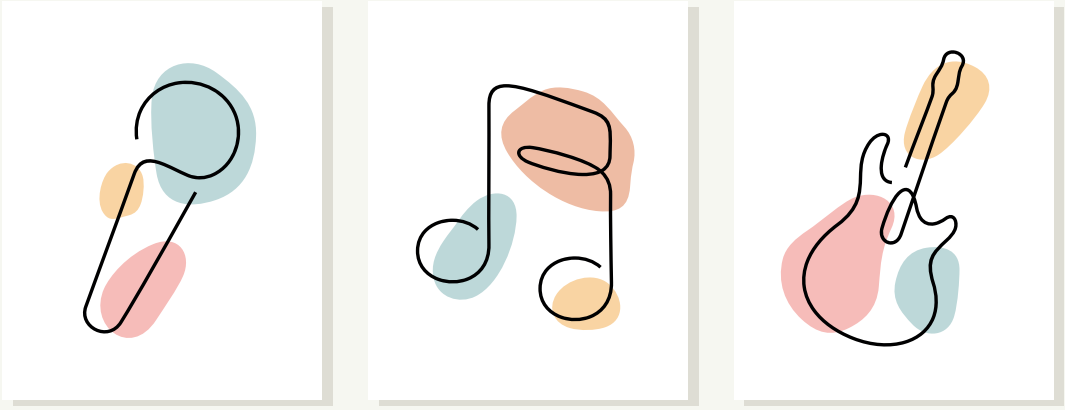
Connected by music

Research on music interventions with adults with intellectual disabilities



Gerianne Smeets

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COLOFON

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VRIJE UNIVERSITEIT

CONNECTED BY MUSIC

RESEARCH ON MUSIC INTERVENTIONS WITH ADULTS WITH INTELLECTUAL DISABILITIES

ACADEMISCH PROEFSCHRIFT

ter verkrijging van de graad Doctor of Philosophy aan
de Vrije Universiteit Amsterdam,
op gezag van de rector magnificus
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volgens besluit van de decaan
van de Faculteit der Gedrags- en Bewegingswetenschappen
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Dit boek draag ik op aan mijn vader, Jos Smeets, die zelf vele boeken las en dankzij wie onze boekenkast gevuld is. Wat had ik je graag ook nog dit boek overhandigd. Je was de meest lieve, vrijgevige, ruimhartige, integere en wijze persoon die ik ken, m'n moreel kompas en bent nog altijd *minge stille Copilot*. Elk woord doet jou tekort. Dank voor alles wie je was, waar je voor stond en wat je me hebt geleerd. Wat had ik graag nog meer van je geleerd en nog langer bij je mogen zijn.
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Kanns do mich sinn, wie ich he stonn?

Wie ich probiere, in dinge Schon wigger zo jonn.

Do fählst mir jede Daach.

Kasalla

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CHAPTER 1

1

Chapter 1

General introduction

Context

Prevalence of Intellectual Disabilities and the Prevalence and Impact of Challenging Behaviour

In the Netherlands, the prevalence of people with intellectual disabilities (ID)[1] is estimated at 1.45% (Cuypers et al., 2021). The degree of ID can be categorized as mild, moderate, severe, and profound ID, based on their intellectual and adaptive functioning (American Psychiatric Association, 2022). Within the population of people with ID, the prevalence of challenging behaviour (CB) is 18.1% (Bowring et al., 2017), which is higher compared to the general population (Dekker et al., 2002). CB can be defined as: "internalizing and/or externalizing behaviour(s) that is perceived to be socially or culturally undesirable in a specific context by the person and/or the environment, and of such an intensity, frequency or duration that it is detrimental, stressful or harmful for the person and/or the social environment" (Wolkorte et al., 2019, p. 1421). Potential explanations for the high prevalence of CB include a cumulative exposure to negative life events (Hatton & Emerson, 2004), community exclusion (Hastings et al., 2013), limited communication (Hastings et al., 2013) and social skills (Kearney & Healy, 2011). Consequently, CB can have a major negative impact on the quality of life (QoL) of people with ID (Bowring et al., 2019). In addition, caregivers and family members perceive the care for individuals with ID and CB as stressful (Maes et al., 2003; Raczka, 2005) and caring for people with CB is associated with increased burnout rates among caregivers (Deveau & McGill, 2019; Nevill et al., 2022; Shead et al., 2016). In sum, CB is a common problem in people with ID and not only has a negative influence on the QoL of the person with ID themselves but it also affects their environment.

Executive Functioning in People with Intellectual Disabilities

People with ID perform worse on executive functions (EF), which are effortful, cognitive processes necessary for executing goal-directed behaviour. These processes are required to adjust adequately to tasks in daily life (Diamond, 2013; Fidler & Lanfranchi, 2022). There is general agreement that there are three core EFs: inhibition, cognitive flexibility and working memory (Miyake et al., 2000). However, others (e.g., Goldstein et al., 2014) use a broader definition of EF which includes attention. People with ID have been found to perform poorer on separate EF tests than controls matched on mental age (Spaniol & Danielsson, 2021; Zagaria et al., 2021). More specifically, existing evidence suggests that people with ID perform worse on working memory (Van Der Molen et al., 2010), cognitive flexibility (Sgaramella et al., 2012), inhibition (Bexkens, Van der Molen, et al., 2014;

[1] Throughout this thesis, the term 'intellectual disabilities' is abbreviated, except in chapters 3, 4, and 6, in accordance with the author guidelines of the journals.

Bexkens, Ruzzano, et al., 2014; Sgaramella et al., 2012) and selective attention (Costanzo et al., 2013). In addition, research indicates that as the severity of ID increases, performance on EF tasks progressively declines (Memisevic & Sinanovic, 2014; Sgaramella et al., 2012) and aging leads to a more significant decline in EF for individuals with ID compared to those without ID (Memisevic & Sinanovic, 2014). Studies have also found an association between CB and EF; when people with ID show CB, they perform worse on EF tasks than people with ID without CB (Schuiringa et al., 2017; Van Rest et al., 2018; Visser et al., 2015). These findings underscore the critical role of EF in understanding and addressing the cognitive and adaptive challenges in daily life faced by people with ID, even more so when they also show CB.

Mental Health Challenges in People with Intellectual Disabilities

Besides the frequent CB and EF difficulties, persons with ID face other problems as well. They have a higher risk of developing mood disorders, such as anxiety and depression (Garaigordobil & Pérez, 2007; Nouwens et al., 2017). When adults with ID also have CB, they are more likely to have more psychiatric symptoms, especially depression (Hurley, 2008; Moss et al., 2000). People with ID also have smaller social networks (Van Asselt-Goverts et al., 2015) and are thus at increased risk for social isolation (Nouwens et al., 2017), which is associated with stress (Zwack et al., 2023). Therefore, it is not surprising that people with ID experience more stress in daily life (Zwack et al., 2023) and often use maladaptive coping skills to deal with stress (Hartley & Maclean, 2008). More stress is, in turn, correlated with more psychiatric disorders and CB in adults with ID (Scott & Havercamp, 2014). Furthermore, the self-esteem of adults with ID seems to be lower than in the general population (Garaigordobil & Pérez, 2007) and even lower when people with ID also have a depression (Dagnan & Sandhu, 1999). Overall, these findings suggest that people with ID face challenges related to mental health, smaller networks, more stress and low self-esteem.

Improving Mental Health and Social Interaction with Music Interventions

Self-esteem and EF have been shown to be positively associated with well-being, whereas CB and psychiatric symptoms are negatively associated with well-being (Kim et al., 2016; Koch et al., 2015; Luerssen & Ayduk, 2017). One way to reduce these inequities is by increasing social support which is a key contributor to health, well-being and overall QoL (Cohen & Wills, 1985; Lunsky & Benson, 2001; Uchino et al., 1996) and it decreases the odds of having a mental illness (Lunsky & Benson, 2001; Scott & Havercamp, 2014). Engagement with others as part of the community and structured activities promotes social well-being, as does creation and maintenance of reciprocal relationships (Eisenman et al., 2017). One potential strategy to improve social interaction and mental health is a

music intervention, which has also been shown to reduce stress. These promising benefits of music interventions will be further explored in the next section.

Characteristics of Music Interventions

Active vs. Receptive Music Interventions

Music interventions can include both active (making music or sounds together) and receptive (listening) intervention techniques (Chung & Woods-Giscombe, 2016; Theorell, 2014). It is believed that active methods generally provide greater individual benefits than passive engagement. Research shows this for example for CB in people with dementia (Gómez-Gallego et al., 2021) and for well-being in the general population (Weinberg & Joseph, 2017). Active techniques also seem to be most promising when targeting EFs (Haering, 2018). It does not seem to make a difference whether playing instruments is used or singing is performed (Bialystok & Depape, 2009). In addition, it is important that music interventions that are aimed at improving EF have 1) a long duration, 2) a sufficient length of individual sessions and enough spacing between the sessions, 3) a high level of in-person interaction and a supportive trainer and 4) meaningful activities that provide joy (Diamond & Ling, 2019). Furthermore, multi-activity music interventions, which for example involve improvisation, instrument playing and singing, show more significant results compared to other music interventions (Després et al., 2024).

Variability in Music Intervention: Length and Number of Sessions

In research the number of sessions can vary substantially, from 5 to 336 (Chung & Woods-Giscombe, 2016; Després et al., 2024; Fusar-Poli et al., 2017) and differ in time spans from one week up to 4 years (Geretsegger et al., 2014; Hennessy et al., 2019). In addition, in research the average duration of a music session is 30-60 minutes (Chung & Woods-Giscombe, 2016; Garrido, 2014; Geretsegger et al., 2022), with some sessions lasting only 12 minutes (Kossyvaki & Curran, 2020) and some two hours (Wilson & MacDonald, 2019). Music interventions range from daily sessions, particularly in a short-term format like one or two weeks, to weekly or biweekly (Bradt et al., 2016; Geretsegger et al., 2022). So, music interventions vary widely in terms of both the number of sessions and their duration.

Three Ways of Providing Music Interventions

The applications of music interventions in clinical settings can be divided into three categories with different intervention providers (Raglio & Oasi, 2015). The first refers to music therapy interventions, which are characterized by a relational component, which is considered essential, and is provided by

a qualified music therapist. The second category concerns music listening, which involves individualized music listening or listening to music based on a music medicine approach. The latter is provided by staff with a medical/therapeutic background who creates specific music listening programs for the receiver, which the client then self-administers. The third category includes general music-making interventions, which are centred on making music and listening to music for various purposes (i.e., improving mood, enhancing cognition) (Raglio & Oasi, 2015). General music-making interventions are often organised by a professional with music skills (Raglio & Oasi, 2015), which can be a music therapist, occupational therapist, professional musician or caregiver with musical expertise (Dorris et al., 2021). These kind of effective music interventions do not always need to be provided by a music therapist (Ito et al., 2022; Van der Steen et al., 2018). Most important in these kind of interventions is the approach and attitude of the music professional involved and the ability to play a musical instrument (Savarimuthu & Bunnell, 2002). This is further illustrated by the finding that caregiver-assisted music events and caregivers incorporating music-making into daily care can have beneficial effects (Gotell et al., 2000). It can even contribute to mutual emotional well-being and reciprocal relationships between the caregiver and the participant (Stuart-Röhm et al., 2023). In addition, training caregivers in live music interventions can reduce the burden of care and increase job satisfaction (Stuart-Röhm et al., 2023).

Individual vs. Group-based Music Interventions

Music interventions can be provided individually or in a group (Degé & Frischen, 2022). Research has shown that engaging with music in the company of others is associated with stronger positive experiences and higher well-being in the general population (Lamont, 1998; Weinberg & Joseph, 2017). However, there is also evidence that an individual music intervention, with personalized attention and targeted support, is more effective than a group intervention (Pelletier, 2004). Overall, whether the music intervention is provided individually or in a group, it is important to take care to provide positive learning experiences which are rewarding for the participant (Hallam, 2010).

(Partly) Digital Music Interventions

Recently, as a response to the COVID-19 restrictions, more attention is directed to (partly) digital music interventions (Dowson et al., 2021; Dowson & Schneider, 2021; Kantorová et al., 2021). For online music interventions, there are additional specific challenges pertaining to, for example, latency and variable sound quality (Dowson et al., 2021) and adaptions are needed, such as adapting the musical instruments or session times (Kantorová et al., 2021). In light of these challenges and adaptations, it is essential to collaborate with the participants to ascertain which approach suits

them best (Keenan & Doody, 2023). However, there are also benefits to online music interventions, such as easier access for clients in remote locations (Kantorová et al., 2021). Overall, it seems that the benefits of digital music interventions outweigh the challenges and should be seen, at least in pandemic times, as a necessary short-term intervention (Dowson et al., 2021; Kantorová et al., 2021).

Research on Music Interventions in the General Population

For several decades, researchers have studied the positive relation between music, health and well-being in the general population (e.g., Dingle et al., 2021; Macdonald, 2013; Raglio & Oasi, 2015; Weinberg & Joseph, 2017). Indeed, music can positively contribute to a flourishing life with greater psychological and emotional well-being (Croom, 2012, 2015; Van Goethem & Sloboda, 2011) and can have considerable benefits for cognition, behaviour, emotion, emotion regulation (Cook et al., 2019; Rentfrow, 2012). Experimental studies show that music interventions can improve the QoL (McCrary et al., 2022), can lead to stress reduction (De Witte, Spruit, et al., 2020) and can reduce depression and anxiety (Leubner & Hinterberger, 2017; Lu et al., 2021; Tang et al., 2020). In line with this, the potential of music to influence mood is described as one of the most important functions of music (Van Goethem & Sloboda, 2011), which is supported by the fact that music can evoke activity changes in the core brain regions responsible for emotions (Koelsch, 2014; Koelsch & Stegemann, 2012). The idea is well established that an improvement of the emotional state induced by music can, in turn, facilitate better performance on a wide range of cognitive tasks (Schellenberg & Weiss, 2013). In sum, the extensive body of research underscores the music's powerful role in enhancing psychological, emotional, and cognitive outcomes, making it a valuable tool for improving one's well-being and mental health.

Typically Developing Children

Multiple studies explored the effect of an active music intervention on various outcome measures in typically developing children. One review concluded that active engagement with music in children and young people had positive effects on their personal and social development, including self-confidence (Hallam 2010). A meta-analysis showed that music-based interventions improve depression and anxiety in children and adolescents with depressive or anxious symptoms (Geipel et al., 2018). With regard to EF, recent systematic reviews found that an active music intervention can enhance EF in children (Degé & Frischen, 2022; Rodriguez-Gomez & Talero-Gutiérrez, 2022), however they included RCTs with and without active control groups. Other studies also did not find a significant effect on EF, when a music intervention was compared with an active control group

(Bowmer et al., 2018; D'Souza & Wiseheart, 2018; Kosokabe et al., 2021). When we look at specific EF components, music interventions improve inhibition (Bugos & DeMarie, 2017; Frischen et al., 2021; Jaschke et al., 2018; Shen et al., 2019), attention (Frischen et al., 2021; Kasuya-Ueba et al., 2020) and working memory (Frischen et al., 2021; Shen et al., 2019), but not always cognitive flexibility and planning where results are mixed (Frischen et al., 2021; Jaschke et al., 2018; Kosokabe et al., 2021; Shen et al., 2019). So, while active music interventions show promise in enhancing various aspects of personal and social development, including self-confidence, depression and anxiety, the effects on EF in typically developing children remain mixed.

Research Addressing Music and Executive Functioning

A specific cognitive ability that is impacted by musical activities is EF (Altenmuller & Schlaug, 2012). In general, neuroimaging results demonstrate that music has the power to entrain neural activity in several brain circuits, including the prefrontal cortex (Carlson et al., 2015; Ferreri et al., 2013), which is associated with EF (Diamond, 2013; Miller & Cohen, 2001). The already mentioned four different EF components are all relevant when making music: 1) inhibition is required, for example to stop playing a known rhythm when the beat changes (Degé & Frischen, 2022; Frischen et al., 2021), 2) cognitive flexibility plays a crucial role during instructions, 3) remembering musical pieces requires the engagement of both visual and auditory working memory (Frischen et al., 2021) and 4) selective attention is needed to listen carefully to the sound of another person making music (Degé & Frischen, 2022). Involving EF while making music could lead to a far transfer effect: EFs required for active music making, such as control, attention, and memory, can have broader benefits for other high-level cognitive activities, enhancing cognitive abilities beyond the musical domain (Moreno et al., 2011). Yet other studies have yielded inconsistent and weak evidence for far transfer of music to other domains, meaning that improvements in EF that are not trained directly do not seem to generalize to untrained tasks or domains (Hogan et al., 2023; Sala & Gobet, 2017). Diamond and Ling (2019) also conclude that benefits of EF training methods in general are fairly specific and rarely transfer to untrained skills. However, they still propose that musical activities would be particularly effective in influencing EF.

Music Interventions in Clinical Populations

Dementia

Several empirically valid studies, reviews and meta-analyses have been carried out with participants who have other cognitive or communication disabilities, such as dementia or autism spectrum disorder (ASD). With regard to people with dementia, the importance of music interventions has

been internationally acknowledged in several systematic reviews and meta-analyses. For example, it has been found that music interventions improve QoL (Vasionyte & Madison, 2013), depression (Gómez-Romero et al., 2017; Ting et al., 2023; Van der Steen et al., 2018), anxiety (Ting et al., 2023) and mood (Guétin et al., 2009; Ing-Randolph et al., 2015; Sung et al., 2010, 2012). Music interventions can also diminish behavioural problems (Gómez-Romero et al., 2017; Van der Steen et al., 2018) and agitation (Duff-Woskosky et al., 2020; Gómez-Romero et al., 2017; Pedersen et al., 2017; Ting et al., 2023). There is evidence from systematic reviews that cognitive functions improve after a music intervention (Dorris et al., 2021; Duff-Woskosky et al., 2020; Moreno-Morales et al., 2020). The evidence specific to EF is, however, ambiguous, since one recent meta-analysis found an effect (Ito et al., 2022), while another meta-analysis did not (Fusar-Poli et al., 2017). In general, the effects of a music intervention go beyond the improvement of behavioural, psychological or cognitive symptoms in people with dementia (McDermott et al., 2014) and can help to create a meaningful care environment (Melhuish et al., 2017).

Autism Spectrum Disorder

It is also interesting to look at what is known about music interventions for people with ASD. Social functioning is a common outcome measure in this target group with no fewer than six systematic reviews showing that music improves their social functioning (Gassner et al., 2022; Geretsegger et al., 2022; James et al., 2015; Ke et al., 2022; Marquez-Garcia et al., 2022; Mayer-Benarous et al., 2021). There is also evidence that a music intervention leads to an increase in self-esteem (Hillier et al., 2011), EF (Sharma & Mehta, 2024), mood (Shi et al., 2016) and QoL (Geretsegger et al., 2022) and a reduction of anxiety (Hillier et al., 2011). In sum, music interventions have shown promising effects on various outcome measures for people with ASD, highlighting their potential as a valuable intervention for this population.

Music Intervention in People with Intellectual Disabilities

Music Interventions in People with Different Severity or Aetiology of Intellectual Disabilities

Music has been studied among specific groups of people with ID, such as people with severe or profound ID (Johnels et al., 2023; Oldfield & Adams, 1990; Rushton et al., 2023), children with Down's syndrome (Gemma et al., 2020; O'Donoghue, 2017), people with William's syndrome (Dunning et al., 2015; Thakur et al., 2018) and people with ID and ASD (Kossyvaki & Curran, 2020; Lundqvist et al., 2009). All studies reported positive findings on a range of measures (Dunning et al., 2015; Gemma et al., 2020; Johnels et al., 2023; Kossyvaki & Curran, 2020; Lundqvist et al., 2009; O'Donoghue, 2017; Oldfield & Adams, 1990; Rushton et al., 2023; Thakur et al., 2018). For example,

Oldfield and Adams (1990) found that music was effective at drawing attention and evoking appropriate social behaviour among adults with a profound ID and Lundqvist et al. (2009) concluded that vibroacoustic music reduced CB in adults with ID and ASD. Overall, musical benefits are reported for people with severe or profound ID, children with Down's syndrome, people with William's syndrome and people with ID and ASD. When considering people with mild to moderate ID, categories that encompass the majority of individuals with ID (The National Academies of Sciences, Engineering, and Medicine, 2015), it is equally important to explore how music can benefit this group.

Non-experimental Studies among People with Mild or Moderate Intellectual Disabilities

Non-experimental studies that investigated music interventions in people with mild to moderate ID found music to be associated with a better QoL (Andring, 2015; Jaschke & Scherder, 2015; Morris, 2014), higher self-esteem (Lewis, 2019; MacGlone et al., 2020; Wilson & MacDonald, 2019), better mood (Jackson, 2007; MacGlone et al., 2020), and less CB (Jaschke & Scherder, 2015). Music therapists also say that they often use active techniques to reduce stress in this population (De Witte, Lindelauf, et al., 2020). Regarding musical experiences in clinical practice, caregivers note that music activities help to increase the interaction between staff and peers (Jackson, 2007). However, in addition to adopting non-experimental designs, the studies exploring these associations predominantly utilized qualitative designs (Jackson, 2007; MacGlone et al., 2020; Wilson & MacDonald, 2019) with some not being peer-reviewed (Andring, 2015; Lewis, 2019; Morris, 2014). In sum, non-experimental studies show significant associations with multiple other measures in people with ID, but these studies do not examine the effectiveness of music interventions for individuals with ID.

Experimental Studies among People with Mild or Moderate Intellectual Disabilities

There are relatively few experimental studies on the relationship between music and ID compared to the amount of non-experimental literature among people with ID (Hooper et al., 2008; Mino-Roy et al., 2021). Experimental studies that were performed show promising effects on communication skills (MacDonald et al., 1999; MacDonald & O'Donnell, 1994), CB (Kalgotra & Warwal, 2017), attention (Bentley, 2015; Sussman, 2009) and social skills (Duffy & Fuller, 2000). However, one study was not peer-reviewed (Bentley, 2015) and one study did not incorporate a RCT design (Kalgotra & Warwal, 2017).

Review Studies among People with Mild or Moderate Intellectual Disabilities

There are also some review studies about music interventions for people with ID. These reviews show also promising effects on communication skills (Després et al., 2024; Mino-Roy et al., 2021; Savarimuthu & Bunnell, 2002), CB (Mino-Roy et al., 2021; Savarimuthu & Bunnell, 2002), cognition, including EF and attention (Després et al., 2024; Gold et al., 2004; Mino-Roy et al., 2021) and social skills (Duffy & Fuller, 2000; Mino-Roy et al., 2021). In addition, these reviews found positive effects on anxiety (Després et al., 2024; Mino-Roy et al., 2021; Savarimuthu & Bunnell, 2002), well-being (Savarimuthu & Bunnell, 2002), emotion regulation, self-esteem, autonomy and QoL (Mino-Roy et al., 2021). However, three of the four reviews did not distinguish between experimental and non-experimental studies (Després et al., 2024; Mino-Roy et al., 2021; Savarimuthu & Bunnell, 2002), which limits the ability to establish cause-and-effect relationships, and one of these review studies stated that the studies were reported to be of poor quality with low levels of evidence (Mino-Roy et al., 2021). In addition, the other review included children and adolescents with psychopathology (among which children with developmental disorders) (Gold et al., 2004). In sum, despite the relatively limited amount of experimental studies and its limitations, the research on music interventions for people with ID show promising outcomes.

Research Gaps and Methodological Constraints

Most studies addressing people with ID are about music therapy (Brown & Jellison, 2012; Murphy & McFerran, 2017), mostly conducted by a certified music therapist (Mino-Roy et al., 2021). Furthermore, research addressing people with ID tends to investigate passive forms of music rather than active music interventions (Brown & Jellison, 2012; Murphy & McFerran, 2017), although recently, there seems to be an increase in active approaches (Després et al., 2024; Mino-Roy et al., 2021). In addition to these research gaps, there are also methodological constraints in studies concerning the ID population that complicate firm conclusions about the effectivity of the intervention and align with constraints identified in the general population (among which typically developing children) and in other clinical groups. These include the absence of control groups, especially of an active control group (Bowmer et al., 2018; Duffy & Fuller, 2000; Jacob et al., 2021; McCrary et al., 2022; Vink et al., 2013), small sample sizes even in RCTs (Fusar-Poli et al., 2017; Geipel et al., 2018; Haslam et al., 2022; Marquez-Garcia et al., 2022) and no follow-up assessments (Després et al., 2024; Geipel et al., 2018; James et al., 2015; Marquez-Garcia et al., 2022; Mino-Roy et al., 2021). In addition, it is important to incorporate in research a combination of assessment methods, for example proxy-reports, self-reports and neuropsychological tasks to get a more accurate, complete and reliable assessment of the person's cognitive, emotional, and behavioural functioning

(Marquez-Garcia et al., 2022). Furthermore, the type, duration and intensity of music interventions are very diverse among studies (Degé & Frischen, 2022; Marquez-Garcia et al., 2022; McCrary et al., 2022; Rodriguez-Gomez & Talero-Gutiérrez, 2022; Ye et al., 2021), which makes it challenging to assess the influence of these parameters. Given the promising findings of music interventions in the general population and in clinical populations on CB, mental health, self-esteem, well-being and EF, there is a need for robust research to study the effects of an active music intervention for adults with ID on these outcome measures, incorporating a control condition, follow-up measurements and both iproxy and self-report measures.

Aim and Research Questions

This research adds to the existing literature concerning music interventions for adults with ID and points to ways in which music workers and care staff can share skills to work together more effectively. We intentionally move away from a focus on music therapy and instead focus on broader music interventions, carried out by music workers and caregivers. The overall research adopts a mixed-method approach, combining quantitative and qualitative data.

This thesis aims to investigate the effects and the feasibility of music interventions for people with ID who live at residential facilities in the Netherlands. To achieve this, this thesis seeks to answer the following research questions:

Part 1: Pilot cluster-RCT

What are the effects and feasibility of a weekly music group intervention compared to leisure group activities (chapters 2 and 3)?

Part 2: COVID study

What are the feasibility, experiences and short-term outcomes of a partly digital music intervention twice a week (chapter 4)?

Part 3: Cluster-RCT

What are the effects and feasibility of an individual music intervention twice a week compared to care-as-usual (CAU) (chapters 5, 6, 7 and 8)?

Outline of This Thesis

This thesis consists of nine chapters, including a general introduction (**chapter 1**) and a general discussion (**chapter 9**). The chapters 2 to 8 report on the studies performed. Given the many challenges in conducting an RCT with people with ID living in different residential settings, a pilot study was performed first. This pilot cluster-RCT including 29 adults with ID studied the effectiveness and feasibility of a weekly music group intervention compared to a weekly leisure group activities intervention. **Chapter 2** presents the effectiveness of this pilot cluster-RCT on well-being, self-esteem, social acceptance, CB and EF of people with ID. **Chapter 3** focuses on the feasibility of this pilot trial, by the use of six well-known areas of feasibility, i.e., acceptability, demand, implementation, practicality, adaptation, and limited efficacy. This study provides an insight into several challenges when performing a music group intervention and a leisure group intervention for people with ID living in several residential settings. **Chapter 4** reports on the feasibility, experiences and short-term outcomes on the CB and well-being in people with ID involving a partly digital music intervention, that was conducted during the COVID-19 pandemic. Ten participants were involved in live or digital individual music sessions twice a week. The proposed modifications in research design that emerged from chapter 3 are taken into account in the study protocol for a full-scale cluster-RCT, including plans for evaluation (**chapter 5**). The performed cluster-RCT, which included 97 participants with ID living in several residential settings who were, per residential setting, randomized into an individual music intervention or CAU, is described in three chapters. **Chapter 6** describes the effects of this intervention on CB, self-esteem, well-being, anxiety and depression in the participants with ID involved. **Chapter 7** describes the effects on EF in the participants with ID involved. **Chapter 8** explores the benefits and challenges associated with the participation in the cluster-RCT, the implementation of the music intervention, the experiences of those participating in the music intervention and the sustainability of the intervention, by use of a process evaluation with input from participants, caregivers and music workers. The general discussion (**chapter 9)** provides an overview of the main findings, key points and strengths and limitations of the current thesis. Furthermore, implications for clinical practice and future research recommendations are discussed.

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PART I



Part I

Pilot cluster-RCT

CHAPTER 2

2

Chapter 2

Active music making and leisure activities for people with intellectual disabilities: a cluster randomized pilot study

Smeets, G. J. J., Volkers, K. M., Scherder, E. J. A., & Moonen, X. M. H. (2023). Active music making and leisure activities for people with intellectual disabilities: A cluster randomized pilot study. *Research in Developmental Disabilities*, 142, 104606. doi:10.1016/j.ridd.2023.104606

Abstract

Aim: A pilot cluster randomized controlled trial was performed to evaluate the effects of a music group intervention and a leisure activities group intervention on well-being, self-esteem, social acceptance, challenging behaviour, and three components of executive functions (inhibitory control, cognitive flexibility and visual working memory) of adults with intellectual disabilities (ID).

Method: Twenty-nine participants living in four residential facilities for people with ID were randomized into a music or a leisure activities group intervention. Sixteen weekly sessions of one hour were performed with six to eight participants per group. Measurements were carried out at baseline, post-intervention and 11 weeks follow-up. Results were analysed with linear mixed models.

Results: The leisure activities group intervention improved more than the music group intervention on internalizing behaviour overall and on inhibitory control at post-intervention. However, further analyses showed that this improvement was only significant in one of the two leisure activities facilities.

Conclusion: A leisure activities group intervention seems a promising intervention to improve internalizing behaviour and inhibitory control of younger adults with mild ID. Implications for the delivery of leisure activities group interventions, and especially music group interventions, are discussed.

Introduction

People with intellectual disabilities (ID) are more likely to show challenging behaviour (CB), such as aggression, self-injury, and destructive behaviour (Emerson et al., 2001). They also experience lower self-esteem than people without ID (Garaigordobil & Pérez, 2007; Valås, 1999) and lack contact with peers (Nouwens et al., 2017). Their executive functioning (EF) is also impacted (Spaniol & Danielsson, 2021; Zagaria et al., 2021), which is not surprising, since EF is linked to intellectual functioning (Miyake et al., 2001). EF is a collective term for an assembly of interrelated higher cognitive processes and is often divided into three components: cognitive flexibility, inhibitory control (inhibition), and working memory (Diamond, 2013). EF processes play an important role in human functioning, as they are crucial for the planning and execution of activities for daily living (Diamond, 2013). Given the importance of CB, self-esteem, social support, and EF, as well as their association with the well-being of people with ID (Kim et al., 2016; Koch et al., 2015; Luerssen & Ayduk, 2017; Lunsky & Benson, 2001), it is important to look for interventions that can improve these factors.

Involvement in leisure activities is associated with a better well-being (Duvdevany & Arar, 2004) and may improve social skills and reduce emotional and behavioural problems among people with ID (Eratay, 2013). Unfortunately, people with ID participate less in leisure activities than people without ID (Merrells et al., 2018). Moreover, people with ID tend to perform leisure activities that occur in specialist settings like day centres (Reynolds, 2002) and are mostly solitary and passive in nature (Buttimer & Tierney, 2005). People with ID may experience various barriers to leisure participation, such as expenses (for the activity itself and the involved caregivers) and the facility being too short-staffed to adequately meet the activity interests of individuals (Reynolds, 2002). Interventions aimed at facilitating participation in leisure activities may contribute to the well-being of people with ID (Badia et al., 2013). An active leisure life can consist of a variety of enjoyable activities, such as watching a movie, playing bingo, and craft activities (Buttimer & Tierney, 2005; Reynolds, 2002; Umb-Carlsson & Sonnander, 2006). It can also include musical activities (Buttimer & Tierney, 2005; Reynolds, 2002).

Several reviews present a positive association between well-being and musical activities in the general population (Dingle et al., 2021; Macdonald, 2013; Sheppard & Broughton, 2020). Furthermore, it has been suggested that active music making (i.e., making music or sounds together) can be more promising than passive music methods (i.e., listening) for targeting well-being and EF (Haering, 2018; Van Der Steen et al., 2017; Weinberg & Joseph, 2017). In addition, music group

interventions seem to improve some components of EF more than other leisure activities group interventions (e.g., visual arts or Lego brick training) among children (Bugos & DeMarie, 2017; Frischen et al., 2021). Active music making can be used as an effective intervention for increasing self-esteem in community settings (Hallam et al., 2012) and in people with mental health problems (Kwan & Clift, 2018). Furthermore, studies including children, adolescents, and elderly people found that active music making improved CB (Geipel et al., 2018; Raglio et al., 2008; Sung et al., 2006). Remarkably, little attention has been paid to the possible positive effects of active music making on adults with ID. One study showed that performing music activities improved communication and facilitated skill development (MacDonald et al., 1999). Furthermore, a non-systematic literature review concluded that music making interventions can decrease levels of aggression and self-injurious behaviour and can improve well-being (Savarimuthu & Bunnell, 2002). To summarize, while various studies have found an association between making music and an improved well-being, self-esteem, social acceptance, CB and EF in non-disabled and other patient groups, there is limited evidence with respect to people with ID.

