed emotions is a technique used in the music therapy method of therapeutic voice work (Austin, 2008; Baker & Uhlig, 2011).
Playing intrinsic tempo	This intervention uses the patient's intrinsic tempo as the basis for a group music improvisation. The intervention begins with every group member playing their intrinsic tempo on a drum, all at the same time. "*And so you get complete chaos, or at least you should*". Then, the music therapist implements structure into the improvisation. Once the improvisation has been structured, patients are asked to play their rhythm and the rest of the group mimics that rhythm. The patient is not only becoming aware of their intrinsic rhythm, but also how it feels to accommodate someone else's rhythm.	Wigram (2004) describes the use and importance of pulsed improvisation. Juslin, Liljeström, Västfjäll and Lundqvist (2010) describe the process of *Rhythmic entrainment*, where emotion is induced when the external rhythm of the music interacts with an internal body rhythm.
Greek Sirtaki	The music therapist plays a Greek Sirtaki on the accordion and patients play along with the rhythm on various percussion instruments. The pace is slowly increased until it culminates in shouting: "1.., 2.., 3.., 4.., HOPPA!". During playing the Greek Sirtaki the music therapist can increase or decrease the music tempo.	The use of pre-existing music provides a set musical framework which gives the patient a sense of predictability and structure (Baker & Wigram, 2005)
Singing/Playing well-known songs	Well-known songs, such as children's songs (nursery rhymes), folk songs and famous pop songs, are recognizable and provide a sense of structure. The musical preference of the patient and the symbolical meaning of a song can also come into play.	The lyrics and musical style of a well-known songs can have symbolic meaning to a patient (Aldridge, 2005; Austin, 2008; Baker & Uhlig, 2011). The use of pre-existing music also provides a set musical framework which gives the patient a sense of predictability and structure (Baker & Wigram, 2005).

Music therapy intervention	Description by the music therapists	Additional literature
Keeping opening song constant	A short song which the therapist sings with or for the patient at the beginning of every music therapy session. This can be a pre-existing song, or a composed song by the music therapist. "*It's a way of creating rituals, which adds to the amount of structure and predictability".*	-
Recording self-composed songs (and taking home)	Recordings of self-composed songs established during the music therapy sessions which can be taken home by the patient. The patient can use the recorded songs as relaxation material outside of the music therapy session.	The recording of composed songs can be part of the songwriting process (Baker & Wigram, 2005).
Songwriting	The method of songwriting was mentioned in combination with various goals. Songwriting can be used for short term stress release, by way of verbalizing frustrations, anger and fear within the music. However, it can also be offered as a form of exposure therapy for patients who suffer from long term stress as a result of difficult or traumatic experiences. "*The more you work on a song, the less triggering the difficult experience becomes".*	Songwriting as a music therapy method has various forms and goals, but is in essence the process of creating, practicing, performing and recording a song with the patient(s), during which the music therapist acts as a facilitator. (Baker & Wigram, 2005; Baker, 2015; Aasgaard and Ærø, 2016)
Hegi	Hegi's Method of Musical Components is mentioned as a theoretic framework by several music therapists.	Hegi (1988, 1998) developed the Music Therapy Method of Musical Components, based on principles of the Gestalt therapy. Hegi describes the diagnostic and therapeutic value of various musical components, such as tone, rhythm, melody, dynamics and form.
Schumacher	A multi-sensory approach in which music is combined with other sensory inputs, such as visualization on a screen, body movement, or the vibration of an instrument, to activate multiple senses at the same time and subsequently achieve (internal) synchronization.	Karin Schumacher (1994, 2001) uses a combination of musical expression, visual contact, physical touch, and body movement, to emulate the early mother-child relationship to promote emotional development.
Ronnie Gardiner Method	The Ronnie Gardiner Method uses symbols instead of musical notes. Every symbol represents a movement and a word which are executed and said to the beat of the music. Through this manner various areas of the brain are stimulated at the same time. Since the exercise requires a lot of concentration	More information on the Ronnie Gardiner Method, as well as links to various publications, can be found on the website "Ronnie Gardiner Method" (2015).

Music therapy intervention	Description by the music therapists	Additional literature
NMT-MACT	-	The Musical Attention Control Training (MACT) is one of the interventions of the Neurologic Music Therapy (NMT). The MACT described by Thaut and Gardiner (2014) provides "structured active or perceptive musical exercises involving precomposed performance or improvisation in which musical elements cue different musical responses to practice attention functions." The method focusses on three different aspects of attention control: focused attention (one task), sustained attention, and alternated attention.
One Note Symphony (Berman)	-	The One note symphony is a music therapy method by Albert Berman (2016). It is a group improvisation where patients are asked to play a single note on a broad range of instruments, creating a melody only through interaction and utilizing different rhythmic forms. With the help of this intervention, Berman tries to make group improvisation accessible to people who do not have complex musical skills and thus enable more interaction and connection between patients during musical improvisation.
Recording and/or creating personalized relaxation music	Personal playlists can be used by the patient outside of the music therapy session to achieve relaxation. These playlists can include pre-existing songs, recorded compositions by the patient and/or music therapist, and recorded improvisations made during the music therapy sessions.	Creating a personalized playlist for patients to induce relaxation is a form of receptive music therapy which can be used both during and outside of the music therapy session (Grocke & Wigram, 2007).

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Appendix 4 (Chapter 5)

Table A4
Overview of Stress Self-Report Measures Found

Self-Report Measure	Author(s) and publication date	Outcome	Target group	Exclusion reason
Beck Anxiety Inventory (BAI) *	Beck et al. (1988).	State anxiety	Adults and people with ID	
Cognitive Anxiety Scale-State	Weinstein & Smith (1987)	Stress	No information available	No scientific information available
Computerized Stress Inventory	Press & Osterkamp (1984)	Stress (stress levels and life satisfaction)	Adults	No scientific information available
Depression Anxiety Stress Scales (DASS) *	Lovibond & Lovibond (1995).	Stress / state anxiety	Adults	
Derogatis Stress Profile (DSP)	Derogatis (1980)	Stress	Adults	
Glasgow Anxiety Scale* (GAS-ID)	Mindham & Espie (2003)	State anxiety	"People with an intellectual disability" (age not specified)	
Hamilton Anxiety Rating Scale (HAM-A)	Hamilton (1959)	State anxiety	Adults with diagnosis of anxiety disorder	Developed for adults with a specific diagnosis
Index of Clinical Stress (ICS) *	Abell (1991)	Stress	Adults & youths age 12+	
Lifestress Inventory (LI) *	Bramston & Fogarty (1997)	Stress	Suitable for a wide range of people, including MID	
Pikunas Adult Stress Inventory (PASI)	Pikunas (1984)	Stress	Adults & youths age 16+	No scientific information available
Psychological Stress Measure (PSM-9) *	Tessier et al. (1990)	Stress	Adults	
Perceived Stress Questionnaire (PSQ) *	Levenstein et al. (1993)	Stress	Adult	
Perceived Stress Reactivity Scale (PSRS)	Schlotz et al. (2011)	Stress	Adults	Anticipatory & specific situations
Perceived Stress Scale (PSS) *	Cohen et al. (1983)	Stress	Adults	
Recovery-Stress-Questionnaires (RESTQ)	Kallus & Kellmann (2016)	Stress	Adults, separate version for adolescents and children	Specific situations: stress during recreational activities
Stress Arousal Checklist (SACL) *	Mackay et al. (1978)	Stress and arousal	Adults	
Self-Rating Anxiety Scale for adults with Intellectual Disabilities (SAS-ID) *	Zung (1971) ID version: Lindsay & Michie (1988)	State anxiety	Adults with ID	