The aim of the present pilot study was therefore to investigate the possible superiority of active music making activities over non-musical leisure group activities regarding their effects on well-being, self-esteem, social acceptance, CB and EF in adults with ID. Feasibility of this music intervention will be described in detail elsewhere.

Method

Design

We conducted a pilot cluster randomized controlled trial (RCT) with 29 adults with ID. The efficacy of a weekly active music group intervention was compared to the efficacy of a leisure activities group intervention using pre, post and follow-up measurements.

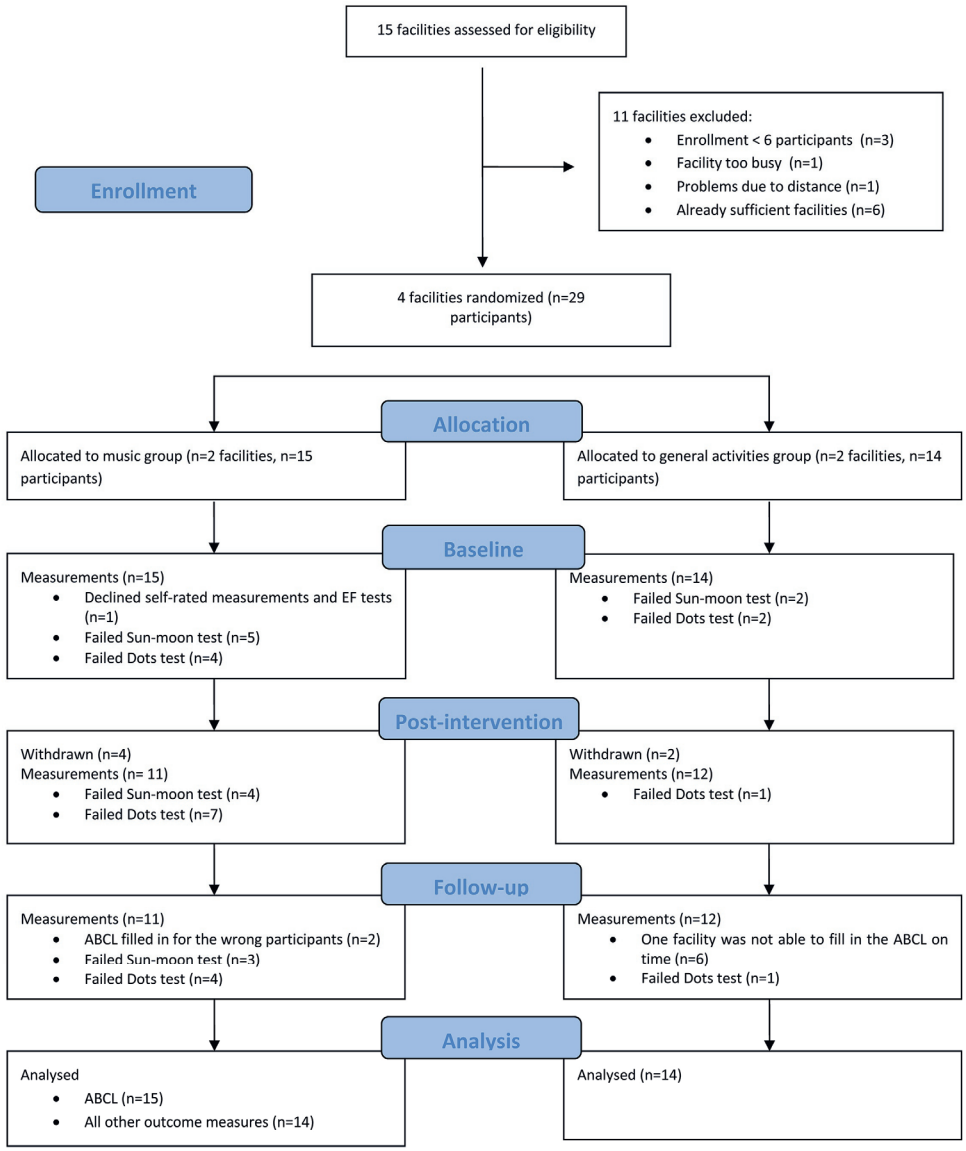
Participants

Participants (n=29) were recruited from four residential facilities of the Dutch service organisation Philadelphia Care Foundation (see Fig. 1). Fifteen facilities expressed an initial interest in participating in this study and were assessed for eligibility (i.e., majority of their residents (>80%) function at mild (IQ 50–69) or borderline (IQ 70–84) ID). Individual residents could participate if they had a mild or borderline ID, were 18 years or older, were able to participate in a group, and were mentally competent to give consent to participate. When people with a moderate ID from the

included facilities expressed interest in the study, they could also participate since it seemed unethical to exclude them from being involved in free enjoyable activities with their peers. This happened more often in the music group intervention than in the leisure activities group intervention. Unfortunately, some participants with moderate ID were not able to complete two of the selected assessment measures (see Fig. 1), and one participant refused all self-rated measurements and tests (see Fig. 1).

Figure 1

Flow diagram following participants through the study



Material and procedure

In this pilot cluster RCT, a comparison was made between two different types of groups: groups in which participants took part in an active music group intervention and groups in which participants were engaged in a leisure activities group intervention. The maximum group size was eight. After we recruited four facilities and all informed consents were collected, we found two music practitioners who would carry out the music intervention. Randomization of the facilities was pragmatic in nature: the facilities that were the nearest to the two music practitioners were allocated to the music group intervention. After the allocation, the special psychologists of the facilities were asked to fill in the characteristics of each participant: age, gender, level of ID and co-morbid psychiatric diagnoses. Data were collected at three time points: prior to the start of the intervention (baseline), the week after completing the intervention (post-intervention), and eleven weeks after the intervention ended (follow-up). Ethical approval for this study was granted by the Medical Ethical Committee of the VU University Medical Center. All participants and their legal representatives (when applicable, n=9) provided written informed consent.

Intervention

In both intervention conditions, 16 one-hour sessions were performed once a week. At each facility, two caregivers were assigned as coordinators for the intervention and during each session at least one of them was present. During the course of the RCT, the facilities could not start new musical or leisure activities (other than the intervention), however, musical or leisure activities that started pre-intervention were still allowed in both intervention conditions.

The music intervention was carried out by two trained music practitioners: one for each facility. A guideline was developed to ensure the intervention's validity and replicability. Prior to each session, the music practitioner and one of two caregivers who functioned as coordinators had a 10-minute meeting to prepare. Each session followed a routine beginning with a welcoming song, a listening exercise and a vocal warming-up. After this, the main part consisted of active music making, with participants choosing instruments to play (a variety of instruments was present) and ended with a cooling-down and always the same closing song. During each session, the trained music practitioner was not only assisted by a caregiver who functioned as coordinator, but also by a second caregiver. This way, most caregivers from each facility experienced the techniques used by the music practitioner and participated actively. The idea behind such a caregiver-assisted music intervention was that this would stimulate the integration of music into daily caring situations (Götell et al., 2000).

Furthermore, participants and their caregivers were encouraged to perform musical activities in-between the music sessions.

The leisure activities intervention was carried out by a research assistant. Per session, the participants could choose which non-musical activities they wanted to engage in (e.g., gaming, playing darts, playing cards, baking, puzzling). They could choose the same activity multiple times. These activities were chosen as several caregivers indicated in a survey that these activities were suitable and enjoyable. In addition, the activities all had an interactive nature in order to mirror the music intervention and to ensure a comparable level of attention and group contact. A manual and list with possible activities were provided to the research assistant.

Behavioural and cognitive tests

Challenging behaviour

The Adult Behaviour Checklist (ABCL) (Achenbach & Rescorla, 2003) was used to assess the level of emotional and behavioural problems of each participant as perceived by two caregivers who work at the facility and know the participant well. The caregivers from each facility could choose whether to distribute the task of filling in the ABCL for all participants from their facility among different caregivers, or to assign two caregivers who fill in the ABCL for all the participants from their facility. The researchers stressed the importance of inter-observer reliability (and lobbied for assigning two caregivers to the task), but only at one facility the ABCL was filled in by the same two caregivers at all three time points. The ABCL has been found to be a reliable (mean Cronbach's alpha = .84) and valid measure of emotional and behavioural problems in an ID inpatient sample (Tenneij & Koot, 2008). The ABCL consists of 118 items scored on a 3-point Likert scale. The items form eight subscales and a total score (maximum 236) can be calculated. A higher score on the (sub)scales indicates more CB. Three subscales concern internalized problems (anxious/depressed, withdrawal, and somatic complaints), three concern externalized problems (aggressive behaviour, rule-breaking behaviour, and intrusiveness), and two concern cognitive problems (thought and attention). In this study, the total score as well as the internalizing subscale (maximum score 64) and the externalizing subscale (maximum score 70) were used.

Well-being

As an indicator of well-being, we chose to determine the level of satisfaction and frustration regarding the need for competence, autonomy and relatedness through administrating the Basic Psychological Need Satisfaction and Frustration Scale – Intellectual Disability (BPNSFS-ID, Frielink et

al., 2019). This is a self-report questionnaire consisting of 24 items that are scored on a five-point Likert scale (Frielink et al., 2019). The BPNSFS-ID has been shown to be a valid and reliable measure for people with ID, with a Cronbach's alpha of .92 (Frielink et al., 2019). Two higher-order constructs need satisfaction and need frustration were assessed, with a maximum score per construct of 60. A low score on need satisfaction is associated with a dissatisfied person, whereas a high score on need frustration is associated with a frustrated person.

Self-esteem and social acceptance

Self-esteem and social acceptance were measured using the adapted Dutch version (Treffers et al., 2002) of Harter's (1988) Self-Perception Profile for Adolescents (SPPA). In this self-report questionnaire, the item format of the SPPA was modified with only one statement per question (Soenens et al., 2006; Vermeir & Van de Sompel, 2014). The two used subscales of the SPPA both consist of five items scored on a four-point Likert scale (maximum score 20), with higher scores representing more self-esteem or social acceptance. The validity and reliability of the Dutch version of the SPPA is acceptable to good with Cronbach's alpha > .70 (Treffers et al., 2002).

Executive functioning: inhibitory control

Inhibitory control was measured by the Sun-Moon Test (Dutch: Zon-Maan Test) from the Cross-Cultural Dementia (CCD) Screening (Goudsmit et al., 2017). Part A consists of sets of pictures of suns and moons which participants with ID have to name as fast as possible. In part B, participants with ID have to name 'sun' as moon and 'moon' as sun, using the standard Stroop paradigm. The time to completion and the number of errors is recorded. Each error results in penalty seconds (3.4 per error) which are added to the completion time. In this study, only the time score on part B was analysed, since this part measures inhibition (Goudsmit et al., 2017).

Executive functioning: cognitive flexibility

Cognitive flexibility was assessed by applying the Dots Test (Dutch: Stippentest) from the CCD Screening (Goudsmit et al., 2017). This test is based on the Trail Making Test (Reitan & Wolfson, 1985), but uses stimuli that resemble domino pieces instead of letters and numbers. In part A, dominoes ranging from 1 to 9 have to be connected by the participant with ID in the right order, as fast as possible. In part B, black and white dominoes have to be connected alternately while also taking into account the ascending numbers, again as fast as possible. To obtain a measure of cognitive flexibility, the time to completion in part A is subtracted from the time to completion in part B (Bugos et al., 2007; Thaut et al., 2009).

Executive functioning: visual working memory

Visual working memory was assessed using the Circle Span Backward Test from the Neuropsychological Test Series for Elderly People with Mild Intellectual Disability (NETOL) (Verberne, 1998). The Circle Span Test resembles the Corsi Block task (Lezak, 1983). Participants with ID have to mimic the research assistant backwards as they 'tap' a sequence of circles. The participant receives 1 point for each correct sequence of 'taps' and there are 8 sequences in total.

Data analysis

Raw scores were used in all analyses. Person mean imputation per subscale was applied for the ABCL and BPNSFS-ID in order to handle missing values. An exception was made for items 56a-56g of the ABCL: if these were left blank, they were scored 0, in accordance with the manual (Achenbach & Rescorla, 2003). Mann-Whitney, chi-square and Fisher's Exact tests were used to explore whether baseline characteristics differed between the two intervention groups (see Table 1).

To examine the effect of each intervention separately, an exploratory linear mixed model was performed to assess the within-group change from baseline to post-intervention and from baseline to follow-up. Since there were significant age differences between the intervention groups, age was added as a covariate in the following analyses. Three linear mixed model analyses were performed for the interaction between the treatment variable and time (overall), post-intervention (T2) and follow-up (T3) (Twisk et al., 2018). This made it possible to adjust for the dependency of the repeated observations within the subjects. If an interaction effect was significant, an extra analysis using Friedman's ANOVA or Wilcoxon signed-rank test was performed between the facilities of the intervention group in which the initial effect was found (to check whether the effect was significant in both facilities or caused by only one facility).

A p-value less than 0.05 was considered statistically significant. Given the exploratory nature of this study, no corrections for multiple tests were made (Streiner & Norman, 2011). All statistical analyses were performed in SPSS version 26.

Results

Three of four facilities completed the full 16 intervention sessions. Due to miscommunication between the caregivers and the research assistant who carried out the leisure activities group intervention, one leisure activities facility completed 14 sessions. Visual inspection of the

standardized residuals for each analysis demonstrated a normal pattern of distribution. Baseline characteristics are shown in Table 1.

Table 1

Participant characteristics per intervention group

	Music group	**Leisure activities group**	**Statistic (degrees of freedom)**	**P-value**
N	15	14		
Median age in years (range)	56 (42-78)	41 (19-73)	54.50	.03
Gender (men)	6	7	.29	.59
Level of ID			4.22	.16
IQ ≥70	3	1		
IQ 50-69	6	11		
IQ 35-49	6	2		
Psychiatric diagnosis			2.98	.64
ASD	3	2		
MD	2	1		
PD	1	0		

Notes: ASD = Autism Spectrum Disorder; MD = Mood Disorder; PD = Personality Disorder

Table 2 presents an overview of the median scores and range of each test per intervention group per measurement, and of the significant within-group differences at post-intervention and follow-up compared to baseline. Participants in the music groups felt significantly less frustrated with their needs and expressed more self-worth post-intervention compared to baseline. Participants in the leisure activities groups exhibited less CB and internalizing behaviour, and felt more social acceptance post-intervention compared to baseline. In addition, participants in the leisure activities group reported more satisfaction with their needs at follow-up and better inhibitory control at both post-intervention and follow-up compared to baseline.

Table 2

Median and range per outcome measure per intervention group at baseline (T1), post-intervention (T2) and follow-up (T3)

Outcomes	Music group			Leisure activities group		
	T1	T2	T3	T1	T2	T3
Challenging Behaviour (ABCL)						
Total (0-236)	48.00 (10-100.58)	39.34 (14.00-109.00)	40.00 (7.00-129.38)	35.46 (12.00-83.26)	29.61* (12.00-58.25)	39.50 (23.00-54.00)
Internalizing (0-64)	13.00 (1.00-36.00)	12.00 (3.00-38.00)	12.00 (2.00-33.00)	11.50 (5.00-28.00)	7.00* (1.00-17.00)	10.00 (5.00-13.00)
Externalizing (0-70)	12.00 (3.00-32.58)	12.00 (2.00-46.00)	10.00 (2.00-40.00)	11.14 (0.00-24.00)	8.50 (2.00-31.00)	11.00 (2.00-25.00)
Well-being (BPNSFS-ID)						
Need Satisfaction(12-60)	58.00 (42.00-60.00)	56.00 (35.00-60.00)	57.00 (31.00-60.00)	52.50 (36.00-59.00)	49.50 (40.00-59.00)	55.00* (48.00-58.00)
Need Frustration (12-60)	28.00 (15.00-60.00)	25.00 (18.00-44.00)	18.00* (15.00-43.00)	22.84 (12.00-52.00)	20.00 (12.00-46.00)	22.00 (15.00-40.00)
Self-esteem (SPPA)						
Global Self-Worth (5-20)	17.00 (8.00-20.00)	19.00* (7.00-20.00)	19.00 (5.00-20.00)	17.50 (12.00-20.00)	17.50 (9.00-20.00)	17.50 (11.00-20.00)
Social acceptance (SPPA)						
Social Acceptance (5-20)	15.50 (12.00-20.00)	17.00 (7.00-20.00)	15.00 (8.00-20.00)	13.50 (9.00-19.00)	16.00* (12.00-19.00)	15.00 (10.00-18.00)
Executive functioning						
Sun-Moon Test (seconds)	58.0 (41.0-117.6)	64.8 (40.0-143.8)	61.0 (42.0-129.8)	39.5 (19.0-137.6)	33.5* (19.0-69.6)	35.5* (19.0-80.2)
Dots Test (seconds)	87.0 (-79.0-134.0)	74.5 (52.0-163.0)	74.0 (38.0-124.0)	44.0 (3.0-258.0)	45.0 (9.0-199.0)	37.0 (11.0-235.0)
Circle Span Backward Test (0-8)	1.5 (0.0-3.0)	0.0 (0.0-4.0)	0.0 (0.0-4.0)	3.0 (1.0-8.0)	3.0 (0.0-8.0)	2.0 (1.0-8.0)

Notes: *p<.05, with exploratory linear mixed models analyses within-group compared to baseline;
ABCL = Adult Behaviour Checklist; BPNSFS-ID = Basic Psychological Need Satisfaction and Frustration Scale – Intellectual Disability; SPPA = Self-Perception Profile for Adults

Table 3 presents the results of the linear mixed model analyses for each outcome variable with the overall interaction effect (Group x Time effect on average over time), and the interaction effect (Group x Time) at post-intervention (T2) and at follow-up (T3) between intervention groups. The overall interaction effect was only significant on the subscale internalizing behaviour of the ABCL, showing that participants of the leisure activities group intervention exhibited a larger reduction in internalizing behaviour than participants of the music group intervention ($F(1, 44.81) = 5.27$, $p = .03$). Further analysis showed that the scores on the internalizing behaviour subscale were only significantly lower at post-intervention compared to baseline for one of the two facilities participating in the leisure activities group intervention ($\chi^2(1) = 5.00$, $p = .03$). The age of the participants in this facility (n = 6, Mdn = 27) was significantly lower compared to the other leisure activities facility (n = 8, Mdn = 58) (Mann-Whitney $U = 46.50$, $z = 2.91$, $p = .00$).

At post-intervention, a significant interaction effect was found on inhibitory control in favour of the leisure activities group intervention ($F(1, 39.64) = 5.67$, $p = .02$). Further analysis showed that there was only a significant effect at the same leisure activities facility as mentioned regarding the subscale internalizing behaviour ($T = .00$, $p = .04$). At follow-up, no significant interaction effect was found.

Table 3

Results of the linear mixed models analyses regarding the overall interaction effect and the interaction effect at post-intervention (T2) and follow-up (T3) between intervention groups

Outcomes	B (95% confidence interval)	F	p-value
ABCL Total			
Overall	-9.48 (-21.24-2.27)	2.64	.11
T2	-12.09 (-25.09-.91)	3.53	.07
T3	-2.39 (-18.25-13.47)	.09	.76
ABCL Internalizing Behaviour			
Overall	-4.43 (-8.31--.54)	5.27	**.03**
T2	-6.11 (-10.34--1.88)	8.51	**.01**
T3	-.96 (-6.13-4.20)	.14	.71
ABCL Externalizing Behaviour			
Overall	-2.49 (-7.69-2.72)	.92	.34
T2	-2.81 (-8.73-3.10)	.92	.34
T3	-1.10 (-8.36-6.17)	.09	.76
BPNSFS-ID Need Satisfaction			
Overall	1.04 (-2.87-4.96)	.29	.60
T2	-1.15 (-5.57-3.27)	.27	.60
T3	3.60 (-.82-8.02)	2.67	.11
BPNSFS-ID Need Frustration			
Overall	2.33 (-2.46-7.12)	.95	.33
T2	.54 (-4.94-6.03)	.04	.84
T3	4.16 (-1.32-9.65)	2.33	.13
SPPA Global Self-Worth			
Overall	.67 (-1.17-2.51)	.53	.47
T2	.98 (-1.27-3.24)	.76	.39
T3	.32 (-1.93-2.58)	.08	.78
SPPA Social Acceptance			
Overall	-.83 (-2.62-.96)	.85	.36
T2	-1.13 (-3.23-1.04)	1.01	.30
T3	-.51 (-2.67-1.65)	.23	.64
Sun-Moon Test			
Overall	-13.98 (-30.48-2.52)	2.91	.10
T2	-22.35 (-41.32--3.37)	5.67	**.02**
T3	-6.43 (-24.76-11.92)	.50	.48
Dots Test			
Overall	2.23 (-37.78-42.26)	.01	.91
T2	-10.88 (-67.14-45.37)	.15	.70
T3	6.96 (-41.10-55.03)	.09	.77
Circle Span Backward Test			
Overall	.50 (-.66-1.66)	.75	.39
T2	.61 (-.80-2.01)	.76	.39
T3	.44 (-.97-1.84)	.39	.54

Notes: ABCL = Adult Behaviour Checklist; BPNSFS-ID = Basic Psychological Need Satisfaction and Frustration Scale – Intellectual Disability; SPPA = Self-Perception Profile for Adults

Discussion

This pilot cluster RCT found significant within-group changes in several outcome measures after executing both types of interventions for adults with ID. However, no evidence was found that active music making in a group improves well-being, self-esteem, social acceptance, CB and EF more than other leisure activities within 16 weekly sessions. In contrast, people in the leisure activities group intervention showed significantly higher inhibitory control and significantly less internalizing behaviour at post-intervention compared to people attending a music group intervention. The improvement on inhibitory control within the leisure activities group intervention is in line with the finding that a leisure time activity programme for people with ID improved self-control (Eratay, 2013) and that a creative movement group for children resulted in more improvement in EF compared to a musical arts program (Park et al., 2015). Concerning the effect on internalizing behaviour, other studies also found less CB in people with ID after general leisure activities (Heyvaert et al., 2012). This promising effect of a leisure activities group intervention on inhibitory control and internalizing behaviour highlights the importance of structurally available caregivers to organize leisure activities and support people with ID participating in them.

The between-group differences on inhibitory control and internalizing behaviour were mainly caused by participants in one leisure activity facility. It therefore seems likely that differences in characteristics between (the participants of) these facilities played an important role. To begin with, the participants of the facility that caused the effect were significantly younger than the participants of the other facility. This seems relevant and is in line with Ferguson et al. (2021) who reported a decline in inhibitory control across the lifespan. Also, these participants were all diagnosed with mild ID, while the participants in the other facility were not (also moderate and borderline ID). Furthermore, it was reported by their caregivers that these participants spent a lot of time on their own before the start of our intervention. These findings suggest that a leisure activities group intervention is more effective on behaviour for younger participants with mild ID, possibly because people with mild ID in institutional settings tend to experience barriers to their social inclusion (Abbott & McConkey, 2006). Further studies which take the variables of age, level of ID and social inclusion into account, are needed to develop a better picture of the effectiveness of leisure activities group interventions.

There are several possible reasons for not finding more improvement after attending a 16 week active music making group on any of the outcomes. A first reason is that it is plausible that a

relatively short-term active music group intervention (<20 weeks) does not improve well-being, self-esteem, social acceptance, CB and EF more than a leisure activities group intervention. The findings of other studies seem to confirm this. For example, one month of musical interventions did not improve well-being and CB among people with dementia more than being engaged in other pleasant activities (Samson et al., 2015). Likewise, D'Souza and Wiseheart (2018) found no improvement in EF after three weeks of music training, compared to a dance training. The only short-term music intervention (8 weeks) in which an improvement in EF (i.e., inhibition) was found, made a comparison with care-as-usual and this positive effect disappeared when compared to art classes (Bowmer et al., 2018). Benefits in CB among children with ID have been seen after 24 weeks of music training, however, in this study participants in the other group received no comparable activities at all (Kalgotra & Warwal, 2017). When looking at musical interventions that were carried out for a longer period, it was found that after 32 weeks of active music making the self-esteem and well-being of people with a severe mental illness improved (Chang et al., 2018). In addition, EF among primary school children increased more after respectively 8 months (Frischen et al., 2021), 1.5 year (Roden et al., 2014) and 2.5 years (Jaschke et al., 2018) of musical involvement when compared to children involved in an arts group or natural science training. In sum, it seems possible that some of the benefits of musical involvement are only achieved after a longer period of time, with a minimum of 32 weeks.

A second reason is that the frequency of the music intervention (once a week) was too low, and that significant changes require a more intensive intervention. Previous studies have shown that 5 days a week of active music making results in benefits in EF among children without ID (Shen et al., 2019) and in CB among children with ID (Kalgotra & Warwal, 2017). However, the latter study made a comparison with care-as-usual only. When a comparison was made with a group that also received an intervention for 5 days a week, both groups showed an equal improvement in EF (Kosokabe et al., 2021). In our study, caregivers were expected to stimulate participants of the music group intervention to also play music in-between the sessions. Unfortunately, this active stimulation was not performed according to protocol and this may have resulted in a frequency of active music making that was too low to realise a detectable impact on the outcome measures.

A third reason is that the music group played music together as a 'one-intervention-suits-all' approach. This seems challenging with regards to the diversity among participants concerning musical preference, cognitive functions, communicative and social skills (Mulhall et al., 2021; Ptomey et al., 2017). It is for example possible that for some participants the musical activity was not

challenging enough, which is a necessary condition to increase EF (Diamond & Ling, 2016). Several authors express that an individual-based music intervention can be more effective (Ogg-groenendaal et al., 2014; Vereenooghe & Langdon, 2013). This is supported by a study among children with autism, whose self-esteem and peer attitudes increased while anxiety decreased following a music intervention with a staff to participant ratio close to 1:1, which ensured enough individual attention (Hillier et al., 2012). An individual-based intervention also prevents the ethical dilemma of withholding an enjoyable group activity to people living at the same facility who do not meet the inclusion criteria, since an individual intervention can be held at the comfort of their own apartment and ensures a more homogeneous inclusion of participants. Within the leisure activities group intervention, it was possible to split different activities in smaller groups, which might explain why this intervention was more effective on outcome variables. In fact, this was indeed done by the two facilities of the leisure activities group intervention: sometimes the group split up (and played for example board games and table tennis in smaller groups) and sometimes they did an activity with the whole group (e.g., making a large drawing together). This finding provides support for the importance of tailoring interventions to the needs and preferences of participants.

Besides the discussed between-group outcomes on EF and CB, significant within-group changes in several outcome measures were found after both types of interventions. First, self-esteem of participants in the music group intervention improved. This is in line with Park et al. (2015), who found that self-esteem improved after 30 hours of arts education composed of musical arts, while it did not improve after a similar amount of creative movement. This effect seems promising and needs further investigation. Second, the need frustration of participants in the music group and the need satisfaction of participants in the leisure activities group improved. These results need to be treated cautiously since the used questionnaire (BPNSFS-ID, Frielink et al., 2019) proved to be quite complex for many participants, especially for people with moderate ID. There are concerns about the usage of self-report questionnaires in general (of which the BPNSFS-ID is just one example) among people with moderate ID (Kooijmans et al., 2022). In addition, it should also be mentioned that the need satisfaction of the participants in the music intervention group was quite high at baseline, demonstrating a well-being homeostasis (McGillivray et al., 2009). Third, the social acceptance of the participants in the leisure activities group intervention improved. This resonates with indications that engagement with others as part of community and structured activities promotes social well-being (Eisenman et al., 2017).

The current study has several limitations. First, the uneven distribution of ID level between the two intervention groups was a consequence of the cluster randomization and the small number of clusters involved. Second, the small sample size limits the generalizability of the results, which is quite common in RCTs that include people with ID (Mulhall et al., 2018). The sample size was further reduced by the fact that the used EF measures proved to be too difficult for some participants, especially for those with moderate ID. This especially reduced the sample size in the music group, and resulted in limited power to detect changes on EF outcome measures. Problems with limited power occur often in ID research studying EF (Spaniol & Danielsson, 2021), especially when the limits of the participants' EF skills are pushed on demanding EF tasks (Diamond & Ling, 2016), which might have occurred with our participants with moderate ID. This 'floor effect' is therefore common when studying EF in people with ID (Willner et al., 2010). Third, the data completion rate was rather low at follow-up for the ABCL, because some caregivers did not deliver this questionnaire on time. Fourth, since the inclusion criteria were broadened for ethical reasons, the group of participants was more heterogeneous with respect to level of ID than anticipated. This made it challenging for the researchers to identify instruments that assess the relevant outcomes and are appropriate for all participants. Heterogeneity and its practical implications and barriers is a common issue in RCTs in the field of ID care (Mulhall et al., 2021). This underlines the importance of both homogeneous inclusion and using questionnaires and tests that are appropriate for all included participants.

Overall, there is a lack of consensus regarding the appropriate length and intensity of the intervention that is needed to reliably improve well-being, self-esteem, social acceptance, CB and EF in adults with ID. Further research is therefore necessary. Future studies should also explore whether an individualized music intervention suits the needs and interests of such a heterogeneous group of people with ID better. Furthermore, to enable the measurement of the planned outcomes in people with mild and especially moderate ID, it is recommended to make more use of custom-developed tests that are adapted to their skills (Willner et al., 2010). For now, this paper shows the promising pilot results of a leisure activities group intervention for younger people with mild ID and contributes to a better understanding of the issues that should be addressed when carrying out an effective music intervention for people with ID.

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CHAPTER 3

3

Chapter 3

A music intervention and a leisure activities intervention for adults with intellectual disabilities: a feasibility study

Swart, E. A. K., Smeets, G. J. J., Volkers, K. M., Scherder, E. J. A., & Moonen, X. M. H. (2024). *A music intervention and a leisure activities intervention for adults with intellectual disabilities: a feasibility study* [Manuscript submitted for publication].

Abstract

***Background*:** This study assessed the feasibility of a pilot cluster-RCT of two group-based interventions: a music group intervention and a leisure activities group intervention in adults with intellectual disabilities.

***Method*:** Feasibility was evaluated using qualitative and quantitative data from participants (people with intellectual disabilities) and providers (music workers, caregivers, research assistant) via questionnaires and log reports. Qualitative data were analysed using a six-domain feasibility framework; quantitative data were analysed with Mann-Whitney U tests.

***Results*:** The music group intervention was deemed suitable, while the leisure activities group was unfeasible due to practical issues resulting in a lack of appeal. The cluster-RCT was unfeasible due to methodological and practical issues.

***Conclusions*:** Major adaptations are needed for conducting a large-scale RCT including: (1) an individual intervention tailored to different cognitive abilities; (2) care-as-usual as a control condition; and (3) improve communication and clarify roles by establishing a project group per facility.

Introduction

Adults with intellectual disabilities often exhibit challenging behaviour (CB), including aggression, self-injury, and destructive behaviour (Emerson et al., 2001). Research suggests several interventions for addressing CB in adults with intellectual disabilities (Heyvaert, Maes, & Onghena, 2010; Bruinsma, Van den Hoofdakker, Groenman, et al., 2020), one of them might be a music intervention. Engaging in musical activities has demonstrated numerous positive effects across various populations, such as enhanced well-being and improved CB in the general population (Macdonald, 2013), children and adolescents (Geipel et al., 2018), as well as in adults with intellectual disabilities (Savarimuthu & Bunnell, 2002). Also, leisure activities have demonstrated beneficial effects on well-being and the reduction of CB among adults with intellectual disabilities (Duvdevany & Arar, 2004; Eratay, 2013). Despite these promising effects on well-being and CB, there has been scant attention directed towards the intellectual disabilities population, especially in Randomized Controlled Trial (RCT) settings.

RCTs play a crucial role in promoting and implementing evidence-based interventions (Mulhall et al., 2018). As noted repeatedly however, RCTs are scarce in intellectual disabilities research literature (Jones & Chaplin, 2020; Koslowski et al., 2016; Vereenooghe & Langdon, 2013). In addition, published RCTs in this domain often encounter methodological and practical challenges, including issues related to participant recruitment (e.g., obtaining a representative sample), study design (e.g., navigating participants' consent), engagement of caregivers and stakeholders, as well as attitudes and perceptions towards research participation from healthcare staff and others involved (Mulhall et al., 2018; Mulhall et al., 2021).

Alongside the methodological and practical challenges associated with RCTs, ethical dilemmas concerning the potential harm to individuals with intellectual disabilities raise considerable concerns about their participation in research in general (McDonald & Kidney, 2012; McDonald et al., 2017). Given the complexity and concerns about RCTs involving individuals with intellectual disabilities, it is crucial to understand the potential challenges that may arise during the planning and execution of such studies. By identifying these challenges, researchers can proactively address them by adjusting study designs, protocols, and procedures as needed. Conducting pilot studies and feasibility testing has proven to be a valuable strategy in preparing for the implementation of large-scale RCTs (Blatch-Jones et al., 2018). In these endeavours, it is essential to consider the perspectives and experiences

of various stakeholders, including those who will affect and be affected by the intervention (Bowen et al., 2009).

The current article focuses on a pilot cluster-RCT aimed at evaluating the impact on CB of adults with intellectual disabilities of two group-based interventions. Both interventions were group-based and caregiver-assisted, since the social aspect of a group was expected to bring additional benefits over an individual approach (Hooper, 2002; Duffy & Fuller, 2000), and caregivers are crucial for successful participation in interventions and research designs (Scott et al., 2019).

Before testing the effect on CB of both interventions in a larger-scale RCT, it is important to evaluate their feasibility in a pilot study. The purpose of this pilot study was to assess the feasibility of a cluster-RCT comparing a music group intervention with a leisure activities group intervention among adults with intellectual disabilities. This paper describes the data analysed, focusing on the six key areas of feasibility according to Bowen et al. (2009): acceptability, demand, implementation, practicality, adaptation, and limited efficacy of the pilot (See Table 2).

Method

Study design

A feasibility study was conducted to evaluate a music group intervention and a leisure activities intervention for adults with borderline (IQ 70-85) to mild (IQ 50-69) intellectual disabilities. This feasibility study was embedded in a pilot cluster-RCT assessing the effects of both interventions on well-being, self-esteem, social acceptance, CB, and three components of executive functions (inhibitory control, cognitive flexibility, and visual working memory). The outcomes of this pilot study have been described elsewhere (Smeets et al., 2023). In the present feasibility study, the experiences and perspectives are assessed of both the people who received (one of) the interventions (adults with intellectual disabilities who will be referred to as ‘receivers’ throughout this article) and the people who provided (one of) the interventions (professional caregivers, music workers, and a research assistant who will be referred to as ‘providers’ throughout this article).

Procedure

Figure 1 presents a flow diagram of the pilot cluster-RCT procedure and the feasibility study procedures. Fifteen residential facilities of the Dutch care provider Philadelphia Care Foundation initially expressed interest in participating in the pilot study. These facilities were assessed for

eligibility (criterion: at least 80% of the residents have mild (IQ 50-69) or borderline intellectual disabilities (IQ 70-85). Four residential facilities were found eligible, and these were randomly assigned to one of the two intervention groups: two facilities participated in a group-based music intervention by a caregiver, and two facilities participated in a group-based leisure activities intervention. Residents of the participating facilities were assessed for eligibility and had to meet the following criteria: having mild or borderline intellectual disabilities, being at least 18 years old, being able to engage in group activities, and being mentally competent to consent to participate in the study. Residents with moderate intellectual disabilities (IQ 35-50) were also accepted to participate when they expressed interest in participating, since the researchers recognized the negative ethical implications of excluding them from potentially enjoyable activities other residents in the same facility would receive.

Interventions

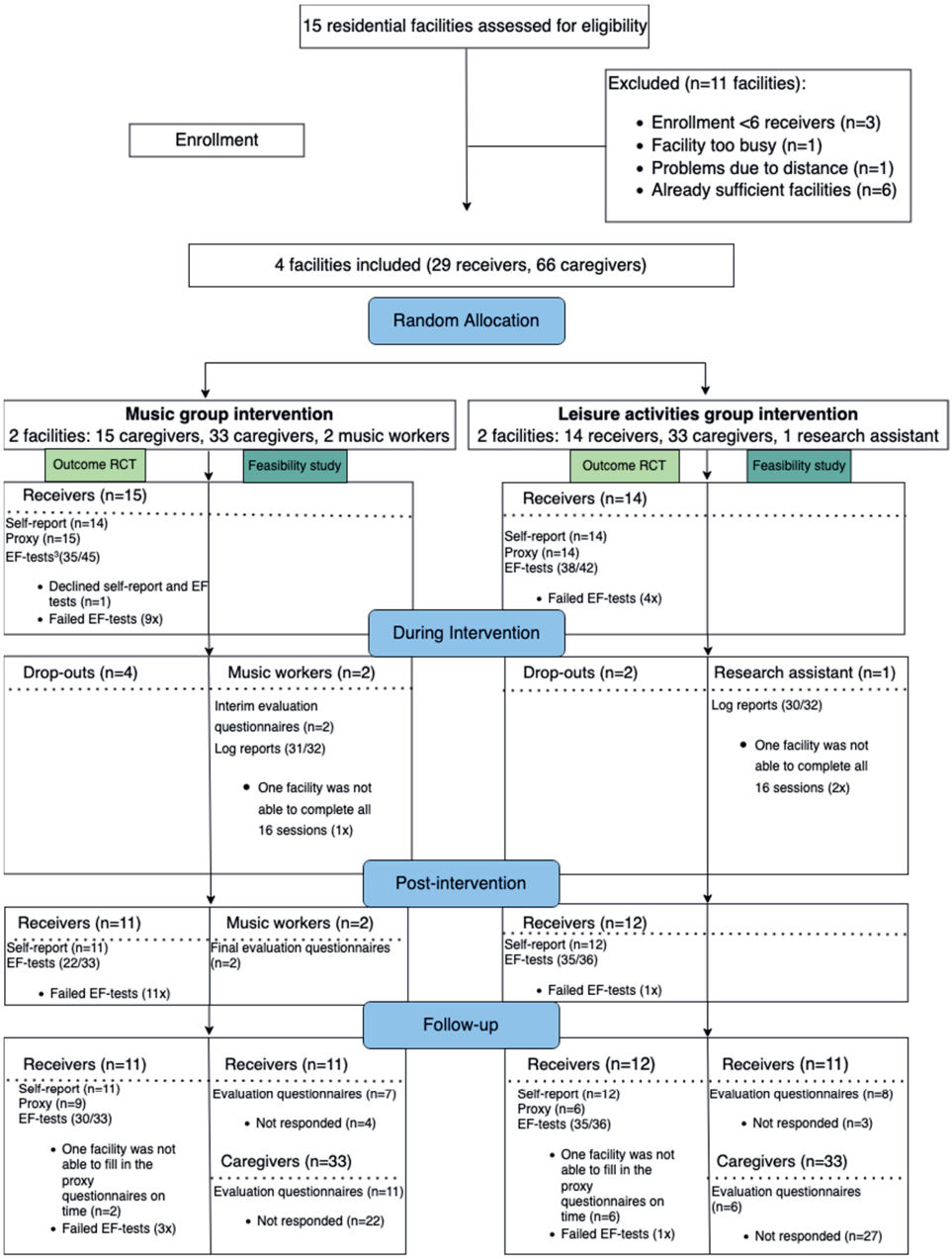
Both group intervention consisted of 16 one-hour sessions, conducted once a week.

Music group intervention

This intervention was provided by professional music workers, assisted by at least one, but preferably two, caregivers (i.e., care staff from the residential facilities). The group engaged in making music together, such as playing instruments and singing. A guideline for these sessions was developed with the help of several professional music workers, caregivers, and adults with intellectual disabilities. The sessions were conducted according to this guideline to ensure the trial's validity and replicability. Prior to each music session, the music worker and caregivers had a meeting in which they prepared the session and discussed any details. After the session they had another meeting to discuss how the session was conducted and to provide feedback on each other's actions. At the end of each session, the receivers were given a homework assignment (e.g., making up different lyrics for a song, singing along with music they liked) and were encouraged to perform musical activities with the support of their caregivers. In both residential facilities that received the music group intervention, one caregiver served as a contact person and attended each session to be able to transfer musical knowledge and skills to the group of receivers. Other caregivers from the facility were asked to attend at least one session.

Figure 1

Flow diagram of the pilot cluster-RCT intervention groups



1 n = number of participants, x = (un)succeeded
2 Self-report = filled in by receivers; Proxy = filled in by caregivers, EF = Executive Functioning
3 Each receiver performed 3 EF tests during each measurement, and each test is considered separately
4 Log reports are per session per facility

Leisure activities group intervention

This intervention was provided by a research assistant, assisted by at least one caregiver (i.e., care staff from the residential facilities). Receivers could choose any type of activity, except activities that involved music (e.g., gaming, playing darts, playing cards, baking, puzzling). The research assistant was given a manual and a list with possible activities, which were selected based on survey results indicating they were both suitable and enjoyable. All activities were designed to be interactive. Receivers were allowed to choose between participating in activities either as a group or the group could be divided into subgroups based on which activity they liked. They were also allowed to choose the same activity multiple times.

Measures and procedure

Feasibility data were collected from both the intervention receivers and the providers. Data were collected in two ways. First, log report forms were developed and used to assess whether the sessions were conducted as intended. The music workers (music group intervention) and the research assistant (leisure activities group intervention) used this form after each session to document the performed activity and any special events that happened. The form included questions based on the sequence of activities and the preconditions of the research design (e.g., the attendance of receivers, key observations, and the content of that session).

Second, several evaluation questionnaires were developed and administered. In the music group intervention, these questionnaires were completed at three time points: (1) halfway through the intervention (8 weeks), (2) post-intervention (16 weeks), and (3) follow-up (11 weeks after post-intervention), see also Figure 1. At time point 1, music workers completed a questionnaire about the intervention after a meeting with the entire team of caregivers from that facility. This questionnaire was designed to gather feedback or insights for upcoming sessions. At time point 2, music workers completed a questionnaire after a final evaluation meeting, also with the entire team. This questionnaire contained questions about their experiences with the music sessions. At time point 3, both caregivers and receivers completed questionnaires featuring a ten-point scale for caregivers and a five-point scale for receivers. These surveys solicited feedback on preferences and concerns regarding the sessions. Additionally, receivers were asked to report any perceived effects on their emotional state, sense of belonging to the group, and, specifically for caregivers, to report any perceived effects in receivers.

In the leisure activities group intervention, evaluation questionnaires were completed at time point 3 only, because leisure activities were more common and well-known by caregivers and receivers, and were not executed by paid, external workers, contrary to the musical activities. The questionnaires were filled in by the leisure activities group intervention receivers and caregivers and contained similar questions to those used at time point 3 for the music group intervention.

In addition, we evaluated the feasibility of the outcome measures of the pilot cluster-RCT. These outcome measures were divided into proxy questionnaires (i.e., filled in by caregivers), self-reports, and executive functioning (EF) tests and were collected at baseline, post-intervention, and follow-up (Smeets et al., 2023).

Participants

The feasibility study explored the experiences of both receivers and providers of both interventions. Characteristics of the receivers are shown in Table 1. The providers (n=69) were 2 music workers, 1 research assistant, and in total 66 caregivers (music group intervention n=33, leisure activities group intervention n=33).

Table 1

Participant characteristics per intervention group

	Music group	**Leisure activities group**	**Statistic (degrees of freedom)**	**P-value**
N	15	14		
Median age in years (range)	56 (42-78)	41 (19-73)	54.50	.03
Gender (men)	6	7	.29	.59
Level of ID			4.22	.16
IQ ≥70	3	1		
IQ 50-69	6	11		
IQ 35-49	6	2		
Psychiatric diagnosis			2.98	.64
ASD	3	2		
MD	2	1		
PD	1	0		

Notes: ASD = Autism Spectrum Disorder; MD = Mood Disorder; PD = Personality Disorder

Data analysis

To analyse and report the data, we used six key areas of feasibility: acceptability, demand, implementation, practicality, adaptation, and limited efficacy (Bowen et al., 2009).

The data were analysed deductively using a stepwise content analysis process (Bingham & Witkowsky, 2022). First, the first and second author (ES and GS) coded the data independently, using the six key areas of Bowen et al. (2009) as described above. Second, they compared and discussed their codes in order to reach consensus. The third author (KV) was consulted in case of doubt or disagreement. Third, after reaching consensus, the original codes were reviewed and potentially relabelled based on further discussion or comparison. Fourth, the codes were grouped into main findings for each key area.

The quantitative data (addressing the key area of Acceptability) were analysed using IBM SPSS Statistics version 24. Mann-Whitney U tests were conducted to evaluate differences between the music group intervention and the leisure activities group intervention. A p-value of $< .05$ was considered significant.

Table 2

Key areas of feasibility according to Bowen et al. (2009)

Key areas	Description	Indicators
Acceptability	How the target group of the implemented program reacts to the intervention	Attractiveness of the intervention for providers and receivers Satisfaction Perceived appropriateness
Demand	The estimated use or the actual use of the intervention	Expressed interest or intention to use Intent to continue use
Implementation	The extent in which an intervention can be fully implemented as planned and proposed	Degree of execution Success or failure of execution
Practicality	The extent in which an intervention can be delivered when resources, time, commitment, or some combination thereof are constrained in some way	Factors affecting implementation ease or difficulty Efficiency, speed, or quality of implementation Ability of receivers and providers to carry out intervention activities Difficulty of outcome measures
Adaptation	How the program contents or procedures can be changed to be appropriate in a new situation	Suggestions for improvement
Limited efficacy	The successfulness or promising effects of the program or process with the intended population	Intended effects of program or process on key intermediate variables

Ethical considerations

Ethical approval for the pilot cluster-RCT (including the feasibility study) was granted by the Medical Ethical Committee of the VU University Medical Center. The statement from the Medical Ethical Committee of the VU University Medical Center indicated that the research was not subject to the Dutch Medical Research Involving Human Subjects Act. All intervention receivers provided written informed consent.

Results

The results are described per key area of interest (see Table 2). For each area, a distinction is made between the experiences with regard to the music group intervention, the leisure activities group intervention and the RCT-design. Where applicable, results are described from the perspective of both intervention receivers and intervention providers.

Acceptability

Music group intervention

Overall, music intervention receivers responded positively to its acceptability. They found the music sessions enjoyable and engaging rating it by a mean score of 4.4 out of 5. They also felt they had benefitted from the clear and simple language used by the music workers during explanations. Their overall recommendation for others to participate in a group to engage in music activities was high, with a mean score of 4.9 out of 5. Their opinion of homework assignments was generally neutral, with a mean score of 3.4 out of 5.

Caregivers' experience was also generally positive, with an average score of 7.4 out of 10. However, personal experiences varied considerably. While some caregivers rated the intervention as positive, others felt the intervention was inappropriate and too complex for receivers, especially when tasks involved discussing emotions through music. Additionally, incorporating musical activities between sessions was deemed impractical, with a mean score of 4.0 out of 10. Three out of 11 caregivers expressed dissatisfaction with the homework assignments as part of the intervention.

Music workers reported that receivers seemed to find the music intervention satisfying and attractive, noting visible enthusiasm during the sessions.

Leisure activities group intervention

Receivers reported a mean score of 3.3 out of 5, which was significantly lower than among receivers of the music intervention (U = 26, z = -2.57, p = .01). They mentioned that their expectations regarding the variety and type of activities were not met, indicating a preference for self-designed activities over those arranged by the research assistant. Their overall recommendation to others to participate in the leisure activities sessions, with a mean score of 3.3 out of 5, was also significantly lower than that of receivers in the music intervention (U = 13, z = -3.22, p = .00).

Caregivers rated the leisure activities group sessions with a mean score of 5.3 out of 10, but this score did not differ significantly from the ratings given by caregivers about the music group intervention (U =20, z =-1.32, p =.19). Caregivers reported that these sessions added little value to the usual activities performed during standard daily care.

RCT-design

Caregivers in the music intervention group gave an average score of 5.8 out of 10 when asked how likely they were to recommend participation in the RCT to others. Caregivers from the leisure activities group rated this question with a mean score of 2.5 out of 10. The difference between these recommendation scores did not reach statistical significance (U = 18, z = -1.75, p = .08).

Demand

Music group intervention

Overall, receivers expressed a desire to continue with the sessions. Some highlighted their enjoyment, with one receiver expressing disappointment at the prospect of the end of the sessions.
Providers also conveyed a positive interest in extending the intervention, recognizing their significant value for receivers. Additionally, caregivers proactively came up with ideas on how to sustain musical activities for individual receivers.

Leisure activities group intervention

Receivers reported a lack of added value of the group-based sessions and expressed disinterest in extending the intervention. Caregivers concurred, emphasizing that the sessions consumed too much time and effort, which could be better utilized for daily care.

RCT-design

Caregivers from both interventions found the organisation of the interventions and the research design demanding. For example, they emphasized the burden of completing multiple questionnaires and the challenges associated with organizing sessions and fostering motivation in the receivers during the sessions.

Implementation

Music group intervention

The vast majority of sessions were successfully completed (97% completion rate). Most sessions adhered to the manual, following the established sequence of activities (87% compliance rate). However, four variations from the protocol were reported. First, in only 67% of the sessions both caregivers were present. Second, the brief preparation meeting prior to each session between the music worker and caregivers, proved to be barely feasible: in only 31% of the cases both caregivers participated, in 58% of the cases one caregiver attended this meeting and in 11% of the cases, no preparation meeting was held. Third, only 29% of the feedback meetings, occurring immediately after each session, were attended by both caregivers. In the remaining 71% of the meetings, only the caregiver who served as contact person attended. Fourth, music workers reported that caregivers did not always complete the scheduled homework. More specifically, only 14% of the homework assignments were completed by all receivers, 32% by most receivers, 5% by half of the receivers, 55% by a few receivers and 18% by none of the receivers.

Leisure activities group intervention

At one facility, only 14 sessions (88%) were completed due to miscommunication between the caregivers and the research assistant. At the other facility, all 16 sessions (100%) were completed. In 80% of the sessions the manual was followed, adhering to the fixed sequence of activities. In the remaining 20%, there were valid reasons for deviating from the fixed sequence of activities, such as certain tasks requiring more attention. In both facilities, the presence of at least one caregiver at each session was 100%, and in 7% of the sessions, an extra second caregiver also attended, which was no strict requirement.

RCT-design

In the music group intervention, all receivers successfully completed the baseline measurement, but the participation rate dropped to 73% for both post-intervention and follow-up measurements. Among caregivers involved, the two caregivers who were part of the project achieved a 100%

completion rate for the baseline measurement and an 82% completion rate for the follow-up measurement (proxy questionnaire). 33% of all caregivers participating in the music group intervention completed the evaluation questionnaires. 100% of receivers in the leisure activities intervention group completed the baseline measurement, 75% completed the post-intervention measurement, and 69% completed all three measurements (baseline, post-intervention, and follow-up). Among caregivers, the two caregivers who were involved in the project achieved a 100% completion rate for the baseline measurement. 50% completed the follow-up measurement (proxy questionnaire). 18% of all caregivers from the leisure activities group intervention completed the evaluation questionnaires.

Practicality

Music group intervention

In general, receivers faced various challenges to be fully engaged in the program as intended. First, it became evident that most receivers struggled to complete homework assignments independently: they required assistance from caregivers. Caregivers did not always spend sufficient time to aid them individually. Second, some receivers found it challenging to keep up with the pace of the group, with two receivers struggling to follow the rhythm of the music, while others sought a more substantial challenge than what the group sessions had to offer. The dropout rate among receivers reached 27%. Reasons for dropout included intense emotional responses evoked by specific types of music (e.g., grief and sadness) and feelings of overburden.

While caregivers generally viewed the intervention positively, they highlighted several factors that made its implementation difficult. First, they found it demanding to integrate the intervention into their regular responsibilities. Second, caregivers doubted the receivers' ability to fully comprehend the music worker's instructions. Third, they highlighted difficulties regarding the considerable amount of preparation time required prior to each session.

On the music worker's side, there was a perception that receivers were capable of carrying out the music activities during sessions. However, they also noted occasional strong emotional displays, emphasizing the importance of caregivers being present to help receivers regulate these emotions. An additional benefit of the presence of caregivers, as mentioned by the music workers, was that they knew the receivers well and could respond adequately to their needs. Finally, a music worker highlighted that some receivers had difficulty participating in the sessions due to additional

disabilities (e.g., dementia or a hearing impairment) along with issues related to variability in the level of cognitive functioning amongst receivers.

Leisure activities group intervention

In general, receivers found the activities too challenging and not tailored to their needs or preferences. They expressed dissatisfaction, noting that the activities were either too demanding or too similar to their usual experiences. The dropout rate among receivers was 14%. Both receivers and caregivers attributed this dropout to a lack of enthusiasm.

Similarly, caregivers reported challenges in assisting with the sessions, particularly in keeping receivers motivated, as well as in the substantial amount of preparation time needed before each session. Their feedback indicated that the activities were unsuitable and lacked diversity.

RCT-design

Caregivers from both intervention groups considered timely completion of proxy-questionnaires unfeasible. Moreover, some caregivers expressed a general aversion to complete questionnaires. Overall, caregivers perceived their participation in the RCT as more demanding than initially expected, citing concerns about the high volume of questionnaires and forms they had to complete. Additional challenges included communication issues with researchers and difficulties leading up to the intervention, which they found unclear and chaotic. When asked to what extent they knew what was expected from them, caregivers scored on average 5.2 out of 10.

Regarding the outcome measures, some difficulties were encountered, especially with the EF tests. This resulted in receivers terminating these tests prematurely or obtaining either a minimum score or no score at all. The incidence of experienced challenges with EF tests increased with the severity of the level of intellectual disabilities (see Table 3).

Table 3

Incidence of challenges with EF tests per measurement, amongst receivers from both intervention groups with different levels of intellectual disabilities

Levels of intellectual disabilities (number of receivers per measurement; baseline, post-intervention, follow-up)	Percentage of failed EF tests per measurement		
	Baseline	Post-intervention	Follow-up
Borderline intellectual disabilities (4,2,2)	8.3	16.7	16.7
Mild intellectual disabilities (17,15,15)	11.7	20.0	8.9
Moderate intellectual disabilities (8,6,6)	41.7	55.6	38.9

Note: Each receiver performed 3 EF tests during each measurement, and each test is considered separately. Consequently, a maximum of three failures per receiver was possible at each measurement.

Adaptation

Music group intervention

In general, feedback from receivers indicated that the intervention did not require substantial modifications. However, two suggestions were made: shorter session durations and no strict attendance of caregivers.

On the caregivers' side, there were several recommendations for modifying the intervention. Some preferred that music workers conduct the intervention independently without being involved themselves. Additionally, caregivers were sometimes confused about aspects regarding project implementation and expectations, particularly about providing support with homework and with attending sessions. Complaints were also raised about inadequate communication between parties involved, which led to a request for clearer instructions.

Leisure activities group intervention

Some receivers suggested more diverse activities and a more collective approach (i.e., discouraging splitting up of the group). On the other hand, some receivers expressed dissatisfaction with activities organized by the research assistant, leading them to engage in individual activities instead.

Caregivers of the leisure activities group confirmed these suggestions and stressed the importance of providers understanding the needs and frustrations of receivers.

RCT-design

Receivers of both intervention groups conveyed a preference for a less demanding RCT-design, particularly concerning the complexity and duration of the questionnaires and tests.

Likewise, some caregivers from both intervention groups also expressed negativity towards the research design and recommended reducing the number of forms and questionnaires. Additionally, they emphasized the need for improved communication about the research process, including clear expectations and a timeframe.

Limited efficacy

Music group intervention

Overall, receivers reported positive experiences both during and after the music sessions, with the majority expressing increased feelings of happiness. Caregivers also reported positive effects on the receivers, including increased happiness and a stronger sense of connection in the group during or after the sessions. However, nearly 40% of the caregivers perceived no noticeable effects on receivers. See Figure 2 for an overview of the perceived effects. Additionally, music workers, although not directly questioned about perceived effects, perceived improved interaction among receivers and a stronger sense of belonging as the intervention progressed. This observation was consistently documented in the log reports, with a specific focus on the increase in making eye contact.

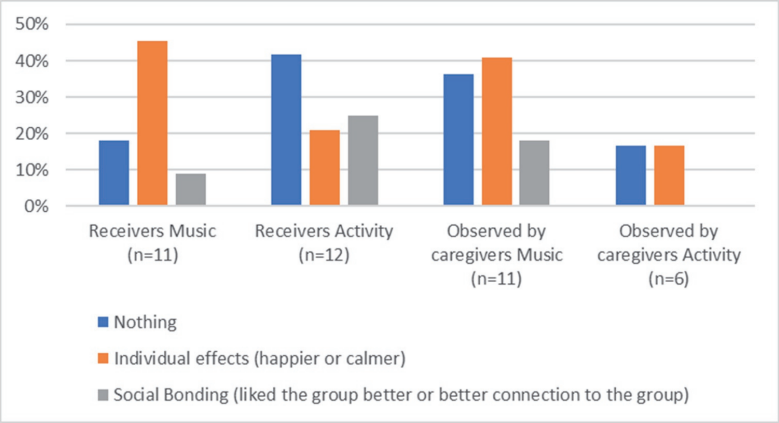
Additionally, we aimed to equip caregivers with musical knowledge and skills that they could transfer to receivers. To assess this, caregivers were asked if they felt they had learned a lot during the music sessions. They rated their experience at 4.2 out of 10.

Leisure activities group intervention

Overall, receivers reported negative experiences either during or after the sessions, citing factors such as lack of enthusiasm and boredom with the activities. The majority reported no noticeable effects. The negative experiences during the sessions were supported by the rating of the caregivers, who reported being negatively affected by the workload and the difficulty of keeping receivers motivated. Nevertheless, certain positive effects were also noted, for example feeling happier and enjoying the group more. See Figure 2 for an overview of the experienced effects.

Figure 2

Perceived effects of participation in the sessions



Discussion

The aim of this pilot evaluation study was to explore the feasibility of conducting a cluster-RCT regarding the effects of a music group intervention compared to a leisure activities group intervention for adults with intellectual disabilities. This study showed that the music intervention met two key feasibility areas of Bowen et al. (2009): acceptability and demand. Receivers responded favourably to the music intervention, which was well-received and effectively engaged adults with intellectual disabilities. Conversely, the leisure activities group intervention failed to meet any of the six key feasibility areas; lacking demand and being deemed non-implementable, unacceptable, impractical, and non-adaptable within intellectual disabilities-care. This difference was supported by positive feedback from both receivers and providers, who reported greater happiness, calmness, and social bonding in the music group comparted to the leisure activities group.

The overall feasibility of the RCT design was found to be low, primarily due to high demand regarding the design. Thus, the findings suggest that the current design of the cluster-RCT based on a group intervention in an existing group home is not feasible from the perspectives of both receivers and providers. We will first discuss a few barriers encountered in this pilot study related to the key areas of Bowen et al. (2009). Second, we provide several recommendations for future cluster-RCT studies. Finally, we discuss the strengths and limitations of this pilot study.

Barriers found in the feasibility study

The first barrier, related to the acceptability and practicality of the study, concerned communication, role clarity, and the level of engagement, commitment, and attitude of caregivers. Insufficient communication between researchers and intervention providers led to uncertainties about the expectations and timelines of the intervention process. Communication was mainly conducted through a contact person via phone and email, with minimal face-to-face interaction. The lack of role clarity resulted in caregivers being less actively engaged in the implementation of the interventions and the execution of the RCT-design. The commitment exhibited by intervention providers played a major role in the successful implementation of both the interventions and the research design. Not everyone showed interest in participating in the research, indicating variability in the attitudes and perceptions of staff members, which is an important factor for successful research implementation (Mulhall et al., 2021).

The second barrier, related to the practicality, and the implementation of both interventions and the RCT-design, involved the high demands of the tasks (i.e., homework assignments, completing forms, questionnaires, and tests) and the excessive workload as perceived by both receivers and providers. This issue aligns with the barriers identified by Lennox et al. (2005), who observed that commitments perceived as an additional workload, such as participating in research, tend to be a low priority for caregivers who already struggle with the demands of daily care. Furthermore, receivers with moderate intellectual disabilities faced more challenges on the EF tests (e.g., terminating the test prematurely or scoring minimal or no points) compared to receivers with mild or borderline intellectual disabilities. Choosing questionnaires and tests that are only appropriate for individuals with a specific level of intellectual disability could potentially exclude individuals with varying ability levels of intellectual disabilities. However, strict exclusion based on IQ criteria is considered unethical, as it may foster feelings of exclusion among participants (Mulhall et al., 2021).

The third barrier, also related to the practicality of the study, arose from the heterogeneity of adults with intellectual disabilities, which made participation in an RCT more complex (Mulhall et al., 2021). Challenges arose in adapting the interventions and RCT-design to accommodate diverse cognitive abilities and in sustaining motivation and engagement. Tailoring activities to the level and preferences of the receivers is crucial, as failure to do so results in a less valued intervention. Moreover, including individuals with additional disabilities, such as age-related issues (e.g., hearing impairment or early-stage signs of dementia), presented practical difficulties, as they struggled to keep pace with group activities or fully participate in assessments.

Recommendations

Several important considerations for future interventions emerged from the positive and negative findings of this feasibility study.

Interventions

- When investigating a music intervention or leisure activities intervention in people with intellectual disabilities, it may be more feasible to compare each intervention with care-as-usual, considering the complexities involved in comparing two different new interventions (Higgins & Greens, 2011).
- Shifting from a group to an individual intervention format allows participants to receive more individual attention and support. This allows intervention providers to better adapt the Intervention to the unique needs, preferences, and abilities of each participant (Beck et al., 2010). Alternatively, if a group format is still desired, efforts can also be made to create a more balanced group, based on factors such as level of intellectual disability, additional diagnoses, interests, and support needs.
- The homework was too demanding and should be designed in a way that it does not put extra burden on receivers and caregivers. For example, homework should consist of tasks that each participating individual with intellectual disabilities can perform without the assistance of caregivers. Nevertheless, it remains uncertain to what extent individuals with intellectual disabilities can independently complete homework assignments without assistance of caregivers.
- It is recommended to start the intervention with caregiver assistance due to its advantages (Scott et al., 2019). However, given the high demand, it should not be mandatory for all sessions, but a point of discussion between the intervention provider and caregiver how many sessions assistance is necessary, based on the expertise of the music worker and the group's performance.
- Caregivers reported not having gained sufficient knowledge and skills from the music sessions by only attending the sessions. Other methods, such as implementing a train-the-trainer program are recommended when transferring knowledge and skills is a goal (Poitras, et al., 2021).

RCT design and measures

- Create a project group per facility to enhance teamwork, clarify roles, and improve communication, as this can facilitate the implementation processes (McGuier, et al., 2023).

Continuously evaluate the effectiveness of communication and make necessary adjustments, utilizing email, face-to-face interactions, or other suitable channels.

- Tailor the RCT to the abilities of the receivers. For example, when adults with moderate intellectual disabilities are included, the questionnaires and tests should be adapted to fit their cognitive abilities (Willner et al., 2010).

Strengths and limitations of this pilot study

The feasibility study is characterized by several strengths and limitations. A first strength is that the perspectives of various stakeholders (i.e., adults with intellectual disabilities, caregivers, music workers, research assistant) were explored to assess the feasibility of the cluster-RCT. A second strength is that both interventions were not only aiming at improving CB, well-being, and EF of adults with intellectual disabilities, but were designed to expand the support repertoire of the caregiver by working together with a music worker or research assistant. This dual focus is crucial for the intervention's adoption and integration into practice (Hithersay et al., 2014; Turk et al., 2012).

The limitations of this feasibility study are mostly inherent to the character of a pilot study. First, this study was conducted in 4 facilities with 29 participants. Therefore, the results may not be fully applicable to other settings with different contexts, populations, or characteristics. Second, the data gathered from log reports and evaluation questionnaires hinders comprehension of specific underlying mechanisms or explanations. Utilizing additional sources like semi-structured interviews could have provided deeper insights into the results of this study (Harris et al., 2019).

In conclusion, this feasibility study provided insights into several challenges that arose during the execution of a pilot cluster-RCT on the effects of a music group intervention compared to a leisure activities group intervention for adults with intellectual disabilities. It outlines various methodological and practical challenges for optimizing interventions and research designs. Overall, these findings underscore the importance of carefully considering feasibility and making necessary adjustments to ensure the success of future interventions, particularly in the context of large-scale cluster-RCTs.