Self-Report Measure	Author(s) and publication date	Outcome	Target group	Exclusion reason
Stress Evaluation Inventory (SEI)	Kulhavy & Dee-Burnett (1984).	Stress (career, family, and personal-social)	Adults	No sufficient scientific information found
Stress Overload Scale (SOS) *	Amirkhan (2012)	Stress	Adults	
Stress Response Inventory (SRI)	Koh et al. (2001)	Stress	Adults	No information on English version available
Stress Symptom Checklist (SSCL) *	Schlebusch (2004)	Stress levels	Adults	
Subjective Stress Scale (SSS)	Bramston & Bostock (1994)	Stress	Adults with ID	Updated version of SSS is included (LI)
State version of the State-Trait Anxiety Inventory (STAI-S) *	Spielberger (1981)	State anxiety	Adults	
Stress Analysis System (SAS)	Nelson et al. (1983)	Stress	No information found	No scientific information available
Subjective Units of Distress Scale (SUDS)	Wolpe (1969)	Stress	Adults, adolescents and children, including persons with ID	Not a standardized measure, but a flexible clinical and research outcome tool

* included in final analysis

Table A5
Characteristics of Included Studies of the Literature Review

Author(s)	Year	*N**	Study Design	Type of setting	Type of patients	Type of stress-related outcome	Intervention description	Active or Receptive	Specification of the Intervention	Instruments
Aldridge et al.	2005	20	RCT	Medical	Multiple Sclerose patients	State anxiety	Very detailed	Combination	Improvisation Listening pre-recorded music Playing/singing existing songs	Voice Nonspecific
Argstatter et al.	2006	83	RCT	Medical	Cardiac patients	State anxiety, stress, BP, HR	Detailed	Receptive	Listening pre-recorded music	-
Bauer et al.	2010	80	RCT	Medical	Pregnant women	Distress	Very detailed	Combination	Improvisation Listening live music Listening pre-recorded music Songwriting/composing *Complemented with:* Breathing exercises Progressive muscle relaxation	Guitar Other Nonspecific
Bradt et al.	2014	31	RCT	Medical	Cancer patients	Stress	Very detailed	Combination	Improvisation Listening live music Playing/singing existing songs Songwriting/composing	Percussion Voice
Browning	2001	20	RCT	Medical	Pregnant women	Stress	Limited details	Receptive	Listening pre-recorded music *Complemented with:* Progressive muscle relaxation Breathing exercises	-
Burns et al.	2008	30	RCT	Medical	Cancer patients	State anxiety	Very detailed	Receptive	Listening pre-recorded music	-
Cassileth et al.	2003	62	RCT	Medical	Cancer patients	Stress	Detailed	Combination	Improvising Listening live music Songwriting/composing	Nonspecific
Chlan et al.	2013	373	RCT	Medical	Patients with respiratory problems	State anxiety	Very detailed	Receptive	Listening pre-recorded music	-

Author(s)	Year	*N**	Study Design	Type of setting	Type of patients	Type of stress-related outcome	Intervention description	Active or Receptive	Specification of the Intervention	Instruments
Chang et al.	2018	49	CCT	Psychiatric day care centre	Schizofrenia, affective disorders	State anxiety	Very detailed	Combination	Listening live music Songwriting/composing *Complemented with:* Reflecting	Piano Percussion Nonspecific
Chen et al.	2015	200	RCT	Prison	Male inmates	State anxiety	Very detailed	Combination	Improvisation Listening pre-recorded music Music games Playing/singing existing songs Songwriting/composing *Complemented with:* Expressive painting	Piano Guitar Percussion Voice
Choi et al.	2008	26	CCT	Mental Health Care	Psychiatric patients	State anxiety	Very detailed	Combination	Listening to live music Playing/singing existing songs Songwriting/composing *Complemented with:* Physical relaxation	Piano Percussion Voice
Clark et al.	2006	63	CCT	Medical	Cancer patients	Stress	Detailed	Receptive	Listening pre-recorded music *Complemented with:* Progressive muscle relaxation Positive self-talk	-
Crawford et al.	2013	38	RCT	Medical	Organ transplant donors and recipients	Stress	Limited details	Combination	Improvisation Listening live music	Guitar Other Nonspecific
Domingo et al.	2015	68	CCT	Palliative Care	Advanced cancer patients	State anxiety	Detailed	Combination	Listening live music Playing/singing existing songs Songwriting/composing	Voice Guitar Percussion Other
Dóro et al.	2016	100	RCT	Medical	Stem cell transplantation patients	State anxiety	Limited details	Active	Improvisation Playing/singing existing songs	Voice Percussion Guitar

Author(s)	Year	*N**	Study Design	Type of setting	Type of patients	Type of stress-related outcome	Intervention description	Active or Receptive	Specification of the Intervention	Instruments
Erkkila et al.	2011	79	RCT	Research and training center	Depression	State anxiety	Detailed	Active	Improvisation	Piano Percussion
Ezegbe et al.	2018	280	CCT	Mental HC	Married couples	Stress	Very detailed	Active	Playing/singing existing songs Songwriting/composing	Voice Nonspecific
Fachner et al.	2012	79	RCT	Mental HC	Depression with comorbid anxiety	Anxiety	Limited details	Receptive	Listening pre-recorded music	-
Ferrer	2007	50	RCT	Medical	Cancer patients	State anxiety, stress, BP	Detailed	Receptive	Listening live music	Guitar Voice
Gallagher et al.	2018	163	RCT	Medical	Patients recovering from total knee arthroplasty	Stress	Very detailed	Combination	Improvising Listening live music Playing/singing existing songs Songwriting/composing *Complemented with:* Breathing exercises Progressive muscle relaxation	Voice Nonspecific
Ghetti	2013	37	RCT	Medical	Cardiac patients	Stress, BP, HR	Detailed	Active	Playing/singing existing songs	Percussion Guitar Voice
Gold et al.	2013	113	RCT	Prison	Prison inmates	Stress	Detailed	Combination	Improvisation Listening pre-recorded music Playing/singing existing songs Songwriting/composing	Nonspecific
Haack & Silverman	2017	29	RCT	Medical	Post-surgery recipients and donors	State anxiety	Detailed	Receptive	Listening live music	Guitar Nonspecific
Hammer	1996	16	RCT	Rehabilitation	Staff members and alcohol and drug addicts	State anxiety	Detailed	Receptive	Listening pre-recorded music *Complemented with:* Breathing exercises Relaxation instructions	-