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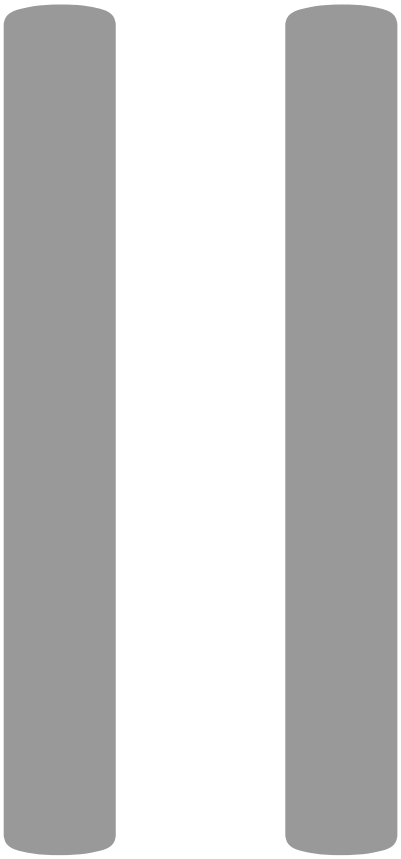
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PART II



PART II

COVID study

CHAPTER 4

4

Chapter 4

Feasibility, experiences and short-term outcomes on challenging behaviour and well-being of a partially online music intervention pilot study for adults with intellectual disabilities

Smeets, G. J. J., Volkers, K. M., Swart, E. A. K., Moonen, X. M. H., & Scherder, E. J. A. (2024). Feasibility, Experiences and Short-Term Outcomes on Challenging Behaviour and Well-Being of a Partially Online Music Intervention Pilot Study for Adults With Intellectual Disabilities. *Journal of Applied Research in Intellectual Disabilities*, 38(1), e13314. https://doi.org/10.1111/jar.13314

Abstract

***Background*:** During the COVID-19 pandemic we implemented a partially online music intervention to examine the feasibility, experiences and short-term outcomes on the challenging behaviour and well-being of adults with intellectual disabilities.

***Method*:** This mixed-methods study included 10 participants with mild or moderate intellectual disabilities who received 16 one-hour individual music sessions in 10 weeks, either face-to-face or online. Data on feasibility and experiences from participants and music workers were collected and analysed using inductive thematic analysis. Challenging behaviour and well-being were measured before and after intervention.

***Results*:** Overall experiences were positive and concerned appreciation, positive feelings, musical abilities, attention span, relatedness and personalisation. Online experiences varied, but most participants preferred face-to-face over online sessions. After the intervention, challenging behaviour scores were better than before.

***Conclusions*:** Engaging in a partially online music intervention is feasible for people with intellectual disabilities and seems to improve challenging behaviour. Experiences are discussed and recommendations for future online sessions are provided.

Introduction

The COVID-19 pandemic had a profoundly negative impact upon the general population (Lai et al., 2020), and especially amongst vulnerable populations such as people with intellectual disabilities (Doody & Keenan, 2021). People with intellectual disabilities were more likely to face social adversity and health inequalities than the general population when being treated in healthcare systems during the pandemic (Hassiotis et al., 2020), which magnified the disparities in healthcare (Baksh et al., 2021). Despite their high-risk status, there was a deficiency in public health guidelines specific to people with intellectual disabilities (Desroches et al., 2021). People with intellectual disabilities faced significant challenges during the COVID-19 pandemic. The restrictions on visits and reduced face-to-face interactions with professionals (Rauf et al., 2021) led to isolation and loneliness (Embregts et al., 2020; Lake et al., 2021). Not only the quantity of formal support networks diminished, but also its quality (Scheffers, Moonen, & Van Vugt, 2021). Furthermore, due to disruptions in their home environment, daily schedules and occupational or recreational activities, it was difficult maintaining structure in their daily lives (Amor et al., 2021; Embregts et al., 2020; Navas et al., 2021). In addition, they perceived difficulty in understanding the COVID-19 preventive measures and adhering to public health measures, which was at least partly caused by an inadequacy in the provision of accessible COVID-19 information online and offline (Chadwick et al., 2022) and increased stress (Desroches et al., 2021; Embregts et al., 2020; Lake et al., 2021). This disruption of services, increased stress, and uncertainty about the future contributed to an increase in psychiatric consultations (Rauf et al., 2021), increased challenging behaviour (Schuengel et al., 2020), and decreased well-being (Amor et al., 2021).

Interventions, either biological, psychotherapeutic or contextual, to decrease challenging behaviour and improve well-being have received considerable attention within the field of intellectual disabilities research (Bruinsma et al., 2020; Heyvaert et al., 2010). One example of a non-pharmacological intervention is a music intervention, which has been shown to have a positive impact upon both challenging behaviour and well-being amongst people with intellectual disabilities (Andring, 2015; Mino-Roy et al., 2021; Savarimuthu & Bunnell, 2002). In addition, active music-based interventions (i.e., playing musical instruments) seem to provide more benefits, for example on challenging behaviour in people with dementia (Gómez-Gallego et al., 2021) and on well-being in the general population (Weinberg & Joseph, 2017), than receptive techniques (i.e., listening to music). Within this scope, a meta-analysis concluded that an individual music intervention, with personalised attention and targeted support, is more effective than a group intervention (Pelletier, 2004). In line

with this, an individualised music intervention, based on personal musical preferences, is most effective in reducing challenging behaviour (Gerdner, 2000) and stress (Sakamoto et al., 2013) for people with dementia. However, during the COVID-19 pandemic face-to-face music interventions required adjustments due to the imposed restrictions upon engaging with professionals in person (Rauf et al., 2021). Therefore, alternatives were required, such as innovative online interventions (Lunsky et al., 2022; Rauf et al., 2021). Online interventions have been successfully applied to people with intellectual disabilities who exhibit challenging behaviour (Oudshoorn et al., 2021; Sheehan & Hassiotis, 2017), and have been found to be potentially beneficial within the social and behavioural domains (Moreno et al., 2021) and as positively impacting upon their well-being (Chadwick et al., 2022). We argue that an online music intervention could also be beneficial for both the challenging behaviour and well-being of people with intellectual disabilities.

Online interventions with people with intellectual disabilities face several challenges, including securing free access to digital devices (Lussier-Desrochers et al., 2017), the digital literacy of this group and finding caregivers who are able to facilitate these online interventions (Chadwick et al., 2022). People with intellectual disabilities must also have the necessary cognitive and sensorimotor abilities in order to take part (Lussier-Desrochers et al., 2017; Sheehan & Hassiotis, 2017). Indeed, video-conference therapy was not suitable for people with intellectual disabilities who experience problems with communication, social interaction or processing difficulties, and therefore, only phone therapy was deemed to be suitable (Rawlings et al., 2021). For online music interventions, there are additional specific challenges pertaining to, for example, latency and variable sound quality (Dowson et al., 2021). In light of these challenges, it is essential to collaborate with people to ascertain which approach suits them best (Keenan & Doody, 2023) as one-size-fits-all solutions will not suffice. Consequently, it is plausible that blended interventions, that is, a combination of online and face-to-face intervention sessions, might be more suitable and acceptable than exclusively online sessions (Sheehan & Hassiotis, 2017; Topooco et al., 2017; Wentzel et al., 2016).

Given the preference for a blended intervention, and the positive effect of music on challenging behaviour and well-being, we conducted a pilot study to evaluate a partially online music intervention. Specifically, this study assessed the feasibility, experiences, and short-term outcomes on challenging behaviour and well-being of this partially online music intervention for people with intellectual disabilities. A concurrent mixed-methods approach was used to evaluate the experiences of participants, caregivers and music workers and quantify aspects of its potential impact.

Method

Study design

This pilot study started during the COVID-19 pandemic when physical visits were permitted only under certain conditions (Vereniging Gehandicaptenzorg Nederland, 2020). The intervention consisted of 16 one-hour individual music sessions that took place over a 10-week period. Data collection from participants as well as from professionals was underpinned by a concurrent triangulation design, with both quantitative and qualitative data being collected at baseline and post-intervention and log reports after each music session and subsequently side-by-side compared (Creswell, 2009).

Ethical approval

Ethical approval was provided by the Science and Ethics committee of the Faculty of Behaviour and Movement Sciences, VU Amsterdam (VCWE-2020-133). An independent medical ethics committee stated the research was not subject to the Dutch Medical Research Involving Human Subjects Act.

Participants

The participants comprised 10 people (50% male) with a median age of 62.5 years (ranging from 53 to 72 years of age) who were living within three residential facilities for people with intellectual disabilities in the Netherlands. Eight participants had mild intellectual disabilities (IQ 50–69), whilst the remaining two had moderate intellectual disabilities (IQ 35–49). Four of the participants had one or more psychiatric diagnoses: two people were diagnosed with autism spectrum disorder, three were diagnosed with a mood disorder, and one participant was diagnosed with an anxiety disorder.

The inclusion criteria were as follows: (1) 18 years or older: (2) must have mild or moderate intellectual disabilities (IQ 36–70); (3) live with intensive assistance, care, and behavioural regulation as described in the Dutch Long-term Care Act (Wet Langdurige Zorg, VG6) and (4) be mentally capable of giving informed consent. The exclusion criteria were as follows: (1) not capable of participating in a one-hour long intervention session, (2) have hearing problems that are not diminished via the use of a hearing aid or (3) have a medical condition (e.g., dementia) that would greatly impact upon their participation. Demographic information about the participants was provided by the special psychologist connected to the facility in which the participants lived.

Both the caregivers and special psychologists from the facilities determined which residents could potentially participate. Subsequently, potential participants and their legal representative (if applicable) were presented with an information letter and informed consent form providing detailed information about the study. Initially, there were 16 potential participants identified, however six of them ended up not participating for various reasons: three failed to hand in their informed consent form before the deadline, two stated they did not want to participate, whilst one participant withdrew on medical grounds. After the participants provided informed consent (n=10) and, if applicable, their legal representatives also did so (n=5), music workers who lived near the facility were contacted and the first informal meetings between music workers, caregivers and participants were organised.

Procedure

Initially, several care managers (n=14) from the Dutch service provider Philadelphia Care Foundation were contacted about participating in the study. There were various reasons for their non-participation: the facility had no people who met the inclusion criteria (n=3), COVID-19 related problems (n=3), contact ceased after multiple attempts at communication (n=3), or the facility had other priorities/organisational issues (n=2), which resulted in three participating facilities.

The music intervention

The sessions were guided by a music worker, who could be any individual who played an instrument and was capable of transferring their musical enthusiasm to someone with intellectual disabilities. More specifically, there were three music educators (‘muziekagoog’ in Dutch), one music therapy student, one psychology student with more than 15 years of experience as a musician and one professional caregiver with qualifications for music education. Most music workers had prior experience working with adults with intellectual disabilities, while others had experience working with people who have other cognitive disabilities (i.e., people with dementia). The sessions took place in a quiet setting, mostly the participants' apartment or another room of the facility where there would be no disturbances. During the sessions, the participants could choose from a box with a variety of instruments they had on loan during the entire intervention (e.g., hand drum, bongo, triangle, keyboard, and guitar). Each participant chose their own preferred music genre and songs, sometimes with some help from the music worker.

Each music session started with a welcoming song (2 min), followed by a listening exercise (5 min), a warming up (5 min), active music making (35 min), cooling down (5 min) and a closing song (2 min).

The music worker often personally adjusted the welcoming song to the participant and this song remained the same in each session. In the listening exercise, a music piece was introduced in which the participants were instructed to focus and answer questions based on that music piece. This music piece was different in each session and was played by the music worker. An alternative was for them to put on some music. Warming up exercises for both body and voice were included, tailored to match the subsequent components of actively making music: mostly instrumental performance afterwards, then the warming-up focused on movement, whereas when singing was the main component afterwards, then the warming-up included vocal exercises. In the active music making part, participants played on their preferred instruments and/or sang with the music worker. This was followed by a cooling down, in which the amount of beats per second was reduced. The music session ended with a closing song chosen by the participant. Each session was closed by the same song. The participants received a diploma after completing the final session.

The music workers and participants decided prior to each session whether the session would take place face-to-face or online, albeit under the condition that some of the sessions (no exact number was mandated) needed to take place online, in order to be able to compare the experiences of face-to-face to online music sessions. In those instances when the session was held online, a secure virtual environment was organised and the assistance of a caregiver from the facility was required. During online sessions the participant had access to all musical instruments.

The above-mentioned structure and guidelines were provided in a manual with several example exercises for the music worker. This manual was prepared by the authors of the study in order to ensure consistency between the different sessions and participants (available upon request from the corresponding author). No guidelines were provided on the amount of decibel levels nor on who should be in control of the volume.

Outcome measures

Three non-standardised measures were used to evaluate both the feasibility of and experiences with the music intervention. Two standardised measures were used to measure challenging behaviour and well-being, both at baseline and post-intervention. Given the restrictive measures in operation during COVID-19, all questionnaires and reports were sent and received digitally.

Non-standardised measures

Log Report by the Music Workers

After each session, every music worker completed a log report containing a series of questions about each session: whether the session was face-to-face or online, whether the session was carried out in accordance with the manual, which instruments were used, how much time was used for active music making, as well as any additional remarks they wished to make.

Personal Evaluation Questionnaires

Both the participants and music workers were asked to complete an evaluation questionnaire after the final music session. This questionnaire sought to collect data on the perceived benefits, barriers and intended musical activities after the intervention period as well as their respective experiences with the face-to-face and online music sessions. The music workers also answered questions about any potential behavioural or emotional changes that they had observed in the participants over the intervention period.

Final Meeting Questionnaire

After the final music session, the music worker, caregiver, and participant had a final meeting in which they evaluated the music sessions together with the help of a questionnaire. This helped them to discuss how the overall experiences with the music intervention were. Questions focused on what the participant, music worker and caregiver liked or disliked about the music intervention, perceived differences between the face-to-face and online sessions, possible observed changes in the participants' behaviour or emotions during the intervention and ideas for future improvement. The music worker noted down their joint answers.

Standardised measures

The Aberrant Behaviour Checklist (ABC)

The ABC (Aman et al., 1985) assesses the challenging behavior of people with intellectual disabilities against 58 items rated on a 4-point Likert scale, with 0 being ‘not at all a problem’ and 3 being ‘the problem is severe in degree’. It comprises five subscales: (1) Irritability, Agitation, Crying (15 items); (2) Lethargy/Social Withdrawal (16 items); (3) Stereotypic Behaviour (7 items); (4) Hyperactivity/Noncompliance (16 items) and (5) Inappropriate Speech (4 items). Higher scores indicate more challenging behaviour, with the maximum score being 174. The ABC has been found to be an adequately reliable and valid tool (Paclawskyj et al., 1997). Within the present study, the ABC was rated by caregivers of the participants. Although we stressed the importance of inter-observer

reliability, unfortunately in 30% of the cases the post-intervention ABC was completed by a different caregiver than the one who completed the baseline ABC.

The Personal Wellbeing Index-Intellectual Disability (PWI-ID)

The PWI-ID measures satisfaction within seven life dimensions (Cummins & Lau, 2005). Normally, there is a pre-testing protocol to determine both if and to what level of complexity scale a participant is able to understand the PWI-ID. However, we skipped this pre-testing protocol in order to reduce the test administering burden upon the participants. Therefore, we used the reduced PWI-ID version with three choices for the participants which use drawings of three faces (sad, neutral, and happy). As indicated in the manual (Cummins & Lau, 2005), we rescaled the items to scores from 0 to 100, to facilitate interpretation of the results, in which 100 is the most optimal overall well-being score. It has been found to be a reliable and valid tool for measuring the subjective well-being of people with intellectual disabilities (McGillivray et al., 2009).

Data analysis

Qualitative data were analysed with inductive thematic analysis (Braun & Clarke, 2006). For the first three participants, the text sections from all of the non-standardised measures that were relevant to answering the research questions, were identified and coded independently by two researchers. This was performed in steps to allow the two researchers (GS and KV or ES) to discuss the codes. In the event of doubt, the third researcher was consulted. Consensus about the codebook was reached after coding the information of the third participant, after which the first researcher (GS) coded the information of the last seven participants. All codes were grouped into themes (GS) and interpretations were jointly discussed and refined by the three authors (GS, KV and ES) in order to ensure that the codes within the themes were coherent and that the codes between the themes were distinctive.

The quantitative data from the log reports, personal evaluation questionnaires, and final meeting questionnaires were analysed via descriptive statistics. For the two standardised measures (i.e., ABC and PWI-ID) the descriptive statistics were reported. Due to the nature of this feasibility study, it was decided not to conduct any efficacy statistical tests on this data. In the event that there were two answers noted on the ABC or PWI-ID, the intermediate answer was calculated. This occurred five times for the ABC and two times for the PWI-ID. Two participants had one missing value on the PWI-ID, in which case a total score was calculated by adding up the domain scores and dividing them by 6

(rather than 7). Three participants had one missing value on the ABC, which was subsequently replaced with the mean of the non-missing values on the subscale.

To ensure internal validity of the data, the researchers used data triangulation by combining multiple data sources (i.e., informants) and methods (i.e., qualitative and quantitative) (Carter et al., 2014; Creswell, 2009).

Results

The results are described in the order of feasibility, experiences (first overall and subsequently focused on online music sessions) and short-term outcomes. Verbatim quotes are used as typical examples and are labelled using their unique identifier.

Feasibility

Recruitment started in July 2020 with a consent rate (participants/approached) of 62.5% and data collection ended in June 2021. Two participants did not take part in any online sessions and one participant only took part in online sessions. If we leave these three participants out, then the average number of online sessions was 4.4 (range 1–13). The reasons that were given for not conducting (more) online sessions were the illness of an assisting caregiver, the workload of the caregiver who needed to be available to assist, problems with one participant's sight (cataract), the continuously changing work schedule of one of the participants, and technical issues. The reason for not conducting any face-to-face music sessions with one participant was the choice of the music worker who had not yet received a COVID-19 vaccination at the time of the music sessions. Eight participants received all the sessions within the 10-week period, one participant needed a few days more, whilst one participant required a 14-weeklong intervention due to them often cancelling sessions at late notice.

Experiences

After analysing the data, six themes emerged regarding experiences of the combined face-to-face and online music intervention: appreciation, positive feelings, enhanced musical ability, attention span, sense of relatedness, and finally, personalisation.

Appreciation

All of the participants (n=10) stated they would recommend the music sessions to others and everyone reported that they would like to continue making music. Eighty percent of the participants indicated that the music sessions meant a lot to them.

Positive feelings

For all the participants, pleasure was mentioned the most by both participants and music workers. Both the participants and music workers reported that they really enjoyed making music, that they had a lot of fun during the sessions, were very enthusiastic about the music sessions and really looked forward to them. According to the music workers, the participants opened up, got goosebumps, got carried away by the music and were able to put their thoughts or feelings into the music, because some participants cried or nearly cried during sessions. Most music workers indicated that the music soothed and relaxed the participants and that the participants became surer of themselves during the music sessions. Some participants reported feeling a sense of pride, especially after receiving their diploma. In addition, caregivers mentioned that the participants showed a more pleasant and active participation in the group directly after having the music sessions. The caregivers indicated that they saw that the participants were happy and joyful, talking constantly about the music sessions and singing songs.

> *The enjoyment increased with every session. [Name of participant] can really enjoy music and doing it himself without restrictions and inhibitions really gave him wings. You could really see him shine. (music worker 5)*

> *[Name of supervisor] noticed that when she brought [name of participant] his medicines in the morning, he was already looking forward to the music session scheduled later that day. (music worker 5)*

Enhanced musical ability

Various music workers stated that the participants' musical abilities improved over the duration of the intervention, namely in terms of their rhythm, harmony, musical scales when playing on the piano and progress with singing.

> *Then I explored the things he really liked in greater depth, and this greatly improved his sense of rhythm and harmony. He now plays and sings almost everything in the right time and key. (music worker 5)*

Attention span

Several music workers reported on the attention span and focus of the participants: (1) one participant had a very short attention span (around 10 s), (2) one participant actually fell asleep during the music session, (3) one hour proved to be too long for one participant and she needed a pause in between, (4) one participant was easily distracted and (5) the other participants were also visibly tired at the end of the music session, but were able to stay focused for the entire hour.

Sense of relatedness

An overwhelming majority of music workers felt that the contact between the music worker and participant was very important. Both music workers and participants commented on the collaboration, positive vibe, trust, and warm bond between them. They both felt a sense of connection over the course of the sessions. According to music workers, an important indicator of this connection was by making more and more eye contact with one another over the course of the sessions.

> *She said she thought the contact was worthwhile and might want to do more with music. She said she saw me as someone she could confide in (which I was really pleased to hear) and always liked having a chance to say what was on her mind. (music worker 4)*

> *Initially, I had to get used to the music worker, but after some time, I had created a trusting relationship and looked forward to it every time. (participant 7)*

Personalisation

The importance of personalising the sessions to the individual wishes and needs of the participant, such as adjusting the tempo, was a major theme according to music workers. The music workers emphasised the need to both dig deeper into the things the participants mentioned and allow space for their emotions or daily struggles to come to the fore.

> *He sometimes talked about his emotions, but then said we should move on to another song, and so I did. (music worker 2)*

Additional focus on experiences with online sessions

Only one participant (10%) viewed the online sessions as pleasant, whereas 40% found them unpleasant or very unpleasant and 50% were either neutral or non-applicable. If there were only online sessions, then only 33% of the participants said they would participate again, whilst 44% said they did not know and 22% stated they would not participate. According to music workers, the online music sessions were unpleasant or very unpleasant for 78% of the participants. For themselves, 44% found the online music sessions to be unpleasant, 33% scored them as neutral or neutral/not pleasant, and 22% reported finding them pleasant (those who held the most online sessions).

The qualitative data analysis revealed four themes which will be discussed in turn below: negative online experiences, positive online experiences, comparison between the online and face-to-face sessions, and finally, conditions for being able to take part in the online music sessions and how to improve them.

Negative online experiences

The most important negative online experiences were the bad connection, impersonal contact, problems with synchronicity and sound, and the need to adjust musical methods, which will each be described below.

Music workers

The most reported negative online experience was the bad connection. In some instances this had to do with the specific program that was being used to enable a secure video call, which led to either insecurity about whether it would be possible to get in touch with the participant, or not being able to begin the session at the specified time. This, in turn, could sometimes lead to mutiny from the participants. The music workers expressed that the staff also often could not help with these issues. At other points, the connection was suddenly lost and participants had to either join again independently or go get help from a caregiver to do this if they were unable to do it themselves. One music worker argued that this was not helpful for the self-esteem of the participants. Another recurring point in the dataset pertained to the contact between the music workers and participants. Most of the music workers reported finding the online music session impersonal, namely because it was harder to understand the other person, harder to clarify what the participant had to do and harder to adjust to the participant. Because a lot of attention needed to be paid to the technical aspects of online sessions, it was difficult to also observe the participant. They reported being able to see only a limited part of what was going on and, hence, that they missed the bigger picture. This, in turn, made it harder for them to react to attitudes and expressions. It was also reported that there

was a lack of a sense of being together in the online sessions, which made it harder to build a bond or trust. The majority of music workers also experienced issues with synchronicity. There was a delay in the sound, making it difficult to play or sing together. Regarding the sound, in some instances the sound was too soft, whilst at other points there was reverb. Overall, there were many technical concerns, which underscores the fact that the online sessions required specific insight and technological know-how, both from the music workers and the participants.

> *As a music worker, you are also less able to respond to the situation if you have online sessions. One time, [name participant] had to go to the toilet for example and it took her ages to get back to the computer. At a moment like that, you have no way of seeing what's happening and I couldn't go and get her. Fortunately, she came back in the end and carried on singing enthusiastically (music worker 1).*

> *Then we sang some more songs, along with the YouTube clips. This was quite tricky, as the client has trouble finding the songs with lyrics on YouTube by herself. When we have a face-to-face session, I normally do that for her. As it was a lot of hassle, we ended up singing 'la-la-la' to another song (music worker 1).*

It was reported that musical methods routinely had to be adjusted, that in some instances it proved not to be possible to compensate for the problems the participants experienced. Making music together proved to be hard, as did explaining the exercises to the participants or showing them the right keys on a keyboard, for example. Ultimately, it was found that repeating known exercises (from the face-to-face sessions) proved to be the most effective method for some of the participants. Some music workers stated that it was exhausting and annoying to stare at a screen for long periods of time, both for themselves and participants, which in some instances led to sessions ending prematurely.

Participants

The participants also found the online sessions to be impersonal, or as one participant referred to it: 'not real'. Some of the participants stated being more easily distracted and that their concentration was worse in these sessions. The online music sessions were a 'hassle', as one participant put it.

> *I can't concentrate. Very crazy, with her being on the other side. (participant 3)*

Positive online experiences

Music workers

Several music workers stated that participants were cooperative during the online sessions. Two music workers explained that the online sessions were an easy and fast way to reach a person, or that, as one of them stated, the fact that the online music sessions could take place in the evening or during the weekend, meant that it better fitted their own attention span. The latter of these music workers also reported experiencing a greater sense of equivalency with the participant, because they were also able to see her own living room. Given the restrictive measures in place at that juncture, one music worker commented that the online music sessions helped their participant to not feel so isolated.

> *Even so, I recently noticed that [name participant] really does his best. He takes the instruments out of the box when I ask him to during the online session, and sometimes plays along wholeheartedly with the music we play together. (music worker 2)*

Participants

Only one participant was positive about the online sessions. The online sessions were a welcoming distraction and gave this person energy.

Comparison between online and face-to-face sessions

Music workers

The music workers reported that it did not feel like they were making music together with the participants when the sessions were online. They also felt that they had to work harder during the online sessions, and that participants exhibited greater agitation, showed less initiative
and engaged in less eye contact. Conversely, the face-to-face sessions were experienced as being more personal, and as making it easier to explain and engage in exercises together. However, one music worker also reported that it was just as easy (or hard) to make contact with the participant during the online sessions as it was face-to-face.

Participants

The participants answered the questions: 'What did you think of the face-to-face music sessions?' and 'What did you think of the online music sessions?' on a 5-point Likert scale. Enjoyment levels were higher for the face-to-face music sessions than they were for the online music sessions. The participants experienced the online sessions as being more impersonal and more rigid.

Conditions for taking part in the online music sessions and how to improve them

Music workers

Several conditions were mentioned by music workers with regard to being able to participate in the online music sessions which were related to the participants, technique or procedure. Regarding participants, it was mentioned that when a participant has bad sight, online sessions are practically impossible. Furthermore, online music sessions were more appropriate for participants who were capable of handling the potential stress of a bad connection. Regarding technique, several music workers noted that a system for secure video calls that works easily and that can be operated by everyone (i.e., music workers, caregivers, and participants) is necessary. They added that providing clear instructions to everyone involved would help to make this work in a more straightforward way. Another improvement that was suggested by the music workers was to use a system that would allow participants to turn on a musical fragment themselves. The final technical point mentioned was the necessity of a computer with a stable internet connection as well as a fully charged mobile phone in some instances. With regards to procedures, music workers recommended developing a bond and sense of trust between music workers and participants prior to starting the online sessions. In addition, they suggested that more time should be reserved for engaging in regular conversation in the programme, and that the intervention should start with face-to-face sessions for several reasons. The first reasons are to help participants understand the structure of each session, familiarise themselves with the available music instruments, and enable them to repeat exercises they already know. The final reason is for the music worker to get familiar with the living situation of their participants, for example, to help the participants navigate to plug the keyboard in the wall outlet.

Participants

One participant reported that there needs to be a stable internet connection.

Short-term outcomes on challenging behaviour and well-being

Descriptive statistics for the outcomes on the ABC and the PWI-ID are outlined in Table 1. Improvements were noted on almost all subscales from the ABC and on the total ABC score.

Table 1

Demographic information of the participant and scores on the ABC and PWI-ID

			ABC[b]												PWI-ID[c]	
			Total		Irritability		Lethargy		Stereotypy		Hyperactivity		Inapp. speech		Total	
Participant	Age range	Level of intellectual disability	base	post	base	post	base	post	base	post	base	post	base	post	base	post
1	50-60	mild	21,3	15,4	8,0	7,0	5,3	2,0	0,0	0,0	8,0	6,4	0,0	0,0	64,3	50,0
2	60-70	mild	31,0	26,0	15,0	11,0	3,0	3,0	0,0	0,0	11,0	10,0	2,0	2,0	78,6	85,7
3[a]	70-80	mild	20,0	11,0	6,0	3,0	5,0	2,0	1,0	0,0	3,0	2,0	5,0	4,0	92,9	83,5
4	50-60	mild	13,3	6,0	6,0	3,0	3,0	1,0	0,0	0,0	4,3	2,0	0,0	0,0	85,7	100,0
5	50-60	mild	8,0	9,5	3,0	4,5	3,0	2,0	0,0	0,0	2,0	3,0	0,0	0,0	100,0	78,6
6	50-60	mild	22,0	13,5	4,0	3,0	11,0	6,5	2,0	1,0	4,0	3,0	1,0	0,0	35,7	53,6
7[a]	60-70	moderate	27,0	26,0	13,0	11,0	9,0	6,5	3,0	1,0	2,0	10,0	0,0	3,0	85,7	50,0
8[a]	60-70	moderate	2,0	5,0	0,0	0,0	1,0	5,0	0,0	0,0	1,0	0,0	0,0	0,0	78,6	100,0
9	60-70	mild	17,0	7,0	1,0	1,0	13,0	6,0	0,0	0,0	1,0	0,0	2,0	0,0	78,6	92,9
10	70-80	mild	8,0	6,0	4,0	0,0	0,0	0,0	0,0	1,0	4,0	3,0	0,0	2,0	92,9	100,0
total (mean)			17,0	12,5	6,0	4,4	5,3	3,4	0,6	0,3	4,0	3,9	1,0	1,1	79,3	79,4

Notes: ABC = Aberrant Behaviour Checklist; PWI-ID = Personal Wellbeing Index-Intellectual Disabilities; base = baseline score; post = post-intervention score; inapp. = inappropriate

[a] ABC was filled in by a different caregiver post-intervention than baseline

[b] Higher scores indicate more challenging behaviour

[c] Higher scores indicate a more optimal overall well-being score

Discussion

This pilot study evaluated a partially online music intervention for people with intellectual disabilities. Our findings suggest that such an intervention is feasible and acceptable and can improve challenging behaviour. However, the intervention protocol, trial design and one of the outcome measures will need refinements.

Feasibility

The findings from our study show that the intervention we implemented is indeed feasible. The consent rate seemed rather low, which is not uncommon in the intellectual disabilities research field (range 37–100) (Cleaver & Sakar, 2010). Not all participants participated regularly in online sessions. This finding was due to various factors, for example, participants' bad sight, illness/workload of caregivers and technical issues, and one participant only had face-to-face music sessions because the music worker was not yet vaccinated.

Experiences

Participants appreciated the partially online music intervention. Both the participants and music workers reported positive, joyful experiences with the music sessions, and the overall satisfaction levels were high. Indeed, the participants really looked forward to the music sessions. In addition, the caregivers noted that participants showed a more pleasant and active participation in the group directly after their sessions. It seemed that the participants' musical abilities improved. The observations provided by several music workers highlight a range of attention span and focus levels amongst the participants during the music sessions. However, most of the participants were able to maintain focus for the duration of the hour-long sessions. The feedback from both music workers and participants underscores the relevance of the interpersonal connection between them during the sessions. This sense of relatedness that developed over the sessions was evident, with increased eye contact cited as a key indicator of this connection. In addition, personalisation emerged as a central theme, with an emphasis on tailoring the sessions to the individual preferences and needs of each participant, which is in line with previous research (Van Der Geer et al., 2009; Van Schijndel-Speet et al., 2014).

Additional focus on experiences with online sessions

Overall, the face-to-face music sessions were preferred to online sessions, which is in line with results from other studies, including with people with intellectual disabilities (Rawlings et al., 2021;

St. John et al., 2022). Both the music workers and the participants indicated that it was very important to establish a connection with each other. Starting with face-to-face sessions before scheduling online sessions was experienced as being helpful in this regard. The importance of a prior face-to-face meeting first or working together once a connection is established has also been confirmed in other research (Lunsky et al., 2021), which makes it simpler to adapt interventions from face-to-face to online delivery (Rawlings et al., 2021). Most music workers and participants found the online sessions to be impersonal and that there was a lack of togetherness. According to the music workers, the online sessions were more intense and tiring, both for themselves and the participants, than the face-to-face sessions. In light of this, they recommended shortening the online sessions. Other challenges with online sessions were also identified, including bad internet connections, problems with synchronicity and sound and having to adjust musical methods. Based on these challenges, it is recommended to build capacity and technical know-how regarding the use of straightforward and easy-to-use digital systems amongst care personnel, as suggested by Dowson et al. (2021). It is also important to check whether participants have a medical condition which might impinge upon their ability to engage online (as in the case of the participant with cataracts in this study). In light of these overall negative online music experiences, it is important to consider that our sample consisted predominantly of older individuals. Younger people with intellectual disabilities have, in general, more experiences with technology (Näslund & Gardelli, 2013), which may lead to different findings. Positive experiences of online sessions centred on the fact that they are efficient time-wise and provide a convenient way to reach people with intellectual disabilities, which was also found by Lunsky et al. (2021).