Author(s)	Year	*N**	Study Design	Type of setting	Type of patients	Type of stress-related outcome	Intervention description	Active or Receptive	Specification of the Intervention	Instruments
Hanser et al.	2006	70	RCT	Medical	Women with advanced breast cancer	State anxiety	Very detailed	Combination	Improvisation Listening live music Playing/singing existing songs Songwriting/composing *Complemented with:* Breathing exercises Relaxation suggestions	Guitar Piano Voice Percussion Other
Hernandez--Ruiz	2005	28	CCT	Shelter	Abused women	State anxiety	Limited details	Receptive	Listening pre-recorded music *Complemented with:* Progressive muscle relaxation	-
Horne-Thompson & Bolger	2010	21	CCT	Medical	Patients with Amyotrophic Lateral Sclerosis / Motor Neuron Disease	State anxiety, HR	Limited details	Combination	Listening live music Listening pre-recorded music Playing/singing existing songs Songwriting/composing	Voice Nonspecific
Horne-Thompson & Grocke	2008	25	RCT	Palliative Care	Terminally ill patients	State anxiety, stress, HR	Detailed	Combination	Listening pre-recorded music Playing/singing existing songs	Voice Non-specific
Kim et al.	2011	18	RCT	Medical rehabilitation	Stroke patients	Stress	Very detailed	Active	Improvisation Playing/singing existing songs Songwriting/composing *Complemented with:* Using pictures, flowers, scents	Piano Percussion Other Voice
Mandel et al.	2007	103	RCT	Medical rehabilitation	Cardiac rehabilitation patients	State anxiety, stress, BP	Very detailed	Combination	Improvisation Listening pre-recorded music Music games Playing/singing existing songs Songwriting/composing *Complemented with:* Relaxation suggestions	Percussion Voice Other

Author(s)	Year	*N**	Study Design	Type of setting	Type of patients	Type of stress-related outcome	Intervention description	Active or Receptive	Specification of the Intervention	Instruments
McKinney et al.	1997	28	RCT	Mental Health Care	Healthy adults	Stress	Very detailed	Receptive	Listening pre-recorded music	-
Mohammedi	2011	19	RCT	Nursing home	Elderly	Stress	Very detailed	Combination	Improvisation Listening live music Playing/singing existing songs *Complemented with:* Movement interaction Relaxing activities	Percussion Voice Other
Mondanaro et al.	2017	60	RCT	Medical	Patients recovering from spine surgery	State anxiety	Detailed	Combination	Improvisation Listening live music Singing/playing existing songs *Complemented with:* Breathing exercises Reflecting	Percussion Voice Nonspecific
Morgan et al.	2011	60	CCT	Pychiatric ward	Psychosis	Stress	Very detailed	Active	Improvisation Playing/singing existing songs Songwriting/composing *Complemented with:* Writing poem	Piano Percussion Voice
Ogba et al.	2019	142	CCT	Universities	Nigerian students	Stress	Very detailed	Receptive	Listening live music Listening pre-recorded music *Complemented with:* Breathing exercises Physical relaxation	Percussion Voice Guitar Piano Other
Palmer et al.	2015	207	RCT	Medical	Women with breast cancer or potential risk	State anxiety	Very detailed	Receptive	Listening live music Listening pre-recorded music	Voice Guitar Piano Other
Pitts & Silverman	2015	10	RCT	Mental Health Care	Discharged psychiatric patients	Stress	Undetailed	Active	Music games	Nonspecific

Author(s)	Year	*N**	Study Design	Type of setting	Type of patients	Type of stress-related outcome	Intervention description	Active or Receptive	Specification of the Intervention	Instruments
Raglio et al.	2017	38	RCT	Post-acute stroke rehabilitation hospitalization	Stroke recovery patients	State anxiety	Very detailed	Active	Improvisation	Percussion
Raglio et al.	2016	30	RCT	Medical	Amyotrophic Lateral Sclerosis patients	State anxiety	Detailed	Active	Improvisation Songwriting/composing	Percussion Nonspecific
Ribeiro et al.	2018	21	RCT	Medical	Mothers of preterm infants	State anxiety, stress, HR	Very detailed	Receptive	Listening pre-recorded music *Complemented with:* Quiet reflection	-
Romito et al.	2013	62	RCT	Medical	Breast cancer patients	Stress	Very detailed	Combination	Listening pre-recorded music Playing/singing existing songs *Complemented with:* Creative journaling Using pictures Making collage of pictures	Voice Nonspecific
Rosenow & Silverman	2014	68	RCT	Medical	Patients recovering from bone marrow transplant	Stress	Detailed	Receptive	Listening live music	Voice Percussion Nonspecific
Rossetti et al.	2017	78	RCT	Medical	Cancer patients	State anxiety, stress	Very detailed	Receptive	Listening pre-recorded music Listening live music	Guitar
Schmid & Aldridge	2004	20	CCT	Medical	Multiple Sclerosis patients	State anxiety	Detailed	Combination	Improvisation Listening live music Listening pre-recorded music Playing/singing existing songs	Piano Percussion Other Voice
Selle & Silverman	2017	36	RCT	Medical	Cardiovascular disease patients	Stress	Detailed	Receptive	Listening live music	Guitar Nonspecific
Smith	2008	80	RCT	Workplace	Employees	State anxiety	Detailed	Receptive	Listening live music *Complemented with:* Progressive muscle relaxation Breathing exercises	Guitar

Author(s)	Year	*N**	Study Design	Type of setting	Type of patients	Type of stress-related outcome	Intervention description	Active or Receptive	Specification of the Intervention	Instruments
Teckenberg--Jansson et al.	2018	102	RCT	Medical	Pregnant women	Stress	Very detailed	Receptive	Listening live music	Other Voice
Verstegen & Silverman	2018	13	RCT	Medical	Patients undergoing bone marrow transplantation	Stress	Detailed	Receptive	Listening live music	Guitar Voice
Volpe et al.	2018	61	RCT	Psychiatric ward	Psychosis patients	Stress	Very detailed	Active	Improvisation Playing/singing existing songs Songwriting/composing	Percussion Voice Nonspecific
Warth et al.	2015	84	RCT	Palliative care	Severely ill patients	Stress, HRV	Very detailed	Receptive	Listening live music *Complemented with:* Mindfulness exercise Reflecting	Voice Other
Wu	2002	26	CCT	College	Taiwanese undergraduates	State anxiety	Detailed	Combination	Improvisation Listening pre-recorded music Playing/singing existing songs *Complemented with:* Body movement Muscle relaxation Creating stories Drawing Psychological drama	Voice Nonspecific
Yates & Silverman	2015	22	RCT	Medical	Cancer patients	Stress	Undetailed	Receptive	Listening live music	Nonspecific

Note: BP = blood pressure, HR = heart rate, HRV = heart rate variability. Instruments can be used by the music therapist and/or patient. Nonspecific: (more) instruments seem to have been used but are not specifically named.

Appendix 6 (Chapter 6)

Table A6
The Final Description of the Micro-Intervention

Micro-Intervention for Music Therapy

Title of intervention:	**Music therapy technique for direct stress reduction, based on changes in tempo and dynamics.**
Goal of intervention:	Reduction of tension and stress directly during the music therapy session.
Description of target population and field:	This micro-intervention can be considered a transdiagnostic music therapy technique, as it can be offered to all client populations in a variety of different clinical settings when stress reduction is needed during the music therapy session. Stress plays an increasingly important role in everyday life, which is particularly the case for vulnerable populations such as people with impaired cognitive functions, including people with mild intellectual disability (MID). This is due to the fact that people with MID are more likely to experience stress, and generally have less adequate coping strategies [39]. For these reasons, they may particularly benefit from a micro-intervention for stress reduction. We also expect potential in other client populations who experience stress, with or without cognitive impairments. The micro-intervention can be broadly applied within existing music therapy modules and treatment programs in both inpatient and outpatient healthcare settings. The micro-intervention contains an active and a receptive variant, allowing the music therapist to choose the form that best suits the needs and possibilities of the client.
Specific domain that is targeted or treated:	Almost everyone experiences stress from time to time. Stress can be caused and sustained by *personal factors* such as having high demands or poor time management skills [8, 63], *environmental factors* such as relationship problems or difficulties at work [55, 58, 62], and *behavioral factors* such as not setting boundaries and lacking adequate coping strategies [8, 67]. Long term stress can lead to psychopathology such as anxiety disorders, depression, addiction and burn-out [1, 45, 61, 62, 63], and physical health issues such as high blood pressure, cardiovascular diseases, insomnia, and weight gain or loss [4, 45, 61, 63]. In the short term, stress can lead to decreased concentration and difficulties in learning new information [61]. This reduces the ability to work towards the client's treatment goals. It is therefore important to reduce the stress and tension as quickly as possible so that the previously set treatment goals can be worked on with better results.
Applicable treatment phase of intervention:	The micro-intervention can be applied in every treatment phase. The micro-intervention can be applied during the music therapy session when the client indicates experiencing stress or tension, or when the client's perceived stress is observed by the music therapist. This micro-intervention is intended for all cases in which the stress is not desired and is a possible impediment to the therapeutic process. The intervention can also be used during the *observation/diagnostic phase* to gain more insight into the client's ability to regulate stress. Understanding the client's stress regulation skills can be important in designing the treatment plan and in gaining a better understanding of the client's needs. Notably, when the micro-intervention is used during the observation/diagnostic phase in clients with anxiety disorders or trauma-related issues, it is important to take into consideration that anxiety-triggering stimuli may not yet be fully identified. In some cases, it may be beneficial to allow the client's present stress to exist for a while so that the client can gain insight into their own stress experience. s

Specification of setting:	Individual: The choice for using the micro-intervention individually or in a group setting depends on the characteristics of the target group.	Group: When applied in a group the micro-intervention focuses on the stress level of one specific client, with the group acting as support (such as in "containment").		Number of participants: The optimal group size depends on the characteristics of the target group such as stimulus sensitivity.
Type of music intervention: (Cross out what does not apply)	Improvising	Recreating	~~Composing~~	Listening
Main instruments: (Cross out what does not apply)	~~Free choice~~	Voice	Instrument group	~~Band/pop~~
Music triggered mechanisms on the emotional brain: (Cross out what does not apply)	Brain stem reflex	Rhythmic entrainment	~~Evaluative conditioning~~	~~Emotional Contagion~~
	~~Visual imagery~~	~~Episodic Memory~~	Musical Expectancy	~~Aesthetic judgement~~
Function of music during intervention technique (physiology, neurology, emotional, social):	In physiology and medicine, the general definition of stress is introduced by Selye (1956): "Stress is a general activation reaction to a stimulus that could mean both a challenge (in a positive way) and a threat (in a negative sense)" [57]. The responses to stress can be categorized as physiological arousal and emotional responses [2, 33, 44]. Together, the underlying systems of these responses regulate stress and affect each other during stress [17, 35, 36]. <u>Effects of music on stress</u> Both music listening and music-making are strongly associated with stress reduction. Recent neuroscientific studies provide insights into how music therapeutic interventions may lead to stress reduction by affecting physiological arousal and emotional states related to stress. Scientific studies show that during and/or after offering music interventions, lower cortisol levels, lowered heart rate and decreased arterial blood pressure are measured [10, 26, 28, 30, 34, 40]. Research also shows that music can modulate activity in brain structures, like the amygdala, that are known to be crucially involved in emotional processes [7, 24, 26, 27, 32, 37]. This may suggest that music can affect stress-related emotional states, defined as subjective worry, anxiety, restlessness or nervousness [1, 13, 45, 46]. <u>Possible explanations for these effects</u> There are several possible explanations for the positive effects of music on the stress experience. Firstly, listening to pleasant music may have a positive influence on emotional valence, which can be explained by the degree of attraction that an individual feels towards a specific object or event, or in this case: a song or music piece [22, 25]. Music experienced as pleasant increases the intensity of emotional valence (the felt happiness), which has a stress-reducing effect [23, 49, 65]. An increased dopamine activity in the mesolimbic reward brain system has been shown to be associated with these feelings of happiness in response to high valence music [7, 52, 53, 66]. Secondly, listening to music can provide "distraction" from stress-increasing thoughts or feelings [6, 11]. The beneficial property of music to distract people from aversive states has been supported by short-term music interventions for acute stress reduction [20, 34]. Finally, empirical evidence shows that synchronization during music making, in which people jointly and simultaneously harmonize musical elements such as melody, rhythm and dynamics, evokes positive feelings of togetherness and bonding, and decreases stress levels [35, 60, 68]. Group music-making or singing together may result in social bonding, which may be explained by the release of the neurotransmitters endorphin and oxytocin [19, 60, 64]. These neurotransmitters play a role in the defensive response to stress [3, 18, 38].			