Short-term outcomes on challenging behaviour and well-being

The finding regarding the decrease in challenging behaviour after the music intervention is in accordance with previous research amongst adults with intellectual disabilities (Savarimuthu & Bunnell, 2002), children with intellectual disabilities (Kalgotra & Warwal, 2017), and people with dementia (Han et al., 2010; Pedersen et al., 2017). Moreover, this decrease in challenging behaviour was also apparent amongst participants who received relatively more online music sessions, thus lending further support to the idea that interventions using digital devices can be used to improve challenging behaviour (Moreno, Sans & Fosch, 2021). This pilot study was unable to corroborate the results of previous studies concerning music interventions improving well-being, including amongst people with intellectual disabilities (Savarimuthu & Bunnell, 2002), people with mental illness (Chang et al., 2018), people with schizophrenia (Geretsegger et al., 2017), and those with dementia (Moreno-Morales et al., 2020; Van Der Steen et al., 2017). There are several potential explanations

for this result. First, well-being scores were already high at baseline, which is in accordance with both Jaschke and Scherder's (2015) results and the 'Theory of Subjective Well-being Homeostasis' (Cummins, 2005). Second, the three-point differential scale version of the reduced PWI-ID overly limited the intra-individual variability (Cummins & Lau, 2005), whilst the 10-week period might have also been too short to improve well-being. Nevertheless, it did appear that the participants maintained their well-being, despite the fact that the COVID-19 pandemic was worsening during that time (the increasing number of infected persons had led to more restrictions). Whether or not this intervention contributed to the maintenance of participants' well-being despite the tumultuous circumstances at that time, as is suggested in other research (St. John et al., 2022), remains unclear. Future research with a control group is recommended to explore this further.

Although this study placed more emphasis on the qualitative findings, converging the qualitative and quantitative data leads to a more comprehensive analysis of the research questions (Creswell, 2009). Comparable findings are visible regarding challenging behaviour. Information from the log reports, evaluation questionnaires and final meeting questionnaire showed that the music sessions soothed and relaxed the participants and resulted in more pleasant behaviour in the group directly afterwards. In addition, participants were found to be happy and joyful and felt a sense of connection with the music worker. However, an expected improvement in well-being was not demonstrated and was likely impacted by the aforementioned issues with the PWI-ID and the measurement of well-being. Based on the predominantly negative experiences with the online music sessions, it is crucial to address the implications of these findings. While the sessions overall appeared to have a positive impact on challenging behaviour, as evidenced by both qualitative and quantitative data, and is positively associated with well-being, as evidenced by the qualitative data, most participants did not enjoy the online music sessions. Although caregivers perceived benefits from the intervention (i.e., reduction of challenging behaviour), it is essential to critically evaluate which aspects of the sessions truly benefitted the participants and are worthwhile for them. It is evident that participant enjoyment and engagement are vital factors to consider in designing and implementing interventions tailored to this population. Reducing the number of online sessions to a minimum for those who do not prefer online sessions could be a viable approach to better align with participants' preferences and needs.

There are a number of limitations in this pilot study. Besides the aforementioned limitations of the reduced PWI-ID and lack of a control group, this study was also limited by its small number of participants and their relatively older age, which limited the generalisation of the findings to all

adults with intellectual disabilities. Furthermore, there might be selection bias in our sample because the participants were interested in a music intervention and willing to perform the music sessions, further limiting the generalisation of the results. In addition, the use of a self-constructed questionnaire to gather participant and music worker experiences, presents additional considerations regarding the reliability and validity of our findings. Furthermore, the reliance on self-report measures introduces the possibility of response bias, whereby participants and music workers may provide more favourable or socially desirable responses. In our study, the ABC introduced another additional challenge for potential inter-rater bias, because for three participants, the ABC at post-intervention was scored by another person than at baseline. Despite this difference in raters, the difference in scores for these three participants seemed consistent with the scores from the other participants. Further, one music worker and three participants only experienced one type of session (i.e., face-to-face or online), which limited their feedback only to that type of session. Lastly, we did not collect data on the genre of music that participants or music workers chose during the music intervention nor on the music worker's skillset, which both could have impacted the findings. Music genre preferences are associated with emotional regulation strategies (Cook et al., 2019). Furthermore, given the variability in music workers' experiences and education, future studies should consider using observational measures. This would help investigate the music workers' approach, attitude and skills, which is important for the successful implementation of music interventions (Savarimuthu & Bunnell, 2002) and could shape participants' experiences. Given these limitations, the findings of this study should be interpreted with caution and it calls for replication studies on a wider scale with a more diverse sample.

With regard to future research, the findings of this study suggest some necessary modifications in order to tailor an online music intervention appropriately. For example, it would be valuable to investigate the experiences and effects of a (partially) digital music intervention that starts with face-to-face sessions to build up a relationship first, incorporates shorter online sessions and employs easier digital systems that deal with the problem of synchronicity. These modifications could potentially enhance participant enjoyment and clinical practice. Additionally, further research is needed to examine how individual characteristics interact with the format and content of online music sessions to influence engagement and outcomes. In light of the challenges posed by the COVID-19 pandemic and the negative impact it has had on people with intellectual disabilities, there is a growing recognition of the importance of establishing flexible support options, including online interventions. However, it is crucial to establish a balance and ensure that the shift to (partially) online formats is implemented wisely, particularly for older individuals with intellectual disabilities

who may face additional barriers to engaging with digital platforms. By adapting interventions based on participant feedback and preferences, we can enhance the overall quality of support provided to this population.

Conclusion

Overall, this study explored the feasibility of a partially online music intervention consisting of 16 sessions over a 10-week period for adults with intellectual disabilities living in residential facilities. This initial data shows that both the participants and music workers generally reported positive experiences concerning taking part in the music intervention, and the overall satisfaction of the participants was high. The face-to-face music sessions were preferred to online music sessions. Furthermore, the challenging behaviour of these participants showed improvements, whereas the results concerning well-being were not conclusive. Balancing between making optimal use of online formats and addressing the unique needs and challenges faced by adults with intellectual disabilities is paramount. This study helped in terms of identifying common issues associated with conducting online music sessions for especially older adults with intellectual disabilities and providing recommendations for music workers to successfully adopt this type of support for people with intellectual disabilities. As such, although it was a pilot study with a small sample size, it has important implications for future research in this area given the restrictive measures due to the COVID-19 pandemic and the disparities of access to interventions by people with intellectual disabilities.

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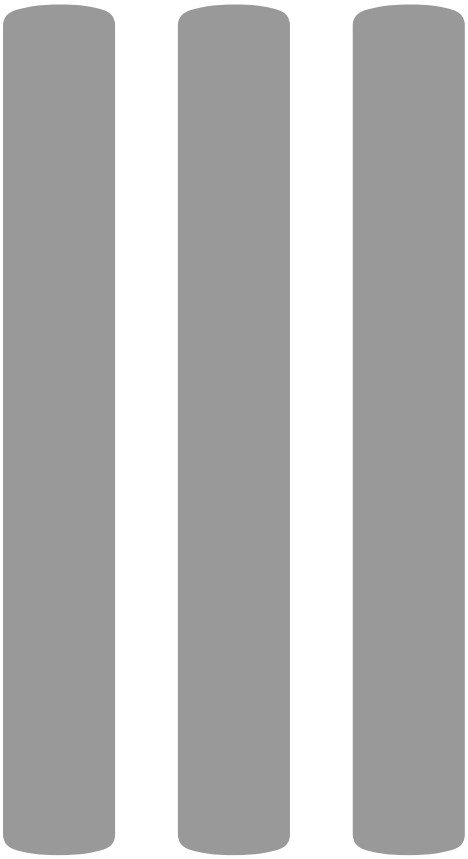
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PART III



PART III

Cluster-RCT

CHAPTER 5

5

Chapter 5

An individual music intervention for adults with intellectual disabilities and challenging behavior: protocol for a randomized controlled trial

Smeets, G., Volkers, K., Scherder, E., & Moonen, X. (2024). An Individual Music Intervention for Adults With Intellectual Disabilities and Challenging Behavior: Protocol for a Randomized Controlled Trial. *JMIR Research Protocols*, 13(1). http://dx.doi.org/10.2196/52497

Abstract

***Background*:** Individuals with intellectual disabilities (ID) are more likely to have problems with executive functioning (EF) and challenging behavior (CB), which are negatively linked to well-being. Among clinical populations, music interventions have been shown to improve various outcome measures, such as CB and EF. Until now, no randomized controlled trials (RCTs) have been conducted to examine the effectiveness of an individual music intervention for adults with ID and CB.

Objective: The study aims to identify the effect and feasibility of an individual music intervention compared with care-as-usual for people with ID and CB.

Method: In this study, a 2-group RCT with a pretest, posttest, and follow-up assessment after 8 weeks is presented. Participants in the music intervention condition will receive 16 individual music sessions within 8 to 10 weeks. The music intervention will be guided by a manual for music workers, in which every session will have a different focus (introduction, emotions, different EF, and end performance). Participants receiving care-as-usual will function as a control group. After the research is finished, they will be offered a budget, which they can spend on musical activities or musical instruments as they wish. Assessments will include caregiver rating scales and self-report questionnaires and tests, which will assess outcome measures of CB, well-being, depression, anxiety, self-esteem, and four domains of EF. A process evaluation will be conducted after the completion of the study, which entails the analysis of data on multiple aspects of the intervention and the study overall.

Results: Enrollment commenced in July 2021, and data collection ended in May 2023. A total of 97 participants were recruited, with 44 participants allocated to the intervention group and 53 allocated to the control group. Data will be analyzed after this protocol has been accepted for publication.

Conclusions: Because there are currently no published RCTs of an individual music intervention for adults with ID and CB, this study will provide insight into the effectiveness of, and experiences with, an individual music intervention for this target group.

Introduction

Background

Intellectual disability (ID) is characterized by significant limitations in both intellectual functioning and adaptive abilities, such as conceptual, social, and practical skills. ID originates before the age of 18 years and can be categorized as mild, moderate, or profound (Diagnostic and Statistical Manual of Mental Disorders (DSM-5), 2013). Besides having a lower IQ and problems with adaptive functioning, people with ID are more likely to develop mental health problems, for example, affective and anxiety disorders and challenging behavior (CB) (Dosen, 2010; Richards et al., 2001). Mental health and CB are negatively linked to quality of life (QoL) (Koch et al., 2015).

People with ID not only have a higher chance of developing mental health problems and CB, but there is evidence that their executive functioning (EF) is also impacted (Zagaria et al., 2021). EF refers to the set of abilities involved in planning, self-monitoring, and purposive action, which are "at the heart of all socially useful, personally enhancing, constructive, and creative activities" (Lezak, 1982, p. 281). There is broad consensus that there are 3 core EFs: inhibition, working memory, and cognitive flexibility (Diamond, 2013; Miyake et al., 2000). Some studies in the ID field use a broader definition that includes attention (Costanzo et al., 2013; Spaniol et al., 2022), planning, and categorization (Costanzo et al., 2013). A few studies suggest that regarding EF, people with ID perform at levels commensurate with their mental age (Henry, 2001; Van Der Molen et al., 2007). Mental age can be calculated from raw scores on intelligence tests: mental age = (IQ score × chronological age) / 100 (Van Der Molen et al., 2009).

Poorer performance on EF tests is linked to more CB, especially externalizing behavior problems, among children, adolescents, and adults with ID. This holds for tests that measure inhibition (Van Rest et al., 2019), working memory (Schuiringa et al., 2015), and cognitive flexibility (Visser et al., 2015). Because CB has a major impact on the lives of people with ID and their caregivers, attempts to help diminish CB cover a wide range of different approaches, from environmental approaches (Tyrer et al., 2017) and support staff training (Randell et al., 2017; Bosco et al., 2019; Hassiotis et al., 2018), to pharmacological interventions (Ramerman et al., 2019). In 2 meta-analyses, effects were found in reducing CB among people with ID for biological and nonpharmacological (such as psychotherapeutic and contextual) interventions (Heyvaert et al., 2010; Bruinsma et al., 2020). Another example of such a nonpharmacological intervention is music therapy, which a recent review found to have a positive effect on CB, anxiety, self-esteem, management of emotions, cognitive measures, and QoL of people

with ID (Mino et al., 2021). However, this review concluded that most of these studies had several shortcomings, such as no control group, small sample sizes, lack of follow-up measurements, and no use of self-reports.

Several empirically valid studies have been carried out with participants who have other cognitive or communication disabilities, such as dementia, acquired brain injury, or autism spectrum disorder (Mulhall et al., 2018). In these studies, beneficial effects of music interventions on various outcomes were found, such as self-esteem (Hillier et al., 2011), anxiety (Hillier et al., 2011; Gómez-Romero et al., 2017; Guétin et al., 2009; Ing-Randolph et al., 2015), CB (Gómez-Romero et al., 2017), mood (Guétin et al., 2009; Särkämö et al., 2014; Gassner et al., 2022), QoL (Särkämö et al., 2014; Gassner et al., 2022), and EF (ie, working memory, attention, and EF in general) (Särkämö et al., 2014). However, even in these well-researched fields, there seems to be a paucity of high-quality empirical studies (Magee et al., 2017; Geretsegger et al., 2022), which emphasizes the need for more rigorous randomized controlled trials (RCTs) with sufficiently large sample sizes studying the different effects of music interventions.

Although music interventions seem effective on CB, anxiety, mood, self-esteem, management of emotions, cognitive measures, and QoL, it is worth noting that the type of music interventions used can vary widely, depending on the goal and the characteristics of the participant. Some interventions are group sessions, whereas others are individual music sessions that can be tailored more to a participant's individual needs and that show tangentially higher effects, for instance, on agitation in persons with dementia (Matziorinis & Koelsch, 2022). Some interventions include music therapy (which involves a trained professional using music to address specific therapeutic goals), music listening, or general music-based interventions (Raglio & Oasi, 2015). It is not uncommon to follow a guideline, with specified principles and procedures for the music intervention, while still leaving enough flexibility to tailor the intervention to the characteristics and needs of the participant and the specific requirements of the situation (Geretsegger et al., 2022). Music interventions range from daily to weekly, lasting 10 to 60 minutes, and can have "passive" (listening) or "active" (making music or sounds together) components (Geretsegger et al., 2022; Hearing, 2018). Active music interventions generally provide greater individual benefits than passive music engagement among older people (Pedersen et al., 2017; Sakamoto et al., 2013), people with autism spectrum disorder (Hearing, 2018), and in the general population (Weinberg & Joseph, 2016).

Overall, evidence suggests that active music participation can enhance one's emotional, psychological, and social well-being and even increases EF (e.g., working memory and attention) in specific clinical populations; however, to the best of our knowledge, no RCTs are available assessing the effectiveness of an individual music intervention for adults with ID, let alone for adults with ID and CB. In this protocol paper, the design and rationale of an RCT are described, in which a manual for an individual active music intervention for adults with ID and CB is presented, matched with persons with ID and CB who receive care-as-usual (CAU) functioning as a control group.

Objectives

Primary objective

The primary objective of the proposed RCT is to evaluate the effect of receiving 16 individual active music sessions compared with CAU in adults with mild or moderate ID with CB on various outcome measures such as CB, well-being, depression, anxiety, self-esteem, and 4 domains of EF.

Secondary objective

A secondary goal of this study is to evaluate the implementation, impact, and context of the individual music intervention with input from participants, caregivers, and music workers.

Method

Study design

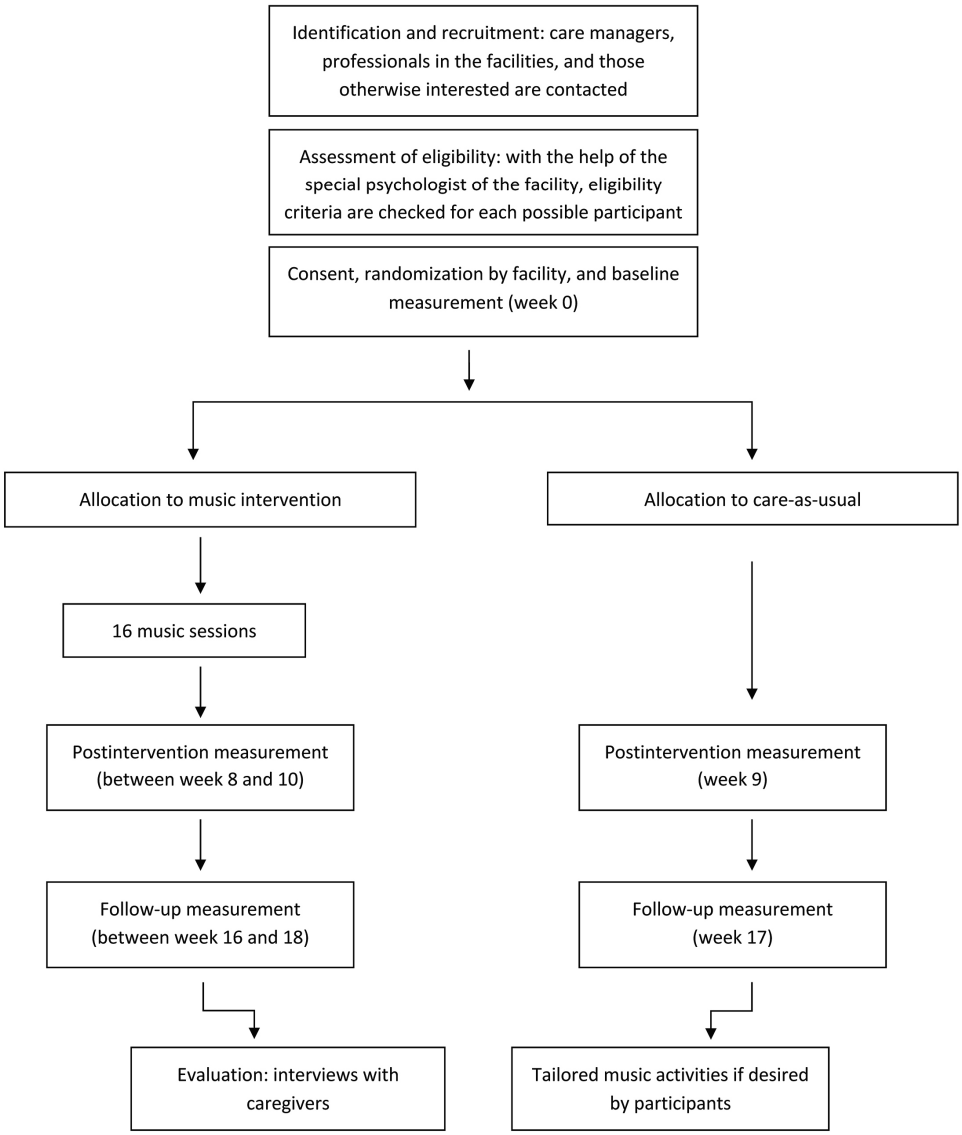
This study is an RCT with a pretest (T1), posttest (T2, 9 weeks after T1, unless the last intervention session was not yet completed; posttest was postponed by 1 or 2 weeks until the week after the final session), and follow-up assessment (T3, 8 weeks after T2), including 2 groups of individual participants (see figure 1).

Participants belonging to the music condition will receive 16 individual music sessions within 8 to 10 weeks, and participants belonging to the control condition will receive (individual) CAU. Randomization will be performed at a facility level for 3 reasons: first, to prevent possible interference between participants belonging to both groups in a facility; second, it is deemed unethical and difficult to explain to people with ID who are assigned to the control condition that they have to wait for the music sessions, whereas fellow residents already receive them; finally, it can also promote recruitment (Tyrer et al., 2017). For randomization, a computerized random number generator will be used. There is no maximum number of participants per facility. Blinding is

not possible because it is clear to the caregivers, participants, and research assistants which type of intervention a participant receives. The independent research assistants are also not blinded because the participants in the music intervention have to fill in an extra questionnaire at the second and third assessment.

Figure 1

Study flowchart



Study population

Participants will be included based on four criteria and three exclusion criteria. The inclusion criteria are as follows: (1) participants must have a mild or moderate ID; (2) they are 18 years or older; (3) they show either internalizing or externalizing CB; and (4) they are mentally competent to give consent to participate in this research. The exclusion criteria are as follows: (1) inability to participate in an intervention for at least one hour; (2) a hearing impairment that cannot be corrected with a hearing aid; and (3) a serious medical condition that limits participation, such as dementia. Already being involved in prior musical activities is not an exclusion criterion.

Procedure

Participants will be recruited from residential facilities for people with ID of the Philadelphia Care Foundation throughout the Netherlands. Because participating in this study requires time and effort from their staff, care managers of facilities serving adult residents with mild to moderate ID are contacted about their willingness to participate in the study. Managers with interest will receive an information letter explaining the research aims and responsibilities of all parties involved. Next, the special psychologist of the facility will check which residents are eligible for inclusion, who then will receive an information letter and an informed consent form in a Dutch, easy-to-read format (Moonen, 2021). Caregivers will be asked to read the letter together with potential participants and help them to fully understand the text, if needed. Legal representatives of the eligible residents will also receive an information letter and an informed consent form, whereas nonlegal representatives of the eligible residents will only receive an information letter. Residents will only become participants in the study if (both) informed consent forms are signed. Subsequently, the special psychologist of the facility will be asked for the participant characteristics, which are age, gender, level of ID, syndrome (when applicable), and comorbid psychiatric diagnoses.

After all assessments are completed, participants will receive a compliment postcard from the Philadelphia Care Foundation and a gift card of €15 (US $17.82) when enrollment commenced.

Intervention

In this RCT, two conditions are compared: a music intervention and CAU.

Music intervention group

The music intervention consists of 16 individual music sessions held twice a week within 8 to 10 weeks. Besides this individual music intervention, regular musical activities that participants already

are involved in before the start of this study will continue, as music participation is a central theme of the Philadelphia Care Foundation. The 16 music sessions will be conducted by a music worker, who can be anyone who is able to play an instrument and is enthusiastic and capable of conducting musical sessions with a person with ID. Music workers are not music therapists because the focus is not on therapy but on a general active music intervention. Before the start of the music intervention, the facility in which it will take place will receive a box with an assortment of different kinds of musical instruments, including rhythm instruments (e.g., cajon, djembe, and small percussion) and harmony and melody instruments (e.g., keyboard and guitar). In general, during the sessions, the music worker adapts the genre or specific pieces of music to the wishes and preferences of the participant. The music intervention will be guided by a training manual for the music workers, in which the frequency, average time per (elements of the) session, and the content of every music session are outlined. The manual is designed based on the input of different advisory groups with professional music coaches, caregivers, and people with ID.

In the manual, intervention procedures are specified, such as the setting, general goals, contents of the music sessions, basic principles of the intervention, and exemplifications. An outline of the music session can be seen in Table 1. Every session will be centered around a different aspect (introduction, emotions, different EF, and end performance), and in the training manual, example exercises are described, focusing on every aspect. These examples are provided by advisory groups of professional music coaches and stem from different other manuals (Het Koninkrijk Concertgebouw, 2016; Houtman et al., 2017; Storms, 2005). Each session will start with a welcome song, after which a listening exercise will take place. Then, a warm-up of the voice and body is scheduled, and subsequently, participants actively engage in music activities by singing or playing an instrument, after which a cool-down and a farewell song are performed. The manual provides a fixed frame for the intervention, but it leaves enough flexibility to tailor the intervention to the characteristics and needs of every participant. Participants will receive a diploma at the final session.

To study for exploratory purposes the impact of the music sessions' frequency, some participants will receive extra music sessions, resulting in 24 to 27 sessions within the same time period, aiming at approximately three sessions a week.

Table 1

Outline music session

Music Intervention	Time (minutes)	Attendees
Preliminary meeting	30	Music worker, participant, and caregiver
Music session • 1: Welcoming or introduction • 2-6: Emotions • 7+10: EF[a] (Inhibition) • 8+11: EF (Cognitive flexibility) • 9+12: EF (Working memory) • 13-15: Working toward the end performance or song • 16: End performance or song	60	Music worker and participant
Final meeting	30	Music worker, participant, and caregiver

[a] EF: executive functioning.

CAU group

Participants receiving CAU will function as a control group. CAU includes regular care such as assistance with acts of daily living and day care activities. CAU can include musical activities that participants already performed before the start of this study, as music is a central theme of the Philadelphia Care Foundation. However, new individual musical activities will be postponed for these participants until after the follow-up measurement. Participants receiving CAU will be offered a budget at the end of the study, which they can spend on musical activities or musical instruments according to their own wishes, with the help of a musical specialist who works at the Philadelphia Care Foundation.

Measures

All assessments (T1, T2, and T3) will take place by applying caregiver rating scales and self-report questionnaires and tests, and all will be conducted in the same week. Students from several Dutch universities will work as research assistants assisting with the data collection. Research assistants receive theoretical and practical training in assessment through a training program in the test administration protocol. They will administer all irregularities and deviations from this protocol in a study log report. The research assistants will not be involved in the intervention itself. An overview of the outcome measures and the process evaluation can be seen in Table 2.

Table 2

Overview outcome measures and process evaluation

Assessment	Proxy or self-report			Measurement moment				
	Caregiver	Music worker	Person with ID[a]	Each music session	Baseline (T1)	Postintervention (T2)	Follow-up (T3)	After follow-up
Outcome measures								
Self-reports and tests			x		x	x	x	
Proxy questionnaires	x				x	x	x	
Evaluation[b]								
Evaluation questionnaire music sessions		x	x			x		
Follow-up evaluation	x		x				x	
Self-report with emoticons			x	x				
Log reports music sessions		x		x				
Semi-structured interview	x							x

[a]ID: intellectual disabilities.
[b]Only in the music intervention.

Outcome measures

Challenging behavior

The Dutch version of the Aberrant Behavior Checklist (ABC) will be used to assess CB, and the form will be filled out by a caregiver (Aman et al., 1985). The ABC consists of 58 items, which are rated on a 4-point Likert scale, ranging from 0 ("not at all a problem") to 3 ("the problem is severe in degree"). The sum of the questions provides a total score (range 0-174), with higher scores indicating more CB. In this study, the cluster structure as suggested by Kaat et al. (2021) will be used. The first cluster (28 items; α=.96, β=.57) represents behavior that is directed outward (i.e., externalizing CB). The second cluster (26 items; α=.93, β=.76) represents behavior that is directed inward (i.e., internalizing CB). For externalizing CB, the score ranges from 0 to 84, and for internalizing CB, the score ranges from 0 to 78. The total score on cluster 1 (i.e., externalizing CB) will be used to assess externalizing CB, the total score on cluster 2 (i.e., internalizing CB) will be used to assess internalizing CB, and the total score on all items of the ABC will be used to assess overall CB.

Well-being

Subjective well-being will be assessed using the Dutch version of the 7-item Personal Well-being Index—Intellectual Disability (PWI-ID) (Cummins & Lau, 2005). An example of an item is as follows: "How happy do you feel about the things you have? Like the money you have and the things you own?" Each item is scored on a 3-point Likert scale (0=sad, 1=neither happy nor sad, and 2=happy). Three pictures of colored smileys are used to represent the 3 answers. The pretesting protocol, which screens for acquiescent responding, will not be applied, to diminish the data collection burden for participants. PWI-ID scores range from 7 to 24, with higher scores indicating a better subjective well-being. The PWI-ID seems to be an appropriate measure for people with a mild or upper moderate level of ID (Cummins et al., 2010).

Depression and anxiety

Symptoms of depression and anxiety will be assessed using the Depressed Mood and General Anxiety subscales from the Dutch version of the Anxiety, Depression and Mood Scale (Hermans et al., 2018). This questionnaire will be completed by the caregiver. The Depressed Mood subscale consists of 13 items, for example, "somber mood," which are scored on a 4-point Likert scale (0=behavior has not occurred or is not a problem, 1=behavior sometimes occurs or is a slight problem, 2=behavior often occurs or is a relatively large/moderate problem, and 3=behavior occurs a lot or is a severe problem). Depression scores can range from 0 to 39. The General Anxiety subscale consists of 7 items, for example, "nervous or anxious," and is also scored on this 4-point Likert scale. Anxiety

scores range from 0 to 21. For both subscales, a higher score indicates more symptoms. Its reliability and validity are satisfactory to good (Esbensen et al., 2003).

Self-esteem

Self-esteem will be assessed using the 5-item Global Self-Worth subscale of the Self-Perception Profile for Adolescents (Harter, 1988). In this study, the Dutch adaptation of the Self-Perception Profile for Adolescents (Treffers et al., 2002) will be used, with a Cronbach's α of >.70 and 1 statement per item (Wichstrøm, 1995). An example of an item is "I am quite happy with myself." Each item is scored on a 4-point Likert scale (1=completely untrue for me, 2=a little untrue for me, 3=a little true for me, and 4=completely true for me). The total score can range from 5 to 20, and a higher score indicates higher self-worth.

EF tasks

Overview

EF will be assessed on a tablet using three computer-based games to assess cognitive flexibility, attention, inhibition, and working memory, which were custom-developed in earlier research and are adapted to the needs of individuals with ID, that is, fewer trials, shorter tasks, visual support during test instructions, and attractive stimuli (Van Rest et al., 2019; Schalock et al., 2010). After each game, the research assistant will complete two questions about how often the participant needed to be motivated and how often the participant was distracted during the games.

Cognitive flexibility

The game to assess cognitive flexibility is based on the flanker task (Eriksen & Eriksen, 1974). The principle of the flanker task is that participants have to respond to a target arrow, which is flanked by distractor arrows. In this study, this task will consist of five arrows per trial, with 32 random trials per condition, across three conditions. In the first condition, there are five green arrows pointing in different directions (left or right). Participants have to ignore the irrelevant stimuli and press the button in the same direction as the green arrow in the middle. In the second condition, the participants are presented with red arrows. Unlike the previous condition, participants have to press the button with the opposite direction as the arrow in the middle. In the third condition, participants are presented with alternating trials of red or green arrows and are requested to press the button with the opposite direction if the arrow in the middle is red and to press the button with the same direction if the arrow in the middle is green. All three conditions will have a practice round before

the official task. The score used to measure cognitive flexibility consists of the correct responses in the third condition (range 0-32). A higher score implies a higher degree of cognitive flexibility.

Attention

Attention will be assessed with the first condition of the already mentioned flanker task, where participants have to respond to the green arrow in the middle, regardless of the nature of the flanking distractor items, by pressing the right button for right-faced arrows and the left button for left-faced arrows. Both correct and incorrect (ie, missing or lack of on-time response) responses will be added, standardized as z scores, to create a composite total attention score. The incorrect responses will be reversed to create a variable in which a higher score represents better attention.

Inhibition

Inhibition will be assessed using two different tasks: (1) the second condition with red arrows of the already mentioned flanker task, which measures interference control (Eriksen & Eriksen, 1974) and (2) the go or no-go paradigm, which measures the inhibition of a prepotent motor response (Trommer et al., 1988). The z scores on both outcome variables will be added to create a composite total inhibition score for each participant. This new variable will be reversed, so a higher score represents better inhibition skills. The outcome variable from the flanker task that will be used is the number of faults in the task with only red arrows, representing the inability to suppress an initial response. In this task, participants have to press the arrow corresponding to the opposite direction of the central arrow.

The go or no-go task consists of two parts. In the first part, the participant is requested to press the button as soon as a green apple, that is, a “go” stimulus, appears on the screen. In the second part, participants have to tap on the button when the green apple appears, but they have to inhibit their response when a red cross appears through the apple, that is, a “no-go” stimulus. An incorrect answer is given when the participant presses the button, and the green apple appears with a red cross. Both conditions had a practice round before the official task. The outcome measure for the go or no-go task is the total incorrect answers in part two.

Working memory

To assess the working memory construct, a visuospatial computerized task, based on the Klingberg principles for working memory, will be used (Klingberg et al., 2002; Olesen et al., 2004). In the Klingberg task, participants have to remember the order that a circle shifts over a 4-by-4 grid of open

squares. The task consists of two conditions that both start with an instruction and a 4-trial practice session before the regular session. The regular session starts with a pattern of two circles on adjacent units and gradually increases in length and difficulty with each trial. Each trial consists of two patterns of the same length and difficulty. In the first condition, patterns with a green circle are presented to the participants who have to tap on the squares in the grid in the same order afterward (forward version). In the second condition, patterns of red dots are shown, which participants have to tap in the reverse order afterward (backward version). When a participant taps a wrong pattern twice on a trial of the same length and difficulty, the task ends. In this study, the average number of total correct trials in the forward and backward task will be used as a measure of working memory (Klingberg et al., 2002; Thorell et al., 2009). A higher score represents better working memory. There is no absolute maximum score; in theory, the task can go on indefinitely.

Process evaluation

Overview

Because conducting clinical trials in the field of adults with ID and CB seems to be challenging (Mulhall et al., 2018), it is also important to share the practical experiences when conducting trials using a process evaluation (Moore et al., 2015). For conducting a process evaluation, different frameworks are developed, which all focus on several process evaluation components. For this study, the following frameworks are used: the Medical Research Council guidance (Moore et al., 2015), process evaluations for cluster-randomized trials of complex interventions as proposed by Grant et al (2013), and the RE-AIM (Reach, Effectiveness, Adoption, Implementation, and Maintenance) framework (Glasgow et al., 1999; 2019). The Medical Research Council guidance and the RE-AIM framework have recently been used in the ID research field (Bosco et al., 2019; Hanzen et al., (2021; Lai et al., 2019; Schaap et al., 2018).