Contra-indications:	As described, the micro-intervention consists of an active and a receptive variant, allowing the music therapist to choose the type of intervention that best suits the client's needs. Since the micro-intervention has not yet been investigated in clinical practice, no contraindications can be identified at this stage. However, it is likely that the micro-intervention might be less suitable for: • Clients suffering from acute psychosis; • Clients who are highly sensitive to sound; • Clients who are hearing impaired or deaf For people with severe autism, advanced dementia, or severe cognitive impairment, the verbal exchange may be omitted after the micro-intervention.
Prerequisites and materials:	General: • A sound-isolated music therapy room (especially within clinical settings) • Chairs for the client and the music therapist. For the active variant: • A piano, guitar, or percussion instrument for the music therapist. • A sufficient selection of accessible/approachable musical instruments, allowing for changes in dynamics and tempo. For the receptive variant: • A piano or guitar for the music therapist. • Sheet music of the patient's preferred songs when needed.
<u>Active variant</u> Stepwise description of procedure:	1. The music therapist chooses to use the micro-intervention when the client indicates feeling stressed or tense, or when the therapist observes stress in the client [6]. The music therapist explains the micro-intervention and its purpose in clear language. The music therapist coordinates physical proximity in such a way that both the music therapist and the client have sufficient space, that there is a comfortable distance between them, and that eye contact can be made [11]. 2. The client and music therapist both choose an instrument [5] by which large dynamic variations can be made, such as a djembé, drum kit, piano or guitar. If this proves difficult, the therapist can suggest a number of instruments to choose from. If the music therapist chooses to use a harmonic instrument, the harmonies should be simple [12] and preferably played in the lower register [14]. 3. The music tempo and dynamics are adjusted to the current stress level of the client [9]. Before proceeding with the micro-intervention, the music therapist checks whether the intensity of the music matches the client's current level of stress. If this is not the case, the music therapist adjusts the intensity based on observations and–when possible–instructions from the client. 4. If the music matches the client's stress level, the volume and music tempo will be increased evenly, such as when releasing/discharging [3, 10]. The client has a leading role in building up the music tempo and dynamics, and also determines when the intensity has reached a peak and can be reduced again [5]. The music therapist actively guides the client in this part of the intervention and helps to evenly bring the tempo back to 60-80 bpm and the dynamics to silence [13]. 5. Afterwards, the music therapist and client discuss how the client experienced any tension during the intervention, and how the stress has changed after the intervention [7]. 6. The improvisation can be recorded and listened to [4] to reflect better, to indicate moments of tension and relaxation more easily, or to compare the intensity and course of the intervention in different sessions. 1. Music based on preferences, 2. Client chooses song, 3. Expressing emotion, 4. Recording music, 5. Client chooses intervention*, 6. Therapist chooses intervention, 7. Verbal processing of emotions, 8. Using familiar song, 9. Music based on emotional state, 10. Accelerating tempo, 11. Keeping appropriate physical distance, 12. Simplicity in harmony, 13. Slowing down tempo, 14. Lower tonal register. *Techniques that have been crossed out do not apply in this variant.*

Receptive variant Stepwise description of procedure:	The music therapist chooses to use the micro-intervention when the client indicates feeling stressed or tense, or when the music therapist observes stress in the client [6]. The music therapist explains the micro-intervention and its purpose briefly. The music therapist coordinates physical proximity in such a way that both the music therapist and the client have sufficient space, that there is a comfortable distance between them, and that it is easy to make eye contact [11]. The client chooses a song [1, 2, 5, 8]. If this proves difficult, the music therapist can suggest a few songs or let the client choose from a song book. The music therapist plays the (if necessary simplified) chords [12] of this song on guitar or piano, preferably in the lower register [14]. The client is invited to listen. The music tempo and dynamics are adjusted to the client's current stress level [9]. Before proceeding with the micro-intervention, the music therapist checks whether the intensity of the music matches the client's current level of stress. If this is not the case, the music therapist adjusts the intensity based on observations and–when possible–instructions from the client. The music therapist can then choose to further increase the intensity of the music [10] and then decrease it, or to let the intensity be equal to the client's stress level and gradually decrease the tempo to 60-80 bpm and the dynamics to silence [13]. Afterwards, the music therapist and client discuss how the client experienced any tension during the intervention, and how the stress level has changed after the intervention [7]. 1. Music based on preferences, 2. Client chooses song, 3. Expressing emotion, 4. Recording music, 5. Client chooses intervention*, 6. Therapist chooses intervention, 7. Verbal processing of emotions, 8. Using familiar song, 9. Music based on emotional state, 10. Accelerating tempo, 11. Keeping appropriate physical distance, 12. Simplicity in harmony, 13. Slowing down tempo, 14. Lower tonal register. *Techniques that have been crossed out do not apply in this variant.*
Therapeutic role and attitude:	While executing the micro-intervention, the therapeutic attitude of the music therapist is based on the index of Smeijsters (2014) [68]: Facilitating; • The music therapist provides a sufficient number of suitable instruments, a sound-isolated music therapy room and a safe atmosphere. • The music therapist adjusts the physical distance and proximity to the client. Structuring; • The music therapist explains the intervention and its purpose to the client in clear language. Supporting; • The music therapist actively guides the client in executing the active micro-intervention together and, at the same time, gives the client sufficient space to express their tension. Observing; • The music therapist remains alert throughout the intervention and observes the client's behavior. • The music therapist constantly adapts the music to the client. The appropriate therapeutic attitude and degree of directivity differs per target group and must always be attuned to the client or client group.
Possible subsequent interventions or variations:	Active variant: • The client also chooses a musical instrument for the music therapist. • At the beginning of the intervention the client plays their own stress level, to which the music therapist can then adjust. Receptive variant: • Asking the client to sing along. • Humming the lyrics instead of singing. • Playing the song instrumentally. After carrying out the micro-intervention, the rest of the session can be spent working towards the previously set treatment goals.

Theoretical and scientific foundations:	This intervention was developed based on both evidence from scientific literature and experiences from clinical practice. Data analysis has led to 14 effective and frequently used intervention components, namely: 1. Music based on preferences (*n* = 10, literature) 2. Client chooses song (*n* = 7, literature) 3. Expressing emotion (*n* = 5, literature) 4. Recording music (*n* = 4, literature) 5. Client chooses intervention (*n* = 4, literature)* 6. Therapist chooses intervention (*n* = 4, literature) 7. Verbal processing of emotions (*n* = 4, literature) 8. Using familiar song (*n* = 4, literature) 9. Music based on emotional state (*n* = 4, literature) 10. Accelerating tempo (*n* = 4, literature and focus groups) 11. Keeping appropriate physical distance (*n* = 4, focus groups) 12. Simplicity in harmony (*n* = 4, focus groups) 13. Slowing down tempo (*n* = 4, focus groups) 14. Lower tonal register (*n* = 4, focus groups) Please consult the article for more context and information on the method of data collection. As many as 10 of the 52 articles made use of music based on the musical preference of the client. Patient Preferred Live Music (PPLM) was particularly common. This is a form of receptive music therapy in which the therapist plays music that is preferred and/or chosen by the client. The objectives of PPLM are to reduce anxiety, pain and fatigue, provide relaxation and improve quality of life [59]. A study by Jiang, Rickson and Jiang (2016) showed that the musical preference of the client plays a crucial role in the relaxing qualities of music during stress. In addition, a 2013 study by Crawford, Hogan and Silverman found that clients also preferred listening to PPLM to a more active form of music therapy, such as a harmonica lesson. Offering moments of choice, in both the active and the receptive variant, gives the client a sense of direction and control, and can increase the client's autonomy [47]. The client chooses a song, an instrument or determines the course and intensity of the improvisation. Autonomy is important in stress reduction because its degree determines whether a stressor influences an individual's functioning [43, 48]. When someone is autonomous, this person is able to make choices that may help them cope with the stressful situation. It is therefore important to increase autonomy. Tempo and changes in tempo can affect different physiological and neurological responses, such as arousal, motor activity and motivation [50]. For example, a fast tempo can lead to increased nervous system activation, muscle tension and heart rate, while a slow tempo can lead to muscle relaxation and a lowered heart rate resulting in more relaxation [5, 9, 12, 25, 41]. This is also the starting point of the so-called Iso-principle that is applied in this micro-intervention and is defined as follows: [Iso-principle is] a technique by which music is matched with the mood of a client, then gradually altered to affect the desired mood state. This technique can also be used to affect physiological responses such as heart rate and blood pressure. (Davis, Gfeller, & Thaut, 2008) By first matching the music to the client and then gradually adjusting parameters such as tempo and volume, certain moods or physiological responses can be provoked or influenced. For example, the music therapist can tune in with a stressed client with a heart rate of 120 beats per minute (BPM) by playing music at a tempo of 120 BPM. After tuning in, the tempo can be reduced to the pulse of a resting heart rate, namely 60 BPM [29, 56]. *Because it is impossible for describing the micro-intervention to give the client complete freedom of choice in regard to the intervention being used, it was decided to offer other moments of choice such as choosing an instrument, a song and having a leading role in the active variant instead of the element "the client chooses the intervention".

Recommended test- or assessment tool:	The effect of this micro-intervention can be measured in several ways: Through self-report questionnaires: • Visual Analogue Scale Stress (VAS-S): the client marks the amount of tension felt on a 10 centimeter line before and after the intervention. For more information, see Lesage, Berjot, & Deschamps, 2012 [31]. • Perceived Stress Scale (PSS): the client indicates on a five-point scale to what extent they agree with the statements. For more information, see Cohen et al., 1983 [13]. Through physiological measurements: • Measuring heart rate (HR) and/or heart rate variability (HRV) using a wristband. • Measuring skin conductance with electrodes on the fingers or hand, or a wristband.
Developed by:	Martina de Witte, Anne Knapen, Susan van Hooren

Date:
March 23th 2021

Appendix:

Definitions from Juslin, P. N. (2013). From everyday emotions to aesthetic emotions: towards a unified theory of musical emotions. *Physics of life reviews, 10*(3), pp. 235-266.

Aesthetic Judgement	"Term used to refer to a subjective evaluation of a piece of music as art based on an individual set of subjective criteria."
Brain Stem Reflex	"A process whereby an emotion is induced by music because one or more fundamental acoustic characteristics of the music are taken by the brain stem to signal a potentially important and urgent event that needs attention. In music, this could involve sounds that are sudden, loud, or dissonant, or that feature accelerating patterns."
Emotional Contagion	"A process whereby an emotion is induced by a piece of music because the listener perceives the emotional expression of the music, and then 'mimics' this expression internally."
Episodic Memory	"A process whereby an emotion is induced in a listener because the music evokes a personal memory of a specific event in the listener's life."
Evaluative Conditioning	"A process whereby an emotion is induced by a piece of music simply because this stimulus has often been paired with other positive or negative stimuli. For example, a particular piece of music may have occurred repeatedly together in time with a specific event that always makes you happy, such as meeting your friends. Over time, through repeated pairing, the music itself will eventually arouse happiness, even in the absence of the friendly interaction."
Musical Expectancy	"A process whereby an emotion is induced in a listener because a specific feature of the music violates, delays, or confirms the listener's expectations about the continuation of the music."
Rhythmic Entrainment	"A process whereby an emotion is evoked by a piece of music because a powerful, external rhythm in the music influences some internal bodily rhythm of the listener (e.g., heart rate), such that the latter rhythm adjusts toward and eventually 'locks in' to a common periodicity."
Visual Imagery	"A process whereby an emotion is evoked in the listener because he or she conjures up inner images (e.g., of a beautiful landscape) while listening to the music."

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Samenvatting (Dutch summary)

Het doel van dit proefschrift is bij te dragen aan de wetenschappelijke kennis over muziekinterventies voor stress reductie. Om het hoofd te bieden aan de negatieve impact van stress, gebruiken miljoenen mensen ter wereld kalmerende medicatie, die vaak geassocieerd wordt met talrijke contra-indicaties en negatieve bijwerkingen. Daarom is juist de ontwikkeling van en onderzoek naar innovatieve en niet-farmacologische interventies voor stress vermindering hard nodig, vooral met betrekking tot patiëntpopulaties waarvan bekend is dat ze kwetsbaarder zijn voor stress en voor wie er nog steeds een schaarste is aan geschikte psychologische interventies. Mensen met een licht verstandelijke beperking (LVB) worden als zo'n populatie beschouwd, en vormen momenteel meer dan 40% van de psychiatrische patiënten in Nederland.

Muziekinterventies, waaronder receptieve interventies (de cliënt luistert naar de muziek) en actieve interventies (de cliënt is actief betrokken bij het maken van muziek), kunnen worden beschouwd als dergelijke niet-farmacologische interventies. Muziekinterventies kunnen worden aangeboden door een gekwalificeerde muziektherapeut in de context van muziektherapie, maar kunnen ook door de cliënt zelf worden toegediend of door andere zorgprofessionals worden aangeboden ("music medicine"). Aangezien de stress verlagende kwaliteiten van muziek in verband zijn gebracht met een breed scala aan positieve uitkomsten, worden muziekinterventies steeds vaker gebruikt om stress te verminderen in zowel de medische als de geestelijke gezondheidszorg. Bovendien is de ervarings- en actiegerichte benadering van muziekinterventies bijzonder goed geschikt voor de mogelijkheden en behoeften van mensen met cognitieve beperkingen, zoals mensen met LVB. Dit proefschrift beoogt daarom de wetenschappelijke kennis te vergroten ten aanzien van (a) de effecten van verschillende soorten muziekinterventies op stressgerelateerde uitkomsten, (b) hoe en waarom muziekinterventies kunnen leiden tot stress verminderende effecten, en (c) hoe de mate van stress accuraat kan worden beoordeeld, specifiek bij mensen die kwetsbaarder zijn voor stress, zoals mensen met LVB.

De eerste studie in dit proefschrift (Chapter 2) onderzocht bestaande empirische studies naar de effecten van muziekinterventies op zowel fysiologische en psychologische stressgerelateerde uitkomsten. Om de groeiende hoeveelheid empirisch onderzoek samen te vatten, werden twee multilevel meta-analyses van 104 RCT's, met in totaal 327 effect sizes en 9.617 deelnemers, uitgevoerd om de sterkte van de effecten van muziekinterventies te beoordelen. Door twee afzonderlijke multilevel meta-analyses uit te voeren, beoogde deze studie de effectgroottes van muziekinterventies op zowel fysiologische als psychologische stressgerelateerde uitkomsten te beoordelen. Daarnaast is er onderzocht welke uitkomst-, studie-, steekproef- en interventiekarakteristieken de sterkte van het