Evaluation per music session

The music worker will ask the participant before and after each music session how he or she feels at that moment: happy, sad, scared, angry, or tired. Sheets representing five emoticons with associated feelings are shown to help the participant. If possible, the participant also indicates whether this feeling is little, normal, or intense. When it is too difficult for a participant to choose a specific emotion, the participant is asked whether he or she feels pleasant or nonpleasant. Music workers will also complete a log report after each music session. The log report contains questions about that specific session: whether the session was performed according to the manual and how much time was spent on active music making.

Evaluation of music intervention

Participants in the music intervention and music workers will be asked to complete an evaluation questionnaire after the last music session. These questionnaires are designed to gain the participants' personal views about the music intervention and their satisfaction with specific elements of it (eg, musical instruments and frequency of the sessions). The music workers will share their observations regarding behavioral, cognitive, and emotional changes within a participant during sessions.

After the last music session, caregivers will be asked to participate in a semi-structured interview. This interview will focus on observed changes in the participants' behavior and the experiences of caregivers with this music intervention study. Topics that will be covered are related to acceptability or satisfaction, reach, and context. The interviews will be recorded with the permission of the respondents and transcribed.

Follow-up evaluation

The participants in the music intervention and caregivers from participants in the music intervention will answer a few questions about the long-term use of musical activities after the last music session. For participants, this questionnaire will be conducted at T3, and for caregivers, it will be conducted 6 months after the last session.

Sample size calculation

The sample size is calculated using G*Power software (version 3.1.9.7) (Faul et al., 2009). The sample size calculation is based on a small effect size of .2, a statistical power of .95, and a type I error probability of α=.05, which results in an unadjusted sample size of 67 participants. Because cluster randomization negatively affects the power, the sample size needs to be adjusted. With the average cluster size of two participants per residential facility and allowing an intraclass correlation of .2, the adjusted sample size is 80. To allow for a dropout rate of 20% over the course of the study, we will aim at recruiting a total of at least 96 participants.

Data analysis

Participant characteristics will be compared between the group receiving the individual music intervention and the group receiving CAU. Further analyses will be adjusted for any potential significant differences. The primary analyses will be undertaken on an intention-to-treat basis, and 2-sided tests will be applied at a 5% α level. In addition, per-protocol analyses will be conducted based

on the sample of participants who adequately adhered to the intervention protocol by completing at least 12 of the 16 music sessions (ie, 75%), which is similar to 67% and 80% of two other intervention studies among adults with mild to moderate ID (Martínez et al., 2019; Willner et al., 2013). In addition, by setting the threshold at 12 sessions, we know for sure that all intervention participants followed at least one or two sessions with a focus on emotions and EF.

To minimize the number of analyses on outcome domains, the 4 different EF measures will be converted into z scores and, according to factor analysis, summed up into an EF domain if possible. Subsequently, to test differences in changes from baseline to follow-up between the 2 conditions, a linear mixed model analysis for each treatment variable will be used. Within each analysis, only time and the interaction between the treatment variable and time will be used (Twisk et al., 2018), making it possible to adjust for the dependence of the repeated observations within the subjects. The effect of the intervention on the two follow-up measurements will be assessed. This analysis allows for participants with only a baseline measurement but with missing data at follow-up to be included. The normality of the residuals from the linear mixed model analysis will be visually inspected.

Secondary analyses will be performed as well. The effect of extra music interventions will be tested applying a Mann-Whitney U test for the between-group comparisons. The self-reported feelings before and after each music session will be compared using a Wilcoxon signed rank test. Descriptive statistics will be used to summarize the quantitative data of the log reports, evaluation questionnaires, and the follow-up evaluation.

Qualitative data of log reports, evaluation questionnaires, follow-up evaluation, recruitment log, trial log, and semi-structured interviews will be analyzed using thematic context analysis on multiple aspects of the intervention and the overall study process, including recruitment, retention, acceptability or satisfaction, maintenance, dose, fidelity, reach, adaptation, and context.

Ethical considerations

Participation in the study will be voluntary. Participants and their legal representatives can withdraw consent at any time during the intervention and have the right to demand the removal of the original data. Random allocation of participants to one of the two conditions is considered reasonable as no adverse effects are expected in any of the conditions. Inconveniences caused by the necessity to attend two or three music sessions per week are considered tolerable in view of the anticipated benefit for the participant receiving music sessions. Participants assigned to CAU will receive a

considerable budget after the study has ended, which they can spend on musical activities or musical instruments according to their own wishes. Ethical approval for this study was granted by the review board of the Faculty of Behavioral and Movement Sciences of the VU University Amsterdam (protocol VCWE-2021-081). The Medical Ethical Committee of the VU University Medical Center stated that the research was not subject to the Dutch Medical Research Involving Human Subjects Act. The trial has been registered in the International Clinical Trial Registry Platform (NL8482).

Data management

To ensure the accuracy of the data, all entered data will be triple-checked by different research assistants. Every participant will be assigned an anonymous code after their informed consent form is received to store and identify their trial data. The coding key and person-related data will be saved in a password-protected file in a restricted folder, which will only be accessible by the study-related researchers. All paper files will be kept in locked cabinets in the researcher's office for the duration of the study, and digital files will be stored on a password-protected and secure system, which is also exclusively accessible by the study-related researchers.

Results

Enrollment commenced in July 2021, and data collection ended in May 2023. The aim to recruit at least 96 participants has been achieved with a total of 97 participants, of whom 44 were randomized in the intervention group. Data will be analyzed after this study protocol has been accepted for publication.

Discussion

In this paper, the design of an RCT is presented, which aims at exploring the effectiveness of and experiences with a music intervention for adults with ID and CB on various outcome measures, including CB, well-being, depression, anxiety, self-esteem, and EF. The first strength of this study is that it concerns a clustered RCT design with a sufficient sample size. The second strength is that the multisource data collection, including the use of self-report questionnaires and tests as well as proxy measures, improves the research quality. Third, because distal outcomes (measured over a longer period of time) and proximal outcomes (measured immediately after the intervention) will be used,

immediate effects can be assessed too. Lastly, as qualitative data are collected alongside quantitative data, further insight into the process or mechanisms of change can be gained.

However, some limitations also have to be taken into consideration. First, blinding is not possible in the design because the nature of the intervention and the different questionnaires at the data collection points for both groups precludes blinding of the assignment for all parties involved. The absence of an active control condition might entail difficulties in ascribing possibly disclosed effects to power factors of the music intervention, such as level of attention (Vink et al., 2013). Furthermore, there are several anticipated challenges, which are based on previous work conducting RCTs with adults with ID. These include challenges with recruiting adequate numbers, resistance to the use of control groups, assessing participants through gatekeepers (ie, the caregivers), and staff turnover that impacts the data collection (Mulhall et al., 2018). The following steps to minimize attrition are considered in this study: (1) the research team follows a flexible participant-led approach to gathering all data—with contacts being at times suggested by participants and at the convenience of the facility of the participant; (2) the incentive to complete all questionnaires and tests is increased with the compliment card that participants receive after every data collection measurement; (3) caregivers will receive multiple reminders, either live, via mail, or telephone, to return the questionnaires on time; and (4) the combination of collecting data from both participants and caregivers reduces the burden for them and diminishes the impact of participant dropout or staff turnover.

In sum, more evidence-based interventions are needed to help improve the mental well-being of this population. It is important to not only fill this evidence-base gap with the outcomes of the ID RCTs but also share the practical experiences conducting trials through the reporting of process evaluations. Therefore, it is expected that this study will provide insight into the effectiveness and experiences of an individual music intervention for adults with ID and CB. If the intervention proves to be effective, broad-scale implementation in the care for people with ID can be considered and results can guide further music intervention guidelines and enrich the work environment of music workers engaging in supporting people with ID.

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CHAPTER 6

6

Chapter 6

The impact of a music intervention on challenging behaviour, mental health and well-being in adults with intellectual disabilities: a cluster-randomized controlled study

Smeets, G. J. J., Volkers, K. M., Hoozemans, M. J. M., Scherder, E. J. A., Moonen, X. M. H. (2024). *The impact of a music intervention on challenging behaviour, mental health and well-being in adults with intellectual disabilities: a cluster-randomized controlled study* [Manuscript submitted for publication].

Abstract

Background: This cluster-randomized controlled trial examined the effects of an individual music intervention on challenging behaviour, mental health and well-being in adults with mild or moderate intellectual disabilities.

Method: Participants were randomized into an individual music intervention condition (n=44), receiving 16 active music sessions over eight weeks, and a care-as-usual condition (n=53). Outcomes were measured at baseline, post-intervention and eight weeks follow-up and analysed with linear mixed models.

Results: The individual music intervention showed significant improvements in challenging behaviour at post-intervention and follow-up, and reductions in anxiety and depression at follow-up. No significant changes were observed in well-being and self-esteem.

Conclusion: An individual music intervention may effectively reduce challenging behaviour and improve mental health in adults with mild or moderate intellectual disabilities, broadening intervention options for this population.

Introduction

For several decades, the relation between music, health and well-being has been studied across different populations (e.g., Dingle et al., 2021; Macdonald, 2013; Raglio & Oasi, 2015; Weinberg & Joseph, 2017). Listening to or making music is associated with enhanced emotional and social well-being, including positive emotions and self-esteem (Daykin et al., 2016; Hays & Minichiello, 2005; Joseph & Southcott, 2014) and can be used as an intervention for emotion regulation (Cook et al., 2019; Van Goethem & Sloboda, 2011). Experimental studies in the general population show that music interventions can improve quality of life (QoL) (McCrary et al., 2022), can lead to stress reduction (De Witte et al., 2020), can reduce depression and anxiety (Geipel et al., 2018) and can evoke activity changes in the core brain regions that underlie emotion (Koelsch, 2014). In several systematic reviews and meta-analyses, music interventions in people with dementia have shown to improve QoL (Vasionyte & Madison, 2013), can diminish depression (Ting et al., 2023; Van der Steen et al., 2018), anxiety (Ting et al., 2023), behavioural problems (Van der Steen et al., 2018) and agitation (Bartfay et al., 2020; Pedersen et al., 2017; Ting et al., 2023). Furthermore, active music making is associated with improving mood in people with dementia (McDermott et al., 2014; Osman et al., 2016).

Positive associations between music, behaviour, mental health and well-being have also been observed in people with intellectual disabilities (Jackson, 2007). Indeed, engagement in music was found to be associated with a better QoL (Andring, 2015; Jaschke & Scherder, 2015; Morris, 2014), higher self-esteem (Lewis, 2019; MacGlone et al., 2020; Wilson & MacDonald, 2019), better mood (MacGlone et al., 2020) and less challenging behaviour (CB) (Jaschke & Scherder, 2015). Musical activities positively affect CB in children with mild and moderate intellectual disabilities (Kalgotra & Warwal, 2017). In a review on music and people with intellectual disabilities covering research published between 1943-2006, it was found that there were several reported benefits of introducing music to people with intellectual disabilities, concerning communication, relations with others, ability to cooperate, peer acceptance and enhancing cognitive capacities, but also on physical, emotional, and psychological (expressing emotion, alleviating agitation, pleasure, self-esteem) functions (Hooper et al., 2008). However, the designs of the studies mentioned above concerning music and people with intellectual disabilities were almost all either qualitative (Jackson, 2007; MacGlone et al., 2020; Wilson & MacDonald, 2019) or cross-sectional (Jaschke & Scherder, 2015) and some even not peer-reviewed (Andring, 2015; Lewis, 2019; Morris, 2014). In other words, the assumption of a causal

relationship between music and mood, well-being, self-esteem and CB needs robust scientific support.

In a recent scoping review on music interventions in people with intellectual disabilities it was found that there were more qualitative than quantitative studies published (Mino-Roy et al., 2021). The number of studies using experimental designs seems to increase, albeit with a substantial increase of single-subject designs (Brown & Jellison, 2012). Savarimuthu and Bunnell (2002) performed a literature review about the effects of music on people with intellectual disabilities and concluded that it reduces CB and improves QoL. However, in their review no distinction was made between quantitative and qualitative studies. Nevertheless, these findings are interesting, since around 10 to 18% of people with intellectual disabilities display CB (Bowring et al., 2017; Lowe et al., 2007) and this is negatively linked to well-being and has a huge impact on caregivers. For example, caregivers experience stress (Raczka, 2005) and are more likely seeking to leave their employment (Robertson et al., 2005). A recent scoping review highlighted a significant reduction of anxiety through musical activities in young people with intellectual disabilities (Després et al., 2023). This is important since people with intellectual disabilities have a higher risk of developing mental disorders, such as anxiety and depression (Garaigordobil & Pérez, 2007). To our knowledge, no studies have examined the effects of music interventions on mood and self-esteem in people with intellectual disabilities. This is a clinically relevant question, since the self-esteem in adults with intellectual disabilities seems to be lower than in the general population (Garaigordobil & Pérez, 2007) and low self-esteem is associated with depression (Dagnan & Sandhu, 1999).

Music interventions in people with intellectual disabilities, living in a residential community home, are often unstructured (Jaschke & Scherder, 2015). The design of music interventions for people with intellectual disabilities varies, ranging from therapeutic sessions with licensed music therapists to engagement in musical activities with music workers. Multi-activity musical interventions, involving improvisation, instrument playing or singing, have shown significant results compared to other musical interventions (Després et al., 2023).

Since most experimental studies concerning music interventions in people with intellectual disabilities are non-randomized and have small sample sizes (Duffy & Fuller, 2000; Kalgotra & Warwal, 2017), there is a need for evidence-based research incorporating a structured, multi-activity intervention, with a control condition, follow-up measurements and self-report measures instead of only proxy measures (Mino-Roy et al., 2021). Therefore, the aim of the present study was to

investigate the effectiveness of an individual music intervention conducted by a music worker in adults with intellectual disabilities and CB compared to care-as-usual (CAU), on CB, well-being, anxiety, depression and self-esteem. Additionally, the study aimed to assess whether treatment success was modified by the level of intellectual disabilities or baseline CB. Secondary objectives were to examine (a) the influence of the intensity of the intervention on the same outcome measures, and (b) – in a non-comparative design – the impact of the music intervention on the participants' feelings.

Method

Study design

A cluster randomized controlled trial was performed including baseline, post-intervention and eight weeks follow-up assessment. Cluster randomization at a facility level was chosen to minimize contamination and for ethical and practical reasons. When in a facility informed consent was given by the people with intellectual disabilities stating that they were willing to participate in the project, the facility was randomised by using a computer-generated randomization list (randomizer.org) to either receiving the music intervention condition or being in the control condition which meant that CAU was continued to be provided to all the participants within that facility. Participants and researchers were not able to influence allocation. Because of the nature of the intervention, blinding was not possible. The research assistants that collected the data could also not be blinded, since the participants in the two conditions completed different questionnaires at different times. As the Medical Ethics Committee of the Amsterdam UMC stated that the research was not subject to the Dutch Medical Research Involving Human Subjects Act, ethical approval was received by the review board of the Faculty of Behavioural and Movement Sciences of the Vrije Universiteit Amsterdam (protocol #VCWE-2021-081). The trial was registered prior to data collection in the International Clinical Trial Registry Platform (main ID #NL8482). More information can be found in the study protocol (Smeets et al., 2024).

Procedure

Participants were recruited from residential facilities (n=96) or an ambulant setting (n=1) of Philadelphia Care Foundation throughout the Netherlands. Several recruitment strategies were used, such as advertisement in a newsletter, a webinar to attract caregivers to refer eligible participants, presentations at management meetings and special psychologists were contacted to refer potential participants. Several facilities chose not to participate due to various reasons, for example no interest

or enthusiasm (from caregivers and/or residents), staffing problems, facility-dependent reasons (e.g., too much turmoil at facility) or relocation of possibly eligible participants.

Written informed consent was obtained from all participants and, if applicable (n=52), also from their legal representatives. After the informed consent procedure was concluded, the caregiver of the facility received information by mail about the intervention allocation and the research assistant contacted the facility to schedule baseline measurement. Participant characteristics (gender, age, level of intellectual disabilities, possible psychiatric diagnoses and syndromes) were provided by the special psychologist of the facility. After the last measurement was completed, every participant received a small monetary gift card.

Eligibility criteria

Participants were eligible for participation based on four inclusion criteria; They (1) had to be 18 years or older, (2) had to have mild or moderate intellectual disabilities (IQ 36-70), (3) had to display CB and (4) had to be mentally capable to give (informed) consent. People were excluded who (1) were not capable of participating in a one-hour long intervention session, (2) had hearing problems that could not be reduced via the use of a hearing aid or (3) had a medical condition that would hinder their participation. Based on these criteria, a special psychologist determined one's initial eligibility for participation.

Intervention

Music intervention

The music intervention consisted of 16 individual music sessions, twice a week, within a period of eight to ten weeks. During the sessions, participants were actively involved in music-making by playing on different musical instruments (e.g., keyboard, bongo, guitar, xylophone) and/or singing. The music sessions were guided by music workers. A music worker could be anyone who was able to play an instrument and was enthusiastic and capable to conduct musical sessions with a person with intellectual disabilities. The music intervention was provided in accordance with a training manual for the music workers, in which the setting, goals and content of the music intervention was outlined. Prior to the first music session, every music worker, participant and the caregiver from the facility who served as the contact person throughout the study had a short meeting in which they got acquainted with each other and discussed all details of the music intervention. Following each music session, the music worker briefly documented some process measures (e.g., active music making time, adherence to session guideline, feelings before and after the music session from the participant

and important observations). To study the impact of the music sessions' frequency five participants received additional music sessions (varying between 24-27 sessions) within the same time frame.

Care-as-usual

Participants in the control condition continued to receive their usual care and support. If before the start of this study musical activities were already part of this care and support, this could be continued. However, new musical activities between baseline and follow-up were not allowed. After follow-up participants in the control condition received a budget for musical activities or instruments according to their own wishes, after consulting their principal caregiver and a music coach from the organisation.

Outcome measures

Outcome measures were collected, based on proxy and self-report assessments. Caregivers received the questionnaires by mail. Self-report questionnaires were collected in the facilities of the participants and were filled in by the participants themselves, if necessary, assisted by the research assistants who visited the facilities. Outcome measures were collected at baseline (T1), in the week after the last music session for the intervention condition and nine weeks after the baseline measurement for the control condition (T2) and at follow up, which was eight weeks after the second measurement (T3).

Proxy measures

CB was assessed using the Aberrant Behaviour Checklist (ABC) (Aman et al., 1985). The ABC is a 58-item rating scale with each item rated on a 4-point Likert scale. Higher scores indicate more problems. In addition to the total score (range 0 – 174), a score for externalizing CB (range 0 – 84) and for internalizing CB (range 0 – 78) was calculated (Kaat et al., 2021). In several studies the psychometric properties of the ABC was demonstrated to be psychometrically sound (for an overview see Farmer & Aman, 2021).

Anxiety and depression were assessed with the Anxiety, Depression and Mood Scale (ADAMS) of which the subscale General Anxiety (range 0 – 21) and the subscale Depressed Mood (range 0 – 39) were used. Higher scores indicate more problems. The ADAMS has been found to be a psychometrically sound instrument (Esbensen et al., 2003). This is a 28-item questionnaire with items rated on a 4-point Likert scale.

Self-report measures

Well-being was assessed using the Personal Well-Being Index – Intellectual Disability (PWI-ID) (Cummins & Lau, 2005), which has shown good reliability and validity (McGillivray et al., 2009). The PWI-ID assesses satisfaction on seven life dimensions. A reduced-choice format using three Likert-type responses (i.e., emotions corresponding with the answer) was used for all participants. The PWI-ID generates an overall score from 0 to 100, with higher scores indicating better well-being.

Self-esteem was assessed using the 5-item subscale Global Self-Worth of the Self-Perception Profile for Adolescents (SPPA) (Harter, 1988; Treffers et al., 2002). An altered version with one statement per item was used (Wichstrøm, 1995). The subscale Global Self-Worth in this revised version has an internal consistency of .77 (Wichstrøm, 1995). Each item is scored on a 4-point Likert scale and the total score for this subscale ranges from 5 to 20. A higher score indicates better self-esteem.

In addition, participants in the music intervention condition were asked about their feelings before and after each music session by the music worker (on the dimensions happy, sad, scared, angry, tired or in general pleasant/non-pleasant). If possible, the participant also indicated the intensity of this feeling (little, normal or intense).

Statistical analyses

Based on a small effect size of 0.2, a statistical power of 0.95, a type-I-error probability of $\alpha = 0.05$, cluster size of 2, an intra-class correlation of 0.2 and a drop-out rate of 20%, 96 participants were needed to detect a significant change (Smeets et al., 2024). Baseline characteristics were reported in the descriptives. Differences between characteristics of the intervention and control condition were tested by chi-square tests (for categorical variables) and independent sample t-tests (for continuous variables). If differences were found, they were reported and were controlled for in further analyses. Baseline differences between all outcome measures were tested with an independent sample t-test. Missing values on the ABC were replaced with the mean of the corresponding subscale for that participant. Missing values on the ADAMS were replaced with the mean of the corresponding subscale when there were less than two missing values on that subscale. For the PWI-ID all questionnaires with only extreme values (i.e., all answers were either ‘1’ or ‘3’) were removed and in the case of missing values the total score was divided by 6 (instead of 7). Subsequently, extreme outliers (Q3 + 3 * IQR) were winsorized by replacing them by the highest or lowest score of the other participants that fell within the normal range.

To answer the research question whether an individual music intervention in adults with intellectual disabilities leads to an improvement on CB, well-being, anxiety, depression and self-esteem

compared with CAU, five linear mixed models (one per each outcome measure) were constructed. The model (equation 1) included time (with three levels: baseline, post-intervention and follow-up) represented by two dummy variables and the interaction between the treatment variable and the two dummy variables for time, from which the treatment effects (at post-intervention and follow-up) could be directly obtained (Twisk et al., 2018).

$$(1)\ Y = \beta_0 + \beta_1 age + \beta_2 dummytime1 + \beta_3 dummytime2 + \beta_4 groupxdummytime1 + \beta_5 groupxdummytime2$$

An advantage of this type of analysis is that subjects with only a baseline value, but with missing data at the two other measurements are still part of the analysis (Twisk et al., 2018). A significant interaction of time and treatment indicated that there was a significant treatment effect for either one of the two times (T2 or T3) on the specific outcome measure. Facility was a potential hierarchical level in the linear mixed model. When a random intercept for facility improved the model, which was tested by means of the likelihood ratio test, this was added to the analyses in order to adjust for the clustering of the data (Twisk, 2006). If a significant treatment effect (i.e., the interaction of treatment with time) for the total CB score was found, two additional linear mixed model analyses were conducted with internalizing CB and externalizing CB as outcome measures. In this way, it would be possible to study on which specific subtype of CB the intervention had the most effect. Furthermore, to explore whether treatment success was different for participants with various levels of CB at baseline, the interaction between treatment and baseline CB was added to the linear mixed model analyses. Likewise, linear mixed model analyses with the interaction between treatment and level of intellectual disabilities (mild vs. moderate) were performed to explore whether the treatment effect was modified by level of intellectual disabilities.

Assumptions for linear mixed model analyses were conducted by inspecting the residuals via different graphs (histograms, Q-Q plots and box-and-whisker plots) to confirm normal distribution of scores. Homoscedasticity was confirmed with scatterplots of residuals and predicted values. PWI-ID was log-transformed due to non-normality.

Primary analyses were conducted using an intention-to-treat approach. In addition, two per-protocol analyses were done. The first one was based on participants who adequately adhered to the intervention protocol by attending at least 12 out of the 16 music sessions. The second one was based on minutes of active music making, which the music worker recorded in the log reports. All

participants who experienced at least 480 minutes of actively making music (16 sessions * 30 minutes) were kept in the analyses.

To explore the second research questions, whether additional music sessions have a bigger effect on the outcome measures than the standard amount of music sessions or CAU, Mann-Whitney tests for between-group comparisons were performed on the change scores, i.e., the difference between the pre-intervention scores and the scores at post-intervention. Effect sizes were calculated, with effect sizes defined as insignificant (<.10), small (.10-.24), medium (.25-.39) or large (>.40) based on guidelines that are commonly used in intellectual disabilities research studies (Dunst & Hamby, 2012). The type of self-reported feelings before and after each music session was compared using a Wilcoxon signed-rank test, given the non-normality of the data. For this purpose, the self-reported feelings were dichotomized into two categories: pleasant feelings (happy or general pleasant) and non-pleasant feelings (sad, scared, angry, tired or general non-pleasant). In addition, to assess whether music can improve already present positive feelings , differences in intensity scores (varying from little, to normal or intense) for participants who scored 'happy' both before and after the music session were examined using a Wilcoxon signed-rank test.

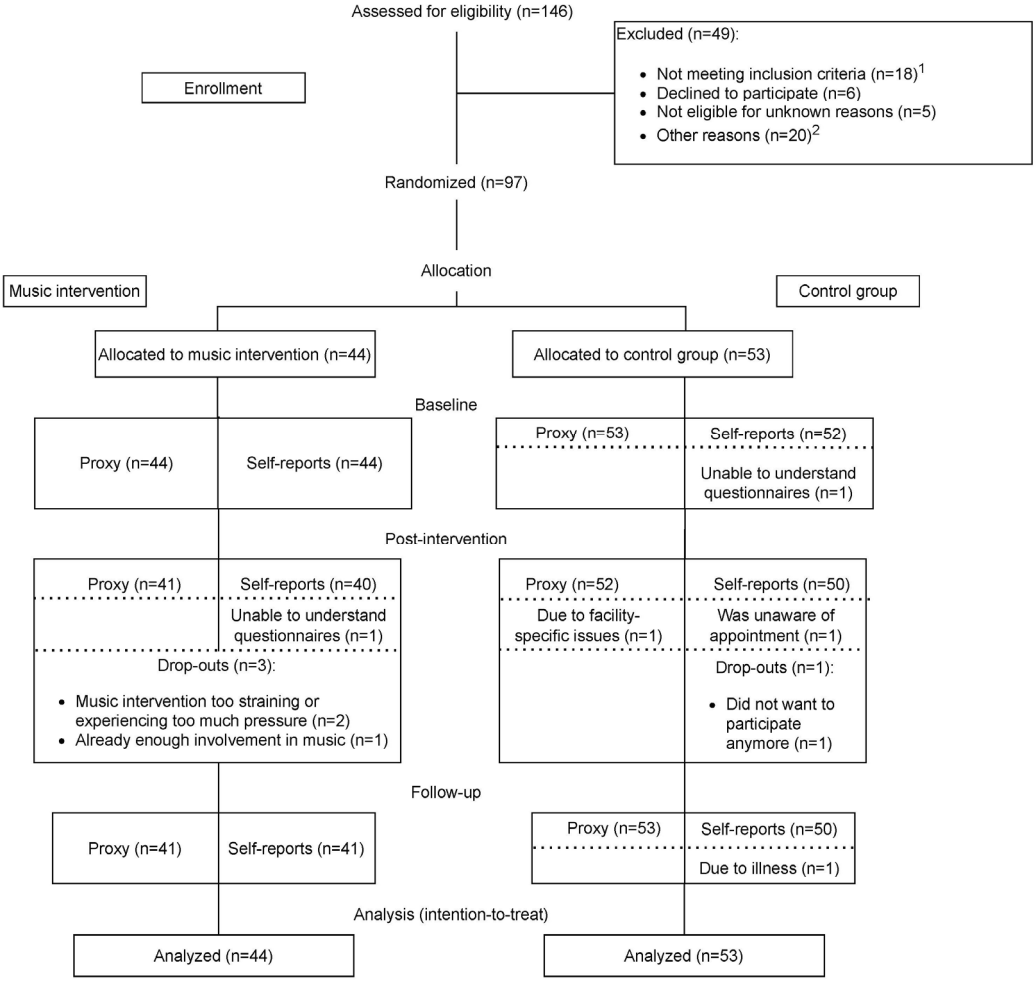
For all analyses, statistical significance was adopted when $p < .05$. Benjamini-Hochberg procedure was applied for false discovery rate control ($q = .20$) to account for multiple comparisons of the primary and secondary analyses (not stated in study protocol) (Thissen et al., 2002). All statistical analyses were conducted using IBM SPSS Statistics for Windows, version 26 (International Business Machines Corp, Armonk, NY, USA).

Results

Recruitment of participants took place between July 2021 and December 2022 and finished when the target sample size was reached. In the present study, 55 facilities were randomized, resulting in 97 participants. Figure 1 presents a flow diagram of the inclusion and exclusion of participants throughout the intervention study, resulting in a cluster-randomization with an unequal number of participants in both conditions. Four participants in the music intervention condition discontinued the intervention, due to various reasons: (1) unable to schedule sessions twice a week/within timeframe (n=2) or (2) not wanting to continue participating in the study (n=2). Of the 55 randomized facilities, in 34 facilities one person participated, in ten facilities two persons participated, in six facilities three persons participated, in two facilities four persons participated, in two facilities five persons participated and in one facility seven persons participated.

Figure 1

Flow-chart following participants through the study



[1] No mild or moderate intellectual disability (n=8), no challenging behaviour (n=8), dementia (n=1), hearing problem (n=1), intervention of one-hour is too long (n=1)
[2] Relocation resident (n=5), too much anticipated stress/arousal (n=3), receiving treatment (n=2), illness resident (n=2), intervention too intense (n=2), legal representative declined (n=1), resident is not stable (n=1), deceased (n=1), too much anticipated work for caregivers (n=1), self-selection of resident after facility is randomized (n=1), presumably unable to understand questionnaires and tests (n=1)

Baseline characteristics and outcome variables

There were no significant differences in the baseline participant characteristics between the intervention and the control condition (Table 1), except for age, which was therefore consequently added as a covariate in all linear mixed models. In Table 2, the mean, standard deviation, and the range of the absolute values of all outcome measures at baseline, post-intervention, and follow-up for both conditions are presented. There was a significant baseline difference between the two conditions on the subscale anxiety.

Table 1

Participant characteristics per group and results of independent samples t-test (age) and chi-square test (sex, level of intellectual disabilities, psychiatric diagnosis, syndrome) for their comparison

	Intervention group	Control group	Statistic (degrees of freedom)	P-value
N	44	53		
Age (years)			2.84 (95)	.006
Mean	51.6	43.0		
SD	14.2	15.4		
Range	24-78	21-73		
Sex (n (%) male)	25 (55%)	29 (55%)	.04 (1)	.84
Level of intellectual disabilities (n (%))			1.76 (1)	.19
Mild	25 (57%)	37 (70%)		
Moderate	19 (43%)	16 (30%)		
Psychiatric diagnosis (n)			3.34 (7)	.91[a]
ASD	11	17		
ADHD	1	4		
MD	4	8		
AD	1	3		
PD	2	4		
SUD	1	2		
Other	13	15		
Syndrome (n)			.11 (2)	1.00
Down's syndrome	3	4		
Other	3	5		

Notes:
ASD = Autism Spectrum Disorder; ADHD = Attention Deficit Hyperactivity Disorder; MD = Mood Disorder; AD = Anxiety Disorder; PD = Personality Disorder; SUD = Substance Use Disorders; Other psychiatric diagnosis = e.g., Schizophrenia, Post-Traumatic Stress Disorder, Attachment Disorder; Other syndrome = e.g., Klinefelter syndrome, Williams-Beuren syndrome, velocardiofacial syndrome
[a]Primary diagnosis

Table 2

Descriptives per outcome measure per group at baseline (T1), post-intervention (T2) and follow-up (T3)

	Intervention group			Control group		
	T1	**T2**	**T3**	**T1**	**T2**	**T3**
Challenging behaviour – Total (0-174)						
Mean (SD)	31.89 (23.82)	25.78 (20.87)	22.98 (21.85)	30.04 (26.52)	30.69 (26.52)	30.83 (30.60)
Minimum -maximum	2.00-86.00	0.00-83.00	0.00-86.00	0.00-108.00	0.00-108.00	0.00-134.00
n	44	41	41	53	52	53
Challenging behaviour - Internalizing behaviour (0-78)						
Mean (SD)	12.55 (11.28)	10.59 (11.00)	9.02 (9.78)	11.94 (12.28)	12.62 (11.57)	12.11 (13.45)
Minimum - maximum	0.00-49.00	0.00-49.00	0.00-44.00	0.00-56.00	0.00-47.00	0.00-52.00
n	44	41	41	53	52	53
Challenging behaviour - Externalizing behaviour (0-84)						
Mean (SD)	16.07 (14.61)	12.54 (11.66)	11.66 (13.20)	15.38 (15.54)	15.29 (15.03)	15.98 (16.52)
Minimum – maximum	0.00-57.00	0.00-53.00	0.00-54.00	0.00-65.00	0.00-64.00	0.00-73.00
n	44	41	41	53	52	53
Self-esteem (5-20)						
Mean (SD)	15.55 (2.82)	16.05 (3.43)	15.80 (3.37)	15.08 (3.90)	15.74 (3.53)	15.74 (3.62)
Minimum – maximum	11.00-20.00	8.00-20.00	8.00-20.00	5.00-20.00	6.00-20.00	6.00-20.00
n	44	40	41	52	50	50
Depression subscale (0-39)						
Mean (SD)	9.82 (6.84)	8.44 (6.25)	6.83 (5.31)	9.33 (8.56)	9.48 (8.06)	9.32 (8.89)
Minimum – maximum	0.00-24.00	0.00-23.00	0.00-21.00	0.00-33.00	0.00-28.00	0.00-33.00
n	44	41	41	52	52	53
Anxiety subscale (0-21)						
Mean (SD)	6.32* (4.12)	5.36 (4.03)	4.37 (3.75)	6.92* (6.15)	6.33 (5.76)	6.36 (6.25)
Minimum – maximum	0.00-16.00	0.00-15.00	0.00-17.00	0.00-20.00	0.00-19.00	0.00-21.00
n	44	41	41	53	52	53

Table 2 *(continued)*

Well-being (0-100)						
Mean (SD)	80.16 (12.83)	77.99 (13.19)	76.68 (12.47)	74.66 (14.13)	73.02 (19.40)	77.03 (18.22)
Minimum – maximum	42.86-92.86	35.71-92.86	50.00-92.86	42.86-92.86	14.29-92.86	21.43-100.00
n	39	37	34	39	36	37

Note: * significant difference between groups at T1 (p<.05)

Primary intention-to-treat analyses

In Table 3 the results of the linear mixed model analyses are presented for each outcome variable with the treatment effect at post-intervention (T2) and at follow-up (T3). Note that the interaction effect is the direct treatment effect in the regression models used (Twisk et al., 2018). For all analyses, except for well-being, cluster was included with a random intercept, since this significantly improved the model. The total CB score in the music intervention condition significantly decreased, and thus improved, more than in the control condition at both post-intervention (difference in improvement of 4.91 on a scale of 0-174) and follow-up (difference in improvement of 7.85 on a scale of 0-174) compared to baseline. Additional analyses showed that both internalizing behaviour and externalizing behaviour significantly improved more in the music intervention condition than in the control condition, again both at post-intervention (difference in improvement of 2.03 on a scale of 0-78 for internalizing behaviour and difference in improvement of 2.75 on a scale of 0-84 for externalizing behaviour) and follow-up (difference in improvement of 3.09 on a scale of 0-78 for internalizing behaviour and difference in improvement of 4.32 on a scale of 0-78). Furthermore, the scores on the subscales depression and anxiety improved significantly more in the music intervention condition than in the control condition at follow-up (difference in improvement of 2.49 on a scale of 0-39 for depression and difference in improvement of 1.99 on a scale of 0-21 for anxiety). To better understand this unexpected finding (i.e., significant improvement at follow-up but not at post-intervention), we performed additional multilevel analyses to check for significant improvements on anxiety or depression at follow-up, compared to post-intervention (instead of baseline). These analyses showed no significant improvements.

All other changes in scores were not significant.

Primary per-protocol analyses

Two per-protocol analyses were performed. First, a per-protocol analysis on the basis of attendance (at least 12 music sessions, n=37) was performed. In Table 3, it is shown that all effects that were significant in the intention-to-treat analyses remained significant, except for internalizing behaviour at post-intervention.

Secondly, a per-protocol analysis on the basis of minutes of active music making (n=33) was performed. All effects that were significant in the intention-to-treat analyses remained significant (after Benjamini-Hochberg correction), except for internalizing behaviour at post-intervention and at follow-up (Table 3).

Table 3

Results of the intention-to-treat and the two per-protocol (based on attendance and minutes of actively making music) linear mixed model analyses, as well as the significant effect modification results by CB and level of intellectual disabilities

	B (95% confidence interval)	p-value
Challenging behaviour - Total		
Post-intervention (T2)		
Intention-to-treat[a,b]	-7.51 (-13.51, -1.50)	**.02**
Per-protocol: attendance	-6.52 (-12.70, -.34)	**.04**
Per-protocol: minutes	-6.04 (-12.46, .38)	.07*
Follow-up (T3)		
Intention-to-treat[a,b]	-9.63 (-15.61, -3.64)	**<.00**
Per-protocol: attendance	-8.84 (-15.00, -2.67)	**.01**
Per-protocol: minutes	-8.33 (-14.73, -1.92)	**.01**
Challenging behaviour - Internalizing Behaviour		
Post-intervention (T2)		
Intention-to-treat[a,b]	-3.14 (-5.98, -.29)	**.03**
Per-protocol: attendance	-2.55 (-5.43, .33)	.08
Per-protocol: minutes	-1.92 (-4.81, .97)	.19
Follow-up (T3)		
Intention-to-treat[a,b]	-3.51 (-6.35, -.68)	**.02**
Per-protocol: attendance	-2.98 (-5.85, -.10)	**.04**
Per-protocol: minutes	-2.38 (-5.27, .50)	.11
Challenging behaviour - Externalizing Behaviour		
Post-intervention (T2)		
Intention-to-treat[a,b]	-3.58 (-6.93, -.23)	**.04**
Per-protocol: attendance	-3.30 (-6.80, .20)	.06*
Per-protocol: minutes	-3.62 (-7.31, .07)	.06*
Follow-up (T3)		
Intention-to-treat[a,b]	-5.09 (-8.4, -1.75)	**<.00**
Per-protocol: attendance	-4.93 (-8.43, -1.44)	**.01**
Per-protocol: minutes	-5.13 (-8.81, -1.45)	**.01**
Self-esteem		
Post-intervention (T2)		
Intention-to-treat[a,b]	-.35 (-1.45, .75)	.53
Per-protocol: attendance	-.42 (-1.56, .72)	.47
Per-protocol: minutes	-.47 (-1.66, .72)	.44
Follow-up (T3)		
Intention-to-treat[a,b]	-.62 (-1.71, .48)	.27
Per-protocol: attendance	-.51 (-1.65, .62)	.37
Per-protocol: minutes	-.32 (-1.50, .86)	.59
Depression Subscale		
Post-intervention (T2)		
Intention-to-treat[a,b]	-1.46 (-3.25, .33)	.11
Per-protocol: attendance	-1.45 (-3.33, .44)	.13

Table 3 *(continued)*

Per-protocol: minutes	-1.30 (-3.26, .66)	.19
Follow-up (T3)		
Intention-to-treat[a,b]	-2.82 (-4.61, -1.04)	**<.00**
Per-protocol: attendance	-2.88 (-4.76, -1.00)	**<.00**
Per-protocol: minutes	-2.90 (-4.85, -.95)	**<.00**
Anxiety subscale		
Post-intervention (T2)		
Intention-to-treat[b]	-.52 (-1.83, .80)	.44
Intention-to-treat (with CB at baseline*treatment)[a1]	-1.17 (-1.48, 1.15)	.80
Per-protocol: attendance	-.35 (-1.71, 1.02)	.62
Per-protocol: minutes	-.32 (-1.76, 1.11)	.66
Follow-up (T3)		
Intention-to-treat[b]	-1.40 (-2.70, -.09)	**.04**
Intention-to-treat (with CB at baseline*treatment)[a1]	-1.05 (-2.36, .26)	.12
Per-protocol: attendance	-1.37 (-2.73, -.01)	**<.05***
Per-protocol: minutes	-1.42 (-2.85, .01)	>.05*
Well-being**		
Post-intervention (T2)		
Intention-to-treat[a]	.07 (-.04, .18)	.18
Intention-to-treat (with level of intellectual disabilities*treatment)[b1]	.03 (-.09, .15)	.65
Per-protocol: attendance	.09 (-.02, .20)	.12
Per-protocol: minutes	.08 (-.03, .20)	.17
Follow-up (T3)		
Intention-to-treat[a]	.00 (-.11, .11)	.94
Intention-to-treat (with level of intellectual disabilities*treatment)[b1]	-.04 (-.17, .08)	.48
Per-protocol: attendance	.01 (-.11, .12	.90
Per-protocol: minutes	-.01 (-.13, .11)	.86

Notes:
The quantified difference in treatment effect (treatment period) after intervention (T2) and after follow-up (T3) between the groups for the subsequent outcome variables, presented as the regression coefficient (B) and the 95% confidence interval and p-value of that difference in line with equation 1, i.e., with T2 and T3 consistent with B4 and B5, respectively.
All reported p-values are before Benjamini-Hochberg correction.
[a] = the treatment effect was not different for participants with different levels of CB at baseline.
[b] = the treatment effect was not different for participants with different levels of intellectual disabilities at baseline.
[a1] = treatment effect was different for participants with different levels of CB due to significant interaction for CB at baseline with treatment (b = -.05 (-.09, -.01), p = .02).
[b1]= treatment effect was different for participants with different levels of intellectual disabilities due to significant interaction for level of intellectual disabilities with treatment (b = .08 (.00, .16), p < .05.).
*Significant after Benjamini-Hochberg correction.
**Log transformed, not adjusted for cluster (i.e., facility).

Effect modification by CB and level of intellectual disabilities

Using linear mixed model analyses, it was explored whether the treatment effect – on all outcomes – was different for participants with different levels of CB at baseline. This interaction between intervention and total CB score at baseline was significant for the ADAMS subscale anxiety (see Table 3). The negative interaction term of CB with intervention indicates that for higher CB values at baseline, the music intervention is associated with a larger difference compared to the control condition, towards a lower – and thus more favourable – level of anxiety. Likewise, analyses were performed to explore whether the treatment effect was different for participants with different levels of intellectual disabilities at baseline. A significant interaction was found for the log-transformed scores on the PWI-ID. Compared to baseline, attending the music intervention by the participants with mild intellectual disabilities was associated with a small but also favourable increase in (the natural log of) PWI-ID at post-intervention, compared to a small decrease in the participants in the control condition. For the participants with moderate intellectual disabilities, this favourable effect was more pronounced and also demonstrated at follow-up.

Effect of additional music sessions

Between participants receiving additional music sessions (varying from 8 to 11 sessions) and those in the original intervention condition as well as the ones in the control condition, no significant differences in the change scores were observed (see Table 4). Effect sizes were either insignificant or small.

Self-reported feelings

Non-parametric Wilcoxon signed-rank tests were performed to assess whether self-reported feelings before and after each music session significantly differed. The values of the self-reported feelings were significantly lower, which reflects more positive feelings after the music sessions (Mdn = 1.00) than before the music sessions (Mdn = 1.25), $T = 33.50$, $p < .001$, $r = -.49$. Even so, positive feelings got stronger, i.e., participants who already indicated that they were happy before the music session (Mdn = 2) felt more happy after the session (Mdn = 2.76), $T = 518.50$, $p < .001$, $r = .49$.

Table 4

Results from the Mann-Whitney U-test on the change scores, i.e., the difference between the pre-intervention scores and the scores at post-intervention, between the small group of participants (n=5) that received between eight to 11 additional music sessions and the remaining participants of the intervention group as well as the control group

	Additional group vs	Effect size (r)	Test statistic (U)	p-value
Challenging behaviour				
Total	Music group	.12	110.00	.45
	Control group	.22	190.00	.09
Internalizing	Music group	.09	104.50	.57
	Control group	.19	180.50	.16
Externalizing	Music group	.11	107.00	.52
	Control group	.19	181.00	.16
Self-esteem	Music group	.10	103.50	.52
	Control group	.02	129.00	.92
Depression subscale	Music group	-.07	79.50	.69
	Control group	-.01	124.50	.93
Anxiety subscale	Music group	.01	92.00	.95
	Control group	.02	135.00	.90
Well-being	Music group	.21	83.00	.24
	Control group	.12	78.00	.51

Discussion

As far as we know, this is the first study of an individual music intervention directed at CB, well-being, anxiety, depression, and self-esteem in adults with intellectual disabilities. In this study, it was demonstrated that a music intervention executed by a music worker as facilitator can be beneficial for adults with intellectual disabilities, as long as the music worker enjoys music, plays a music instrument and is able to relate with adults with intellectual disabilities in a structured way (Savarimuthu & Bunnell, 2002) and in an encouraging environment. Positive intervention effects were seen for CB at post-intervention and follow-up, indicating that an individual active music intervention might be an effective and sustainable way of improving CB in this population. In more detail, this positive effect for CB could be attributed both to internalizing as well as externalizing CB, which were both found to be significantly improved at post-intervention and follow-up. Furthermore, positive intervention effects were found for anxiety and depression at follow-up, indicating that an individual music intervention might be an effective way of improving the mental health in this population. There were no effects found on well-being and self-esteem.

The positive effect on CB found in this music intervention is in line with results found in previous studies, which have shown that music-based interventions can be effective in reducing internalizing CB (Geipel et al., 2018) and externalizing CB in children and adolescents (Ye et al., 2021), and children with mild or moderate intellectual disabilities (Kalgotra & Warwal, 2017). The significant improvements in anxiety and depression at follow-up are, at least partly, in line with two studies showing that music interventions in adults need to last at least 15 weeks to have a significant impact on anxiety and depression (Choi et al., 2008; Guétin et al., 2009), and not just eight weeks (Cooke et al., 2010). Only one music intervention showed a significant improvement after five weeks on depression in adults, but this effect had disappeared three months after intervention (Lu et al., 2013). Although our intervention lasted only eight weeks, we found that the majority of participants in the music group (53%) continued to play music after the last music session, despite our instructions to postpone until after the follow-up measurement. This continued musical engagement could have ensured that the threshold of the minimum number of 15 weeks was reached to have a significant improvement of music on anxiety and depression. Overall, the fact that improvements in anxiety and depression persisted beyond the intervention period and sustained in the long term, strengthen the potential of music interventions for people with intellectual disabilities. Interestingly, the effect of music on anxiety was the highest for participants with more CB at baseline. Perhaps a music intervention is most effective when it comes to anxiety for people with intellectual disabilities with higher levels of CB, in line with research results reporting that music interventions can reduce stress-related symptoms (e.g., anxiety) (De Witte et al., 2020) and that people with intellectual disabilities and CB have elevated stress-related biomarkers compared to people without CB (Symons et al., 2011). However, the relationship and the direction of this relationship between CB and anxiety is quite complex (Koritsas & Iacono, 2015; Pruijssers et al., 2014) and further research is needed to clarify this.

This study was unable to demonstrate a significant effect of music on self-esteem, which is in line with results of another study comparing a rap & sing music therapy among adolescents in a school to a control condition in which no significant difference on self-esteem was found (Uhlig et al., 2018). Studies in which a significant effect of music on self-esteem was found, did not compare the music condition with a control condition (Hillier et al., 2011) or found that in the control condition (reading activity) self-esteem also improved and thus showed no significant difference in effect between the conditions (Cooke et al., 2010). Methodological differences must be taken into account in these cases, and overall, research seems to suggest that a music intervention has no additional beneficial effect on self-esteem. Another possible explanation for not finding a significant effect of music on

self-esteem in our study could be that there was a higher proportion of individuals with moderate intellectual disabilities in the intervention group compared to the control group. This discrepancy in intellectual functioning could have had implications for questionnaire comprehension, since the 4-point Likert scale of this questionnaire might have been too difficult for people with moderate intellectual disabilities (Kooijmans et al., 2022). This might have affected the reliability of responses and the ability to accurately detect intervention effects measured with self-reports, which is also discussed in the next section.

No overall effect on well-being was found, which could be explained by both a ceiling effect at baseline, which limits the potential to observe improvement and reflects a 'subjective well-being homeostasis' (Cummins, 2005) and restriction of range (i.e., limited intra-individual variability) (Coster, 2013). In the manual of the PWI-ID it is stated that a three-point answering option could elicit consistent positive responses to all items on the questionnaire. This consistency could stem from tendencies such as acquiescence or social desirability of responding, leading to a lack of variation in the data (Cummins & Lau, 2005). However, a 3-point Likert scale when measuring well-being is very common in the intellectual disabilities research field (Davison et al., 2022; Schalock, 2010) and is recommended for people with mild to moderate intellectual disabilities (Kooijmans et al., 2022). Despite these psychometric difficulties, it should be noted that a music intervention seems to benefit well-being in participants with moderate intellectual disabilities the most. Overall, self-report ratings seem to be a reliable and valid way to measure subjective well-being (Claes et al., 2012), however, it is important that self-report measures are modified for this target population (Davison et al., 2022; Kooijmans et al., 2022).

Another interesting finding is that no positive effect on all outcome measures was found on increasing the amount of music, which is contrary to results found in previous research (Chung & Woods-Giscombe, 2016). Perhaps the additional benefits of a higher frequency of music sessions are marginal when a certain dosage is reached, which was in the current study two times a week, and the total duration (i.e., number of weeks) might be more important. Further research into the optimal dosage of music sessions is required. Also of interest was the significant increase in positive feelings and the increase in intensity of these positive feelings immediately after each music session. It is known that music can elicit and help regulate already existing positive feelings (Cook et al., 2019; Moore, 2013; Sakamoto et al., 2013; Van Goethem & Sloboda, 2011). This finding provides support for the beneficial effect of conducting music interventions to increase emotional well-being of adults with intellectual disabilities.

The present study has several strengths. In the first place, in this research a randomized design with adequate power was used. Therefore, the positive outcomes of this study were not attributable to passing of time (when no control condition is included) or individual characteristics (when a small sample is used). Another strength is that the intervention was tailored to the cognitive abilities of adults with intellectual disabilities. In addition, the results were obtained in facilities throughout the Netherlands, and therefore, despite the fact that all participants came from the same care organisation, it can be assumed that results found are generalizable to persons with intellectual disabilities living in similar contexts.

Although some of the possible methodological limitations of previous studies were addressed, this study has a number of limitations. First, participants, music workers and research assistants could not be blinded, in line with other RCTs on music interventions (Guétin et al., 2012) and which can lead to biased results. Second, a modified version of the PWI-ID was used to assess well-being, which lacks the sensitivity the full version of the PWI-ID demonstrates. Third, the presence of an occasional significant finding when applying multiple analyses must be considered. However, adjustments for multiple testing were applied. Fourth, the specific benefits of a music intervention were not assessed since no third condition using an active non-musical condition was included.

In conclusion, the results of the present study show that an individual music intervention by a music worker two times a week for eight weeks can have a significant effect on CB, anxiety and depression on adults with mild or moderate intellectual disabilities. In addition, the music sessions helped them to up-regulate positive emotionality. Future research should focus on continuation of improving our understanding of the efficacy of music participation for adults with intellectual disabilities using sound empirical methods including a comparison of effects between music interventions and other non-musical activities. In addition, future research should focus on the optimal duration and number of music sessions required. The issue of pre-existing CB and its moderating effect on anxiety is intriguing and also should be object of future research. From a clinical standpoint, the findings of this study are promising as they encourage the use of music workers as facilitators of active music making as a beneficial activity for people with intellectual disabilities. The lack of effective interventions to improve the mental health of adults with intellectual disabilities (Hastings, 2013) make this recommendation all the more pressing.

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CHAPTER 7

7

Chapter 7

Executive functions in adults with intellectual disabilities: the effects of an individual music intervention and feasibility of testing

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Abstract

Background: People with intellectual disabilities (ID) often demonstrate impairments in executive functioning (EF). In other clinical groups, music interventions have a positive effect on EF. The aim of this study was to examine the effect of an individual active music intervention on EF and to assess the feasibility of EF measurements.

Method: In this cluster randomised controlled trial, a music intervention (16 one-hour sessions in 8-10 weeks, n=44) was compared to care-as-usual (n=53) at baseline, post-intervention, and 8-week follow-up. EF was measured with the flanker test (cognitive flexibility, attention, inhibition), the go/no-go test (inhibition) and the Klingberg test (working memory). Principal component analyses and multilevel models were applied to examine the effect on EF. Feasibility of the EF tests was assessed using a motivation/distraction score and by analysing log reports with EF test deviations from protocol.

Results: The music intervention had no significant effect on EF compared to the care-as-usual condition. Various challenges related to the feasibility of the EF measurements were detected.

Conclusion: The individual music intervention in this study did not enhance EF in adults with mild or moderate ID. Due to several methodological and feasibility issues identified in the study results need to be interpreted with caution.

Introduction

The concept of executive functioning (EF) encompasses a broad cognitive construct related to higher order thinking abilities, which are 'at the heart of all socially useful, personally enhancing, constructive, and creative activities' (Lezak, 1982, p. 281). Executive functions (EFs) may have a distinctive impact on the relation between facing intellectual challenges, effectively managing daily routines and engagement in community activities (Fidler & Lanfranchi, 2022). EF is thought to rely strongly on the prefrontal cortex (Cristofori et al., 2019; Diamond, 2013; Miller & Cohen, 2001), but they are not exclusively dependent on it. More specifically, considerable evidence indicates that these complex cognitive processes are supported by a broader network that includes both anterior and posterior brain regions, as well as cortical and subcortical structures (Bettcher et al., 2016; Heyder et al., 2004; Saban et al., 2018; Whybird et al., 2022). These higher order domains typically consist of several diverse functions, including working memory (i.e., updating, ability to retain and manipulate information over short periods of time), attention (i.e., the ability to retain focus and suppress all other incoming information), cognitive flexibility (i.e., ability to sustain and shift attention, application of different rules) and inhibition (i.e., ability to set priorities and control actions or responses) (Miyake et al., 2000). However, whether EF should be conceptualised addressing these separate yet related cognitive functions (i.e., diversity approach) or as a unitary construct, with one underlying ability, is a matter of an ongoing debate (Karr et al., 2018).

EF, as measured in neuropsychological EF tests, is linked to intellectual functioning (Ardila, 2018). Therefore it is not surprising that people with intellectual disabilities (ID) have been found to perform poorer on separate EF tests than controls matched on mental age (Spaniol & Danielsson, 2021; Zagaria et al., 2021). Existing evidence suggests that this poorer performance holds for diverse EF: working memory (Van Der Molen et al., 2010), cognitive flexibility (Sgaramella et al., 2012), inhibition (Bexkens, Van der Molen, et al., 2014; Bexkens, Ruzzano, et al., 2014; Sgaramella et al., 2012) and attention (Costanzo et al., 2013). When people with ID also show challenging behaviour, they perform even worse on EF tasks than people with ID without challenging behaviour (Schuiringa et al., 2017; Van Rest et al., 2018; Visser et al., 2015). In addition, there seems to be a progressive decrease in performance according to a decrease in ID levels (Memisevic & Sinanovic, 2014; Sgaramella et al., 2012) and it is suggested that the deterioration of EF with aging is more prominent in people with ID than in people without ID (Memisevic & Sinanovic, 2014). Making matters more complicated, measuring EF is challenging, even more so in people with ID. There is a lack of validated, standardised EF tests suitable for use with people with ID (Fidler & Lanfranchi, 2022). Many different tests and

measures are used (Spaniol & Danielsson, 2021) and often in a high proportion of participants floor or ceiling effects at EF (performance) tests are shown (Webb et al., 2020; Willner et al., 2010). Given these complexities, accurately assessing EF in people with ID remains a challenge.

Fortunately, EFs are considered malleable and can be impacted by interventions, regardless of age and cognitive functioning levels (Diamond, 2013; Diamond & Ling, 2019; Lifshitz, 1998; Serpell & Esposito, 2016; Van der Molen et al., 2010). Even more so, people who are most behind on EFs tests can benefit the most from an EF intervention (Diamond, 2013). Musical training could be one such promoting intervention (Diamond & Ling, 2019), since music making can induce brain plasticity in the developing as well as in the adult brain (Altenmuller & Schlaug, 2013). More specifically, music has the power to entrain neural activity in several brain regions, among which the prefrontal cortex (Carlson et al., 2015; Ferreri et al., 2013) and impacts both white and grey matter as well as cortical and subcortical structures (Altenmuller & Schlaug, 2013). Therefore, active music making is a complex activity that draws on several cognitive functions (Bugos, 2019; Haering, 2018) and might stimulate EF (Zuk et al., 2014).

Research addressing the effect of an active music intervention on EF in adults with ID is scarce. In a pilot study on active music making in a group compared with leisure activities no effects were found on EF tests in adults with ID (Smeets et al., 2023). Some research on music interventions in the field of ID focused on attention; however, these studies included children, were non peer-reviewed or concerned case studies (Bentley, 2015; Reza et al., 2011; Sussman, 2009). In multiple behavioural studies it was shown that participation in musical activities stimulates EF in different groups of people, among which people with autism (Haering, 2018), aging adults (Bugos, 2019), people with dementia (Särkämö et al., 2013) and children (Degé & Frischen, 2022; Rodriguez-Gomez & Talero-Gutiérrez, 2022). For example, six weeks of music training twice a week led to enhanced inhibition performance on complex tests in preschool children (Bugos & DeMarie, 2017). Gold et al. (2004) performed a meta-analysis of active music making in children who suffer from either developmental or behavioural disorders and suggested that active music making helps to focus and sustain attention. Selective attention is needed to listen carefully to the sound of another person making music (Degé & Frischen, 2022). Inhibition is required, to stop playing a known rhythm when the beat changes (Degé & Frischen, 2022; Frischen et al., 2021). Cognitive flexibility plays a crucial role during instructions, and remembering musical pieces requires the engagement of both visual and auditory working memory (Frischen et al., 2021). This could lead to a narrow or near transfer effect: music training requires high levels of control, attention and memorisation, and these intervention-related

benefits in EFs could transfer to other contexts where those same EFs are needed (Diamond & Ling, 2016). Therefore, interventions that will most likely improve EF need to tax EFs and be continuously challenging. Besides this point, other suggestions for interventions aimed at improving EF are 1) a long duration, a sufficient length of individual sessions and enough spacing between the sessions, 3) a high level of in-person interaction and a supportive trainer and 4) meaningful activities that provide joy (Diamond & Ling, 2019). An individual active music intervention, aimed at improving EFs and tailored to the specific needs of each participant, seems to have these characteristics.

The present study has two aims. First, to examine the effect of an individual active music intervention on EF in adults with ID compared to adults receiving care-as-usual. Second, to assess the feasibility of EF measurements in adults with ID. To address the first aim we hypothesised that 1) participants receiving an individual music intervention would improve more on EF tests than participants receiving care-as-usual; 2) participants with lower baseline levels of EF would benefit more from the music intervention on EF compared to participants with a higher baseline level of EF; 3) participants with moderate ID would benefit more from the music intervention on EF compared to participants with mild ID; and 4) participants being more frequently engaged in the music intervention would show greater improvements in EF compared to those receiving less frequently a music intervention.

Method

Study design and procedure

The effects of the individual music intervention were tested using a cluster randomised controlled trial (International Clinical Trial Registry Platform NL8482). Adults with mild or moderate ID from residential facilities of the Dutch service organisation Philadelphia Care Foundation in the Netherlands participated between July 2021 and May 2023 in the study. The current findings were part of a larger study, in which also the effects and feasibility of the intervention on CB, mental health, and EF were determined. Full design details have been published in Smeets et al. (2024).

A two-way approach was used for recruitment until the target sample size was reached. Most participants were recruited through the first approach; 1) Managers were informed about the research aims and responsibilities of all parties involved. Subsequently, managers informed caregivers about the study, who then sent the information letter and consent form to potential participants and to their (legal) representatives. 2) Psychologists were informed about the aims of

the study and were asked to refer eligible residents to the caregivers, who then again sent the information letter and consent form to these potential participants and their (legal) representatives. In all cases, eligibility to participate was checked by the psychologist, which ensured that the in- and exclusion criteria were applied appropriately for each potential participant. Randomisation at a facility level by using a list of computer-generated random numbers took place after the participants were screened for eligibility and consented to participate in the trial. Randomisation was concealed, but due to the obvious differences between the two conditions (i.e., music intervention and care-as-usual (CAU)), blinding of participants was not possible. All participants were tested at baseline, post-intervention (after the last music session (intervention group) or 9 weeks after the baseline measurement (CAU group)) and at follow-up (8 weeks after the post-intervention measurement). After the last measurement, participants were rewarded with a small gift. Ethical approval was granted by the review board of the Faculty of Behavioural and Movement Sciences of the Vrije Universiteit Amsterdam (protocol VCWE-2021-081).

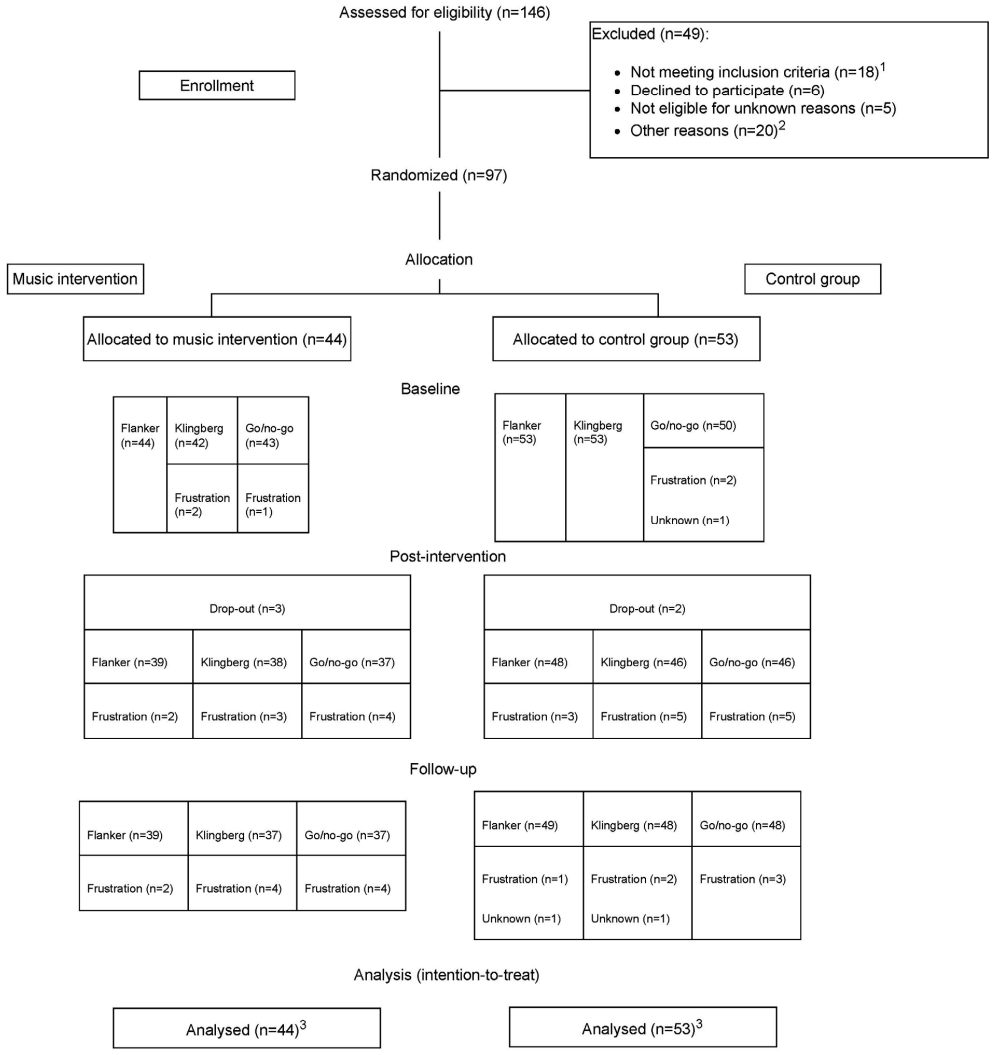
Participants

In Figure 1 the flow diagram with the flow of the participants through the trial is shown. In total, 97 people from 55 facilities, aged between 21 and 78 participated in the study (54 male), of which 44 were randomised to receive the music intervention and 53 to receive CAU (see Table 1). Of the 55 randomised facilities, ten facilities had two participants, six facilities had three participants, two facilities had four participants and two facilities had five participants. In one facility seven participants participated in the study. And in the remaining facilities (n=34) one participant participated.

Inclusion criteria were: mild or moderate ID, ≥18 years, showing CB and being mentally competent to give consent to participate. Exclusion criteria were: (1) not capable of participating in a one-hour long intervention session, (2) hearing problems that could not be diminished via the use of a hearing aid, (3) a medical condition (e.g., dementia) that would majorly influence the participant's ability to engage effectively in the intervention. Informed consent to participate was provided by all participants before data collection started and, if applicable, by their legal representatives (n=52).

Figure 1

Flow-chart following participants through the study



Note: All measurements, valid and invalid, are included

[1] No mild or moderate intellectual disability (n=8), no challenging behaviour (n=8), dementia (n=1), hearing problem (n=1), intervention of one-hour is too long (n=1)

[2] Relocation resident (n=5), too much anticipated stress/arousal (n=3), receiving treatment (n=2), illness resident (n=2), intervention too intense (n=2), legal representative declined (n=1), resident is not stable (n=1), deceased (n=1), too much anticipated work for caregivers (n=1), self-selection of resident after facility is randomized (n=1), presumably unable to understand questionnaires and tests (n=1)

[3] See also Table 2

Table 1

Participant characteristics per group and results of independent samples t-test (age) and chi-square test (sex, level of ID, psychiatric diagnosis, syndrome) for their comparison

	Intervention group	Control group	Statistic (degrees of freedom)	P-value
N	44	53		
Age (years)			2.84 (95)	.006
Mean	51.6	43.0		
SD	14.2	15.4		
Range	24-78	21-73		
Sex (n (%) male)	25 (55%)	29 (55%)	.04 (1)	.84
Level of ID (n (%))			1.76 (1)	.19
Mild	25 (57%)	37 (70%)		
Moderate	19 (43%)	16 (30%)		
Psychiatric diagnosis (n)*			3.34 (7)	.91
ASD	11	17		
ADHD	1	4		
MD	4	8		
AD	1	3		
PD	2	4		
SUD	1	2		
Other	13	15		
Syndrome (n)			.11 (2)	1.00
Down's syndrome	3	4		
Other	3	5		

Notes:
ASD = Autism Spectrum Disorder; ADHD = Attention Deficit Hyperactivity Disorder; MD = Mood Disorder; AD = Anxiety Disorder; PD = Personality Disorder; SUD = Substance Use Disorders; Other psychiatric diagnosis = e.g., Schizophrenia, Post-Traumatic Stress Disorder, Attachment Disorder; Other syndrome = e.g., Klinefelter syndrome, Williams-Beuren syndrome, velocardiofacial syndrome
*Primary diagnosis

Intervention

Music intervention

Prior to the start of the music intervention, every participant received a box with several musical instruments, e.g., a keyboard, guitar, djembe drums, boomwhacker tubes. Before the start of the music sessions, there was an introductory meeting between the participant, their caregiver and their assigned music worker. The music worker could be a trained music therapist, a music therapy student, a musician, a music educator (in Dutch 'muziekagoog') or a caregiver with musical abilities. Every music worker followed a protocol in which the music intervention was outlined in detail, ensuring that the intervention was executed uniformly. However, there was flexibility to adapt the intervention to the specific characteristics and needs of each participant. The protocol addressed the

intervention procedures, such as the setting, general goals, content, and basic principles of the intervention, and included also example exercises. 16 one-hour music sessions were held in 8 to 10 weeks. Each music session had a fixed structure, starting with a welcoming song (2 minutes) followed by a listening exercise (5 minutes), a warming up (5 minutes), actively making music (35 minutes), cooling down (5 minutes) and a closing song (2 minutes). Session one was focused on getting to know one another, sessions 2-6 focused on emotions, in sessions 7 to 12 the focus was on EF and sessions 13 to 15 the focus was on creating their own song as preparation for the 16th session, in which the participant performed the self-created song in front of an audience. The music worker noted the specifics of each session in a log report. The music sessions took place at the apartment of the participant or in another quiet room at the facility.

Care-as-usual

Participants randomised into the control condition received CAU. The CAU condition consisted of the regular supportive care from healthcare professionals and included activities for daily living and social meetings. If participants in the CAU condition were already involved in regular musical activities before the start of this study, these continued, however no additional musical activities were added during the study period. After the study was ended, participants in the control condition were offered a budget to spend on musical activities to their liking, e.g., obtaining an instrument, music lessons or even tickets to attend a concert.

Outcome measures

Outcome measures regarding EF and feasibility.

Executive functioning

At every facility, a research assistant administered the EF tests, consisting of 3 computer-based games on a tablet, specifically designed for children and adolescents with mild ID (Van Rest et al., 2018). To minimise possible observer bias, the research assistants were trained and followed a data collection manual. Furthermore, they were not involved in the programme's execution, which helped prevent any conflict of interest or unintentional influence on the intervention's outcomes. Each EF test lasted around 5 minutes. Four different EF tests were explored: 1) cognitive flexibility, 2) attention, 3) inhibition and 4) working memory. Neither the research assistant nor the software program provided feedback on performance to the participants.

Cognitive flexibility

Cognitive flexibility was assessed using the arrow version of the flanker test (Eriksen & Eriksen, 1974). In general, participants responded to a target arrow, which was flanked by distractor arrows. This test included three blocks: standard flanker, reverse flanker, and a mixed block. Each block began with oral instructions and practice trials. Participants first received the standard flanker block with 5 green arrows lined up, variously pointing left or right. The assignment was to press the arrow key facing the same direction as the middle arrow, while ignoring the outer arrows. In the second block (reverse flanker), participants saw only red arrows and learned a different rule: press the arrow key facing the opposite direction to the middle arrow. In the final, mixed block, participants received stimuli with green (standard flanker) and red (reverse flanker) arrows intermixed. Participants were requested to press the arrow key according to instructions as above, different for green or red arrows. The number of correct responses in the final, mixed, block was used to measure cognitive flexibility (range 0-32), with higher scores indicating better performance. Participants with a high non-response (>3 SD above mean) on the Flanker test were removed, since we made the conservative assumption that this was an indication that they either did not understand the test or did not try to perform the test.

Attention

Focused attention was assessed using the aforementioned standard flanker. Participants had to attend to the task, remember the rule to focus on the middle arrow, and inhibit looking at the distractor arrows. This test yielded two dependent attention variables, correct and incorrect (not late or missing) responses, which were added to create a composite total focused attention score (range 0-32). The incorrect responses were reversed with higher scores indicating better performance.

Inhibition

Inhibition was assessed using the go/no-go test (Trommer et al., 1988). In the first task for processing speed the participants were instructed to press the button (‘go’) when an apple appeared on the screen. The second task included an additional component of inhibition: the participant had to withhold the response (‘no-go’) when a red cross appeared through the apple. The number of incorrect responses in the second task (i.e., pressing the button during a red cross) was used as the dependent variable (range 0-13). In addition, inhibition was measured with the aforementioned reversed red flanker: participants had to suppress their initial response and had to give an opposite reaction. The number of incorrect responses was used as the dependent variable (range 0-32). A

composite total inhibition score was created by the sum of these two tasks, which was also reversed, so that a higher score implicated better inhibition.

Working memory

Working memory was assessed using the Klingberg test (Klingberg et al., 2002; Olesen et al., 2004). Trials of circles were sequentially presented on the screen in a four-by-four grid. Participants had to remember both their location and order and respond by tapping on the screen in the same order (forward version) or in the reverse order (backward version). Each trial consisted of 2 patterns of the same length and difficulty. Trial difficulty was manipulated through increasing the number of circles that had to be remembered and placing circles adjacent or apart, until the participant missed two trials of the same difficulty level in a row. Performance was calculated by the total number of correct trials in the forward and the backward versions. There was no maximum score.

Covariates

Given the development of neurocognitive functioning being age-dependent (Diamond, 2013) and because there was a significant age difference between participants in the two conditions, age was inserted as a covariate in the model. In addition, anxiety and depression scores were also inserted as covariates, given that internalising psychopathology is associated with EF difficulties (Alves et al., 2014; Han et al., 2016; Hollocks et al., 2014). Anxiety and depression scores were administered at baseline using the Anxiety, Depression and Mood Scale (Esbensen et al., 2003; Hermans et al., 2018).

Feasibility

For every EF test, the research assistants noted how often the participant needed to be motivated and how often the participant was visibly distracted during the test. Scores ranged from 1 (‘not’) to 4 (‘constantly’). Data was considered invalid if both scores on an EF test were 3 or if one score was 4, or when the EF test was terminated prematurely due to participant’s frustration.

In addition, a data collection log report was kept, in which all deviations from the protocol for test administration were noted. This log report was analysed to report the remarks about the feasibility of the EF tests.

Data analysis

Sample size calculation requested *n*=96 to detect a significant change between the intervention and control group. The calculation was two-sided, power was set to 95%, the significance level to 5%,

cluster size to two individuals, based on a small effect size of .2, an intra-class correlation of .2 and a drop-out rate of 20%.

Participant characteristics and outcome measures were compared using independent samples t-tests (for continuous variables) or chi-square tests (for categorical data) to identify differences among the two conditions. EF variables were converted into z-scores and, where needed, reversed so that higher scores indicated better performance. Scores of participants that were above or below three SD's from the mean were winsorised to the value of three SDs from the mean.

A principal component analysis (PCA) on all measurements was performed to combine the variables into one variable (Field, 2013). Analysis was based on the 'intention-to-treat' (ITT) principle, with all participants analysed according to randomisation group. Inter-group changes over time (from baseline to post-intervention and from baseline to follow-up) were investigated using linear mixed model analysis with time (represented by two dummy variables) and the interaction between the treatment variable and time. Mixed models are superior for handling missing data, enabling us to include all participants with valid data in our statistical analysis. To adjust for measurements being clustered within individuals, using likelihood ratio test, a random intercept for facility was evaluated as improvement of the model (Twisk, 2013). If positive, the random intercept for facility was added to the model. Interaction terms were used to test whether effects were moderated by baseline level of EF and level of ID. Age, anxiety and depression were inserted as covariates in the model. Assumptions for the linear mixed model analyses were conducted by inspecting the residuals via different graphs (histograms, Q-Q plots and box-and-whisker plots) to confirm normal distribution of scores. Homoscedasticity was confirmed with scatterplots of residuals and predicted values. Additionally, two 'per-protocol' analyses were conducted. In the first one results of participants who adequately adhered to the intervention protocol by attending at least 12 out of the 16 music sessions were analysed. In the second one minutes of active music making were analysed, based on the notes the music worker recorded in the log reports. All participants who experienced at least 480 minutes of active music making (16 sessions * 30 minutes) were included in this per-protocol analysis. These per-protocol analyses provided insight into whether music interventions are more effective in a higher dosage.

Feasibility was assessed using inductive thematic analysis (Braun & Clarke, 2006). Initially, three coders independently analysed a part of the data collection log report using Atlas.ti and compared the results with each other. After a sufficient level of agreement over the codes was established, the

remaining part of the log report was coded independently by two coders. The coders together created a coding framework. In case of disagreement, a fourth researcher (KV) was consulted. Coded data were linked and unified into themes.

By using IBM SPSS Statistics 26, p-values <.05 were considered significant in all analyses.

Results

First, we give general information about the participant flow in the study, reasons for incomplete data or removal of data and descriptive statistics. We then assessed whether the music intervention improved EF more in the intervention group than in the control group. Next, we give the results from the thematic analysis regarding the feasibility of EF measurements.

General information and descriptives

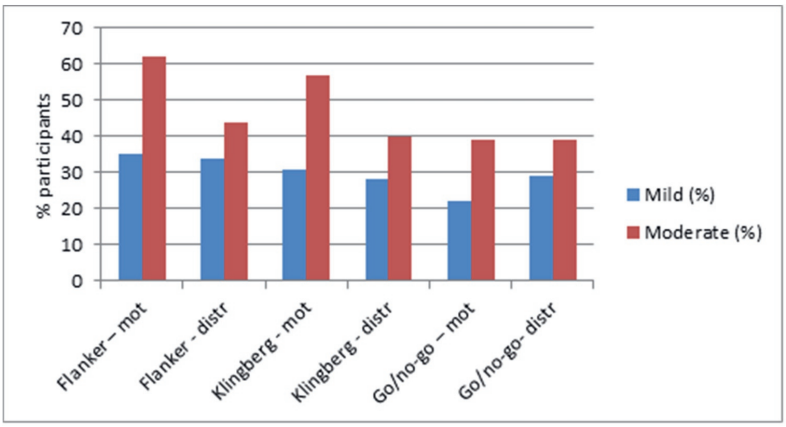
Four participants in the music intervention group discontinued the intervention, because they were unable to schedule sessions twice a week/within timeframe (n=2), or did not want to continue participating in the study (n=2).

Data that were collected, but proved invalid based on the EF test motivation/distraction score were discarded (see Figure 2). Subsequently, for all tasks, distributions of responses were examined to check for ceiling or floor effects and deviations from normality. For the go/no-go test, there was a strong ceiling effect in that 76.5% of the participants made zero mistakes, even after all invalid data were removed (see Figure 3). Consequently, this test was not included in the factor analysis. In the remaining six variables, there were no outliers more than 3 SD away from the mean. Data from two participants on the Flanker test were removed, due to a high non-response. As a result, in the final analyses (see Intervention effects and Table 2) 57 participants were included at T2 and 58 participants at T3.

Table 2 describes the mean, standard deviation, and the range of all remaining outcome measures at baseline, post-intervention and follow-up for both groups.

Figure 2

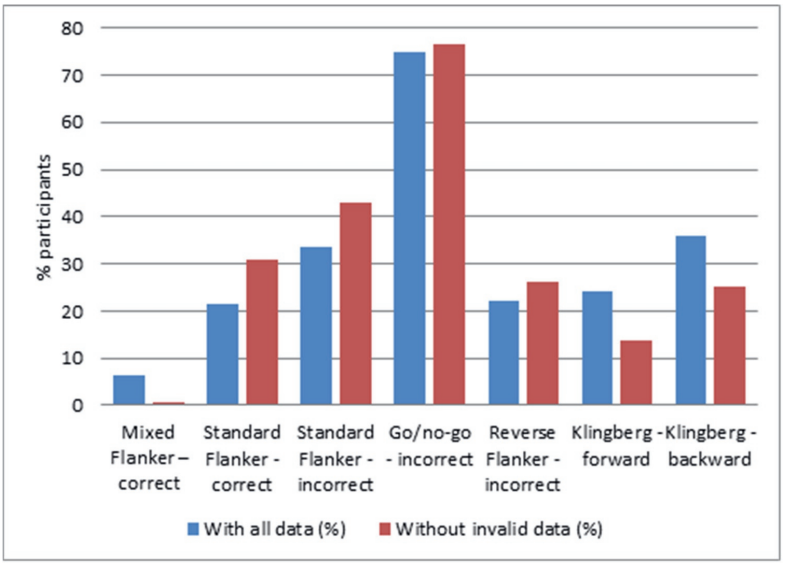
Percentage of invalid EF test scores due to motivation or distraction problems in people with mild and moderate ID



Note: percentage of participants who scored 3 or 4 or terminated the EF test prematurely due to participant's frustration.
mot = motivation, distr = distraction

Figure 3

Floor and ceiling effects of the EF tasks



Note: Percentages are given for either the floor or the ceiling effects of the EF tasks. Mixed Flanker – correct = ceiling effect (maximum score); Standard Flanker – correct = ceiling effect (maximum score); Standard Flanker incorrect = ceiling effect (score 0); Go/no-go – incorrect = ceiling effect (score 0); Reverse Flanker – incorrect = ceiling effect (score 0); Klingberg forward = floor effect (score 0); Klingberg backward = floor effect (score 0).

Table 2

Descriptives per outcome measure per group at baseline (T1), post-intervention (T2) and follow-up (T3)

	Intervention group			Control group		
	T1	**T2**	**T3**	**T1**	**T2**	**T3**
Flanker mixed block, correct responses						
Mean (SD)	18.46 (7.54)	17.57 (5.17)	20.04 (8.01)	20.84 (7.01)	20.62 (8.81)	20.68 (7.48)
Minimum – maximum	7.00-31.00	12.00-30.00	7.00-32.00	8.00-32.00	0.00-32.00	10.00-32.00
n	24	23	23	32	34	34
Standard flanker, correct responses						
Mean (SD)	22.48 (10.18)	23.78 (8.69)	23.13 (9.72)	24.66 (7.82)	25.29 (8.65)	25.09 (7.64)
Minimum – maximum	2.00-32.00	9.00-32.00	3.00-32.00	9.00-32.00	0.00-32.00	6.00-32.00
n	25	23	24	32	34	34
Standard flanker, incorrect responses						
Mean (SD)	3.36 (4.08)	3.57 (5.12)	3.54 (4.78)	3.66 (4.09)	3.74 (5.30)	4.59 (5.08)
Minimum – maximum	0.00-15.00	0.00-19.00	0.00-14.00	0.00-14.00	0.00-16.00	0.00-15.00
n	25	23	24	32	34	34
Reverse flanker, incorrect responses						
Mean (SD)	8.96 (9.62)	9.17 (9.02)	8.96 (9.98)	6.19 (6.99)	7.44 (8.00)	9.35 (8.97)
Minimum – maximum	0.00-31.00	0.00-27.00	0.00-32.00	0.00-23.00	0.00-23.00	0.00-29.00
n	25	23	24	32	34	34
Klingberg, forward						
Mean (SD)	7.76 (4.02)	7.14 (4.16)	7.38 (5.07)	6.73 (4.66)	7.11 (4.68)	7.41 (4.65)
Minimum-Maximum	0.00-13.00	0.00-14.00	0.00-16.00	0.00-14.00	0.00-16.00	0.00-17.00
n	25	21	24	41	37	34
Klingberg, backward						
Mean (SD)	5.36 (3.95)	4.33 (3.26)	3.83 (3.45)	4.41 (3.85)	4.68 (4.16)	4.56 (4.49)
Minimum-Maximum	0.00-12.00	0.00-11.00	0.00-11.00	0.00-11.00	0.00-13.00	0.00-13.00
n	25	21	24	41	37	34

Intervention effects

A PCA with orthogonal rotation (varimax) was undertaken to explore the relationships between the variables. There were no problems with multicollinearity. The factorability of the correlation matrix also met the other assumptions, including a Kaiser–Meyer–Olkin (KMO) measure of .82 and a statistically significant Bartlett's test of sphericity ($p < .001$). All KMO values for individual variables were greater than .79, which is well above the acceptable limit of .5 (Field, 2013). Inspection of the scree plot, as well using the Kaiser criterion (i.e., eigenvalues greater than one) identified one factor, which accounted for 64.47% of the variance. Consequently, in further analyses the single highest-loading variable was used, which was the number of correct responses on the first condition of the Flanker task (i.e., a measure of attention). Table 3 shows the factor loadings.

Table 3

Factor loadings of the principal component analysis

Variable	Factor loading
Standard flanker, number of correct responses	.83
Klingberg, backward	.80
Klingberg, forward	.78
Mixed flanker, number of correct responses	.73
Standard flanker, number of incorrect responses	.72
Reverse flanker, number of incorrect responses	.69

Table 4 presents the intention-to-treat and per-protocol results of the linear mixed model analyses of the number of correct responses on the first condition of the Flanker test, with the treatment effect at post-intervention (T2) and at follow-up (T3) between both conditions. Adding cluster as a random intercept did not significantly improve the model. There were no significant differences between the two conditions and there were no significant differences between the intention-to-treat and the per-protocol analyses. In addition, baseline level of EF or level of ID had no moderating effect (see Table 4).

Table 4

Results of the intention-to-treat and the two per-protocol (based on attendance and minutes of actively making music) linear mixed model analyses, as well as the effect modification results by baseline level of EF and level of ID

	B (95% confidence interval)	p-value
Standard Flanker Task, number of correct responses		
Post-intervention (T2)		
Intention-to-treat	-.18 (-.50, .14)	.27
Per-protocol: attendance	-.23 (-.56, .10)	.17
Per-protocol: minutes	-.20 (-.48, .09)	.18
Follow-up (T3)		
Intention-to-treat	-.06 (-.38, .26)	.72
Per-protocol: attendance	-.03 (-.37, .31)	.85
Per-protocol: minutes	.00 (-.29, .29)	.98
Effectmodification		
EF at baseline*treatment	-.04 (-.20, .12)	.65
Level of ID*treatment	-.01 (-.25, .23)	.91

Feasibility EF measurements

Not all baseline and/or effect measurements were available for each outcome measure. In this paragraph, the most important reasons for incomplete data accountable for both conditions are described (see Table 5). Of all 873 EF tests (i.e., 3 measurements x 3 EF tests x 97 participants), the software malfunctioned 57 times and loss to follow up/discontinuation of the intervention and measurements happened 30 times. Participants showed frustration with the test due to its difficulty and therefore it was decided not to do them again or the participant refused to do the test (again). This occurred 43 times. In addition, there were 3 missings for unknown reasons. Furthermore, 290 times there was missing data because the data proved invalid based on the EF test motivation/distraction score. As can be seen in Figure 2, participants with moderate ID more frequently seemed unable to complete a valid EF test, on all three tests. In addition, a substantial number of participants had scores that showed ceiling or floor effects (see Figure 3).

Table 5

Number of participants with completed datasets and reasons for missing data of all test measurements

	Flanker N (%)	Klingberg N (%)	Go/no-go N (%)
Complete dataset[a]	67 (69.1)	66 (68.0)	65 (67.0)
Reasons for missing data[b, c]			
Software errors	22 (7.6)	20 (6.9)	15 (5.2)
Lost to follow-up/did not want to participate anymore	10 (3.4)	10 (3.4)	10 (3.4)
Tests were too demanding/participant wanted to stop/participant showed frustration[d]	9 (3.1)	16 (5.5)	18 (6.2)
Unknown	1 (0.3)	1 (0.3)	1 (0.3)
Collected, but invalid on basis of EF test motivation/distraction score[e]	111 (37.4)	98 (33.7)	81 (27.3)

[a] T1, T2 and T3 collected, of all 97 participants
[b] T1, T2 and T3 calculated together (3 measurements x 97 participants = 291 tests)
[c] Multiple reasons possible
[d] Assessed or showed during the test administration
[e] Both questions scored 3 or one question scored 4 b or the EF test was terminated prematurely due to participant's frustration

After analysing the data collection log report, several overarching themes regarding the feasibility of the EF tests could be distinguished, which included: 1) Instructional comprehension and test execution challenges, 2) Distraction and concentration, 3) Motor skills, 4) Tablet interaction difficulty and 5) Engagement and motivation struggles.

Theme 1: Instructional comprehension and test execution challenges

Sometimes participants did not comprehend the instructions or showed not to understand the tests. This led to problems with the execution of the tests, which could be subdivided into pushing the button too soon, pushing the button too late, pushing at the wrong spot, or pushing randomly.

> *Flanker explained and when I asked if she understood it she said yes, but when the test started, Ms seemed not to understand it at all. She pushed a few times at random and basically looked at me instead of the tablet the whole time.*

Theme 2: Distraction and concentration

In addition to the EF test motivation/distraction score, it was mentioned several times that participants were distracted in the data collection log report. This could be due to a cat running around or children playing outside. It also occurred that a phone rang during the test administration or a caregiver or resident walked in.

> *But sir got distracted halfway through by his cuckoo clocks, which he wanted to explain briefly.*

Some participants were unable to stay focused during the tests and lost their concentration.

> *Participant seemed to well understand when to push and when not to push, but after a while he seemed to forget a bit what the intention was or he lost concentration which made things a bit worse again.*

Theme 3: Motor skills

Some participants had trouble with their fine motor skills, which affected their ability to perform the tests accurately. In addition, some participants had spasms, showed a trembling hand or held their hand sideways, which resulted in accidental clicks with the rest of the hand.

> *His fine motor skills were very impaired. His finger that he used for clicking moved stiffly/was rigid. The gross movement of bringing the hand to the screen went increasingly smooth, but clicking on the screen precisely was difficult/went often wrong.*

Theme 4: Tablet interaction difficulty

Some participants were not familiar with a tablet, which led to problems with the use of a tablet. Participants pressed too hard or too long, touched the screen with their whole hand or only their nails or dragged with their finger across the screen. In addition, some participants did not keep their hands near the tablet.

> *He also had trouble clicking on the arrows at the bottom of the screen: either he pressed too long or he dragged his finger across the screen, causing him not to click but to drag the tablet's menu upwards.*

Theme 5: Engagement and motivation struggles

Again, in addition to the EF test motivation/distraction score, it was noted that some participants needed continuous encouragement and motivation, which could lead to heightened frustration or terminating the test prematurely. Some participants found the tests boring or childish and sometimes it made them restless and impatient.

During the EF tests, Sir got very frustrated and wanted to put on a song on the tablet. I terminated the EF tests due to sir's frustration.

Discussion

This randomised controlled trial investigated the effects of an individual music intervention on EF in adults with ID. The results showed that 16 music sessions did not significantly improve their EF. Regarding feasibility of the EF tests in people with ID, this study showed that administering the EF tests was challenging for them, which might have influenced the intervention outcomes.

In contrast to our hypothesis and to previous research, we found no effect of an active individual music intervention on EF compared to CAU, regardless of intervention duration (attendance in minutes and number of sessions). In several systematic reviews an improvement in EF was found, especially regarding inhibition, concerning music interventions in preschool children (Degé & Frischen, 2022; Rodriguez-Gomez & Talero-Gutiérrez, 2022) and in people with dementia (Ito et al., 2022). The music interventions administered in these randomised studies lasted from 4 weeks up to 4 years. Even when receiving relatively short interventions an improvement in EF was found in participants, compared to CAU (e.g., Bowmer et al., 2018; Särkämö et al., 2013), which was further evidenced by the lack of a significant effect of music intervention duration on EF found by Fusar-Poli et al. (2017). This negative finding was not due to the intensity of the interventions, as it was higher than the intensity applied in our pilot study (Smeets et al., 2023). Perhaps in people with ID an even longer intervention duration and/or a higher intensity is needed, given the successful (non-musical) intervention observed in other studies (Alloway et al., 2013; Chen et al., 2015; Rodrigues et al., 2019; Wang et al., 2023). These studies employed a racket sports intervention with a longer duration and higher intensity (three times a week for 12 or 16 weeks) (Chen et al., 2015; Wang et al., 2023), a psychomotor intervention once a week for five months (Rodrigues et al., 2019) or a working memory training program four times a week for eight weeks (Alloway et al., 2013). In line with this last study, another explanation for our negative finding might be that only six sessions out of 16 were exclusively focused on EF. Another intervention that focused solely on EF performance, more specifically working memory, found improvements in EF after 20 sessions (Söderqvist et al., 2012). In conclusion, it might be that either a longer intervention with a higher intensity or a greater focus on EF-specific activities within the music sessions may be necessary to achieve meaningful improvements in EF for adults with ID.

Also contrary to our hypothesis, we did not find that participants with lower baseline levels of EF or moderate ID had greater potential for improvement, thus would benefit more from the music intervention, compared to participants with a higher baseline level of EF or mild ID. In our study, examination of these moderator effects was limited due to the relatively small sample size of participants with moderate ID (n=35), which was further diminished by deleted invalid data. Extant research remains conflicted on whether baseline performance is positively or negatively associated with training gains and it seems important to take the design of the training and the EF measures into account (Katz et al., 2021).

Regarding the feasibility of the EF measurements, it was found that the measures used proved to be too difficult for some participants, especially for those with moderate ID. Difficulties were associated with the comprehension of the instructions and the tests, lack of concentration and distraction, motor skills issues, tablet interaction difficulty and engagement and motivation struggles. Furthermore, data from EF tasks often were invalid and high percentages of floor and ceiling effects were found, which is common when studying EF in people with ID (Sgaramella et al., 2012; Willner et al., 2010). However, screening with an EF test motivation/distraction score diminished floor and ceiling effects on almost all tasks. As a consequence of reducing invalid data to improve the overall quality of the data, it also declined the sample size, which compromised the external validity of this study. Due to this removal we exceeded the anticipated drop-out of 20%. In other studies, EF tests were administered to adults with ID, however, their level of ID was higher (Zagaria et al., 2021) and the mean age of participants was lower than in our study (Sgaramella et al., 2012; Willner et al., 2010; Zagaria et al., 2021). In some studies people with IQ < 70 (Meijer et al., 2021) were excluded, however this has ethical implications (Mulhall et al., 2021). Adaptive testing could be a promising approach, allowing customisation of the test experience to match the participants' cognitive level or developmental age. Furthermore, given that there are numerous other EF tests available for people with ID (Spaniol & Danielsson, 2021), it is recommended for further EF-research, to use other paper-pencil or digital EF performance measures that are found suitable for people with moderate ID. These measures include the Behavioral Assessment of Dysexecutive Syndrome battery (Zagaria et al., 2021), Cambridge Executive Functioning Assessment (Willner et al., 2010) or Frontal Assessment Battery (Sgaramella et al., 2012). We chose for three EF performance tests on a tablet, given the administration ease and the earlier positive experiences with these tests with children with mild ID (Van Rest et al., 2018). However, in the PCA we found that all our EF variables could be combined into one 'focused attention' variable. This implies that a unitary construct for EF best fitted our data, which was also found in preschool children (Wiebe et al., 2008). On the other hand, our three EF

tests were possibly too homogeneous to gain a more comprehensive and diverse picture of EF in people with ID. To gain a more comprehensive picture, a multi-informant approach is recommended (Fidler & Lanfranchi, 2022) by use of a proxy-report such as the Behaviour Rating Inventory of Executive Function (BRIEF) (Memisevic & Sinanovic, 2014). We did not administer the BRIEF to reduce the caregiver burden, which was an issue in an earlier pilot study (Swart et al., 2024). However, using proxy measures like the BRIEF can complement EF performance measures by capturing EF-related behaviours in daily life contexts. This approach recognizes the challenges associated with directly assessing EF tests in individuals with (moderate) ID, which might confound performances on EF assessments, and acknowledges the importance of considering their functional abilities and behaviours beyond standardised testing environments (Barton & McIntyre, 2022).

Besides the abovementioned explanations, we should consider that several methodological issues could have hampered the possibility of finding positive intervention effects in our analyses. People with moderate ID often have (major) problems with processing speed (Santegoeds et al., 2022).This could have delayed the processing of the EF tasks, more so than in other target groups assessed by these tests (such as youth with mild ID and borderline IQ, see Van Rest et al. (2018). We did not correct for this delayed processing speed by including a covariate in data analysis, nor during test administration by adjusting the test to a self-spaced format, resulting in a more valid measure specifically for testing in moderate ID. To address these issues in future EF research, it is recommended to allow participants with moderate ID more time to respond, to provide additional learning opportunities, and to offer coaching support if needed (Santegoeds et al., 2022; Schuiringa et al., 2017). Another possible methodological limitation to be considered is that, despite our endeavours to exclude people with dementia, in the log reports from the music sessions we read that some participants had suspected dementia. Related to this, participants in our sample had a relatively older age, especially in the music intervention condition, whereas most of the literature on EF in people with ID included rather young people (Spaniol & Danielsson, 2021). This could have influenced the results, given that the used tests had a strong visuospatial nature. Impairments in visuospatial information processing occur early in the course of Alzheimer-type dementia (Quental et al., 2013) and we know that the deterioration of EF with aging is more pronounced in people with ID compared to those without (Memisevic & Sinanovic, 2014). In addition, older adults face impairments in their learning rate (Nassar et al., 2016) and older people with ID often show a lack of engagement with technology and experience, for example, difficulties with the use of a tablet (McCausland et al., 2017). The last methodological limitation to address is that we did not explore the moderating effect of ID aetiology. People with different syndromes perform differently on EF

tests (Spaniol & Danielsson, 2021), although findings have not always been consistent (Sgaramella et al., 2012). The heterogeneity of the ID population, which encompasses a wide range of syndromes and genetic conditions, can mask potential differences in EF performance among subgroups.

In conclusion, no significant effect of an individual music intervention was found on EF for people with ID. Furthermore, identifying and acknowledging the difficulties encountered during the administration of EF tests is crucial to ensure that EF testing is accurately carried out. The results of this study contribute to our knowledge of EF and of music interventions in people with ID.

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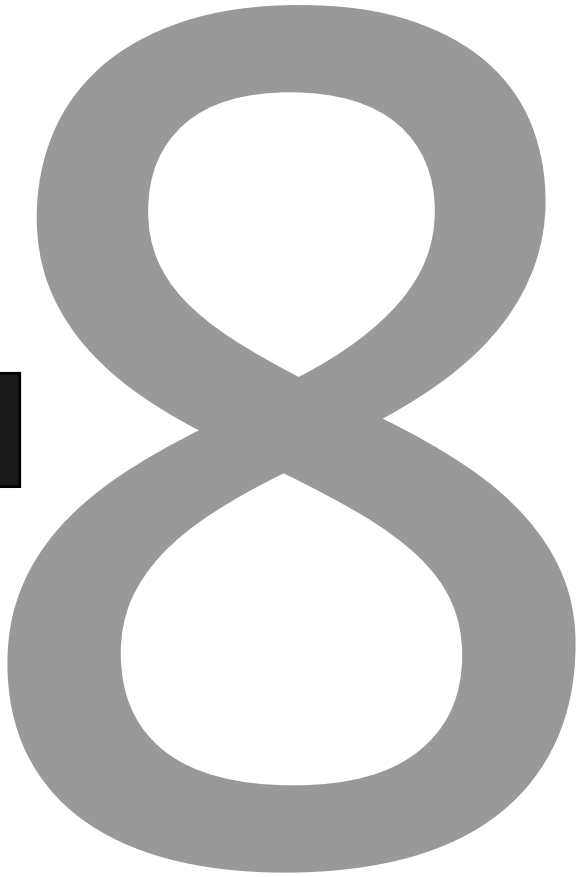
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CHAPTER 8



Chapter 8

Process evaluation of an individual music intervention for adults with intellectual disabilities in the context of a randomised controlled trial

Smeets, G. J. J., Volkers, K. M., Scherder, E. J. A., Moonen, X. M