effect op fysiologische en psychologische stressgerelateerde uitkomsten modereerden. Overall vonden we een significant klein tot middelgroot effect van muziekinterventies op fysiologische stressgerelateerde uitkomsten (*d* = .380; 61 RCT's) en een middelgroot effect van muziekinterventies op psychologische stressgerelateerde uitkomsten (*d* = .545; 79 RCT's), wat betekent dat participanten in de muziekinterventiegroepen een grotere afname van stress lieten zien dan participanten in de vergelijkingsgroepen. Moderatoranalyses toonden vervolgens aan dat het type uitkomstmaat de effecten van muziekinterventies op stressgerelateerde uitkomsten modereerde. Er werden namelijk grotere effecten gevonden op uitkomstmaat hartslag, in vergelijking met de uitkomstmaten bloeddruk en hormoonlevels. Geen van de mogelijke moderatoren ten aanzien van de interventiekarakteristieken bleek significant te zijn, maar de resultaten toonden echter wel enkele opmerkelijke trends van de interventiekarakteristieken. Grotere effectgroottes werden gevonden in studies met een muziektempo van 60-80 bpm (beats per minuut) en in studies die slechts één enkele sessie aanboden in vergelijking met studies waarin meer dan één sessie plaatsvond. De resultaten toonden geen modererend effect voor de verschillende settingen waarin de muziekinterventie werd uitgevoerd, wat er op zou kunnen wijzen dat de effecten van muziekinterventies niet afhankelijk zijn van het type setting waar de interventie wordt aangeboden. Er kan worden geconcludeerd dat deze studie op hoog niveau bewijs levert dat muziekinterventies effectief kunnen zijn in het verminderen van stress en rechtvaardigt daarmee het toenemende gebruik van muziekinterventies in zowel de medische- als de geestelijke gezondheidszorgpraktijk.

De tweede studie (Chapter 3) toont een meta-analytische review die specifiek de effecten van muziektherapie interventies onderzoekt. Om de groeiende hoeveelheid empirisch onderzoek naar muziektherapie samen te vatten, werd een multilevel meta-analyse, met 47 studies, 76 effect sizes en 2.747 deelnemers, uitgevoerd om de effectgrootte van muziektherapie op zowel fysiologische als psychologische stress gerelateerde uitkomsten te beoordelen, en om potentiële moderatoren van de interventie-effecten te testen. Overall vonden we een significant middelgroot tot groot effect van muziektherapie op stressgerelateerde uitkomsten (*d* = .723; 47 RCT's/CCT's). Grotere effecten werden gevonden voor gecontroleerd klinisch onderzoek (CCT) in vergelijking met gerandomiseerd gecontroleerd onderzoek (RCT), wachtlijst-controlegroepen in plaats van groepen waarbij de gebruikelijke zorg (care as usual [CAU]) of andere stress verminderende interventies werden aangeboden, en voor studies die uitgevoerd werden in niet-westerse landen in vergelijking met westerse landen. De geselecteerde interventiekenmerken hadden geen statistisch significante invloed op de effectiviteit van muziektherapie. Er werden echter wel enkele substantiële verschillen in effectgroottes gevonden via moderatoranalyses die het conventionele significantieniveau niet haalden wegens gebrek aan statistische power, meestal veroorzaakt door een ongelijke verdeling van studies (en effectgroottes) over de moderatorcategorieën. Grotere effecten werden gevonden in studies die een groter aantal sessies boden, studies die muziek gebruikten met een tempo van 60-90 bpm, en studies die muziektherapie in een groepssetting aanboden. Deze studie levert bewijs dat muziektherapie

effectief kan zijn in het verminderen van stress en rechtvaardigt daarmee het toenemend gebruik van muziektherapie uitgevoerd door een gekwalificeerde muziektherapeut in zowel de praktijk van de geestelijke gezondheidszorg als in medische settingen.

De derde studie (Chapter 4) geeft meer inzicht in de praktijkkennis met betrekking tot hoe muziektherapeuten het stressniveau van hun cliënten met LVB verlagen tijdens muziektherapiesessies. Drie focusgroep interviews werden gehouden met muziektherapeuten uit verschillende landen en klinische instellingen in Europa, werkzaam met volwassenen met LVB (*N* = 13). De resultaten geven een overzicht van de meest gebruikte interventies voor stressvermindering, gespecificeerd naar therapeutische doelen, interventietechnieken, het gebruik van muziekinstrumenten, en gerelateerde therapeutische veranderingsfactoren ("change factors"). De bevindingen tonen aan dat er twee manieren te onderscheiden zijn om tot stressvermindering te komen, namelijk "spanning loslaten" of "directe ontspanning", welke tevens als twee verschillende therapiedoelen kunnen worden beschouwd. Het therapiedoel "synchroniseren" kan gezien worden als een subdoel omdat het vaak voorafgaat aan het werken aan de andere twee doelen. Daarnaast blijkt uit de resultaten dat muziektherapeuten weinig tot geen receptieve interventies (zoals muziek luisteren) gebruiken voor stressreductie, maar liever actieve interventies gebruikten, die vooral gebaseerd waren op muzikale improvisatie. Het tempo en de dynamiek van de muziek worden beschouwd als de belangrijkste muzikale componenten om stress te verminderen.

De vierde studie (Chapter 5) presenteert een scoping literatuur review om meer inzicht te verkrijgen in de validiteit en betrouwbaarheid van zelfrapportage stressmeetinstrumenten en hun geschiktheid voor mensen met LVB. Bevindingen uit de literatuur werden aangevuld met informatie uit een raadpleging van deskundigen vanuit zowel de klinische- als academische praktijk. Hoewel het gebruik van fysiologische metingen ("biomarkers") om de stressniveaus van mensen te beoordelen aanzienlijke gegroeid is binnen het bestaande onderzoek naar stress, is het geen vervanging voor zelfrapportagemetingen, aangezien de literatuur aantoont dat fysiologische en psychologische stressgerelateerde uitkomsten niet noodzakelijkerwijs direct aan elkaar gerelateerd zijn. Daarom vonden wij het belangrijk om bestaande zelfrapportage meetinstrumenten voor stress te evalueren en zo de kennis over stressmeting in het algemeen te vergroten. In totaal voldeden 13 zelfrapportage stressinstrumenten aan de inclusiecriteria, waarvan er drie speciaal ontwikkeld waren om stress bij volwassenen met een verstandelijke beperking te meten: de Glasgow Anxiety Scale for people with Intellectual Disability (GAS-ID), de Lifestress Inventory (LI), en de Self-rating Anxiety Scale for adults with Intellectual Disabilities (SAS-ID). Vijf van de gevonden zelfrapportage stressinstrumenten leken niet geschikt voor andere populaties dan de gemiddeld begaafde populatie, en op basis van de literatuur zouden vijf van hen mogelijk wel geschikt kunnen zijn (zie Chapter 5 voor meer details). De geraadpleegde deskundigen gaven de volgende belangrijke aanbevelingen, die zowel voor de klinische praktijk als voor toekomstig onderzoek een belangrijke leidraad kunnen vormen: (a) het gebruik van concrete en gemakkelijk te begrijpen woordenschat, eenvoudige grammatica en korte zinnen, (b) het gebruik van een kort tijdsbestek voor het

ophalen van informatie (< een week), (c) antwoordopties in Likert-schalen moeten beperkt worden tot drie, (d) een 'ik weet het niet' antwoordoptie moet worden opgenomen, (e) het gebruik van visualisaties om de betekenis van vragen en antwoorden te ondersteunen, en (f) het gebruik van vooraf uitgeschreven alternatieve bewoordingen als de respondent niet in staat lijkt om de vraag te begrijpen. De bevindingen van deze studie onderstrepen de noodzaak van voortdurende inspanningen om zelfrapportage stressmetingen van hoge kwaliteit en "LVB-gevoelig" te blijven ontwikkelen. Daarnaast draagt deze studie bij aan de toenemende wetenschappelijke kennis over zelfrapportage stressinstrumenten, die gebruikt kunnen worden in meer rigoureuze studie ontwerpen voor toekomstig onderzoek naar de effecten van muziekinterventies op stress reductie.

Het doel van het laatste onderzoek in dit proefschrift (Chapter 6) was het ontwikkelen van een muziektherapeutische micro-interventie voor stressreductie, gebaseerd op en stevig verankerd in zowel "evidence-based" kennis als "practice-based" kennis. Een muziektherapeutische micro-interventie kan gedefinieerd worden als een kort deel van een sessie waarin de muziektherapeut specifieke therapeutische technieken of stappen gebruikt om te werken aan specifieke doelen van de patiënt. Ondanks het feit dat micro-interventies van korte duur zijn, zijn ze systematisch beschreven en volgen ze een stapsgewijze aanpak die zowel gebaseerd is op recente theoretische modellen als op de meest recente wetenschappelijke evidentie. In de eerste fase werd een kwalitatieve analyse uitgevoerd van de interventiebeschrijvingen van 52 gecontroleerde uitkomststudies gefocust op muziektherapie voor stressreductie, wat uiteindelijk leidde tot 14 overkoepelende categorieën van interventiecomponenten. Vervolgens werden praktijkgegevens uit de eerder gehouden focusgroep studie geïntegreerd, wat uiteindelijk resulteerde in een gedetailleerde stapsgewijze beschrijving van zowel een receptieve als actieve variant van de muziektherapeutische micro-interventie. De Delphi-techniek werd toegepast om feedback te verzamelen op de ontwikkelde micro-interventie en om tot een groepsmening te komen, door een panel van 16 muziektherapeuten en onderzoekers samen te stellen en te bevragen. Deze procedure resulteerde in een verbeterde beschrijving van de muziektherapeutische micro-interventie voor stressreductie.

Overall kan worden geconcludeerd dat muziekinterventies in de vorm van zowel muziek luisteren als in de context van muziektherapie, gegeven door specifiek opgeleide en getrainde therapeuten, veel voordeel kunnen opleveren voor patiënten in zowel medische settingen als in de geestelijke gezondheidszorg. Muziektherapie studies lieten over het algemeen grotere effecten zien, hetgeen verklaard kan worden door de gepersonaliseerde, op maat gesneden aanpak van de muziektherapeut die specifiek is opgeleid om muzikaal af te stemmen op de behoeften van de patiënt. Deze effecten kunnen echter ook gedeeltelijk verband houden met bepaalde methodologische tekortkomingen of het type setting. Meer onderzoek is nodig, vooral bij patiëntpopulaties waarvan bewezen is dat ze kwetsbaarder zijn voor stress zoals mensen met cognitieve stoornissen. Wij suggereren dat verder onderzoek van de muziektherapeutische micro-interventie voor stressreductie uit Chapter 6 zich hier goed

voor leent. Aangezien de micro-interventie theoretisch onderbouwd is en stevig verankerd is in zowel evidence-based kennis als practice-based kennis, is het opportuun om zowel de directe effecten binnen een enkele sessie te onderzoeken, als de effecten na meerdere sessies waarin de micro-interventie werd toegepast. Verder zijn goed geoperationaliseerde concepten van stress, onderzoeksopzetten die consistent zijn met RCT-normen, valide en betrouwbare meetmethoden, en het opnemen van een secundaire onderzoeksvraag gericht op het identificeren van therapeutische factoren sterk aan te bevelen in toekomstige wetenschappelijke studies.

Curriculum Vitae

About the author

Martina de Witte (Heerenveen, 31-07-1981) started her studies in Creative Arts Therapies, specialism Music Therapy, at the HAN University of Applied Sciences in Nijmegen in 1999. After completion of her Bachelor degree (2003), she started working in clinical practice as a music therapist. For 14 years, she worked in several clinical settings, including mental healthcare institution Stevig: an expert treatment center for adult (forensic) patients with mild intellectual disabilities (MID) and psychiatric disorders. As a music therapist she specialized in improvisational music therapy for arousal- and stress regulation.

After obtaining her Master's degree (2013) in the Arts Therapies at Zuyd University of Applied Sciences te Heerlen, she has been working as a teacher in music therapy and research methods at HAN University in both the bachelor- and master program. Since 2014, she conducted several research projects, both as part of her doctoral research as well as being part of the Dutch national research center KenVaK (www.kenvak.nl), where creative arts therapy interventions are developed and examined in direct relation to clinical practice. Last years, she presented her research at national and international conferences focused on music interventions, music therapy, creative arts therapies, and the topic of stress.

In collaboration with research colleagues of Artez university, HAN University, and the University of Amsterdam, she yearly organizes an international summer course in research on music-based interventions. She recently collaborated with researchers from all over the world in the "Mechanisms project", which was focused on increasing scientific knowledge on the mechanisms of change in the creative arts therapies. She is a guest lecturer in the topic of music therapy and multilevel meta-analysis at the University of Amsterdam, SRH Hochschule Heidelberg, and Pompeu Fabra University Barcelona. In clinical practice, she serves on an ethics advisory board to advise directors of various practice settings on the quality and ethical considerations of initiated research.

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List of Publications included in this Dissertation

Chapter 2

de Witte, M., Spruit, A., van Hooren, S., Moonen, X., & Stams, G. J. (2020). Effects of music interventions on stress-related outcomes: a systematic review and two meta-analyses. *Health psychology review, 14*(2), 294-324. https://doi.org/10.1080/17437199.2019.1627897

Chapter 3

de Witte, M., Pinho, A. D. S., Stams, G. J., Moonen, X., Bos, A. E., & van Hooren, S. (2020). Music therapy for stress reduction: a systematic review and meta-analysis. *Health Psychology Review,* 1-26. https://doi.org/10.1080/17437199.2020.1846580

Chapter 4

de Witte, M., Lindelauf, E., Moonen, X., Stams, G. J., & van Hooren, S. (2020). Music therapy interventions for stress reduction in Adults with Mild Intellectual Disabilities: Perspectives from clinical practice. *Frontiers in Psychology*, 11. https://doi.org/10.3389/fpsyg.2020.572549

Chapter 5

de Witte, M., Kooijmans, R., Hermanns, M., van Hooren, S., Biesmans, K., Hermsen, M, Stams, G.J., & Moonen, X. (under revision). *The Validity and Reliability of Self-Report Stress Measures and their Suitability for Assessing Stress in Adults with Mild Intellectual Disabilities.*

Chapter 6

de Witte, M., Knapen, A., Moonen, X., Stams, G.J., & van Hooren, S. (under revision). *Development of a Music Therapy Micro-Intervention for Stress Reduction.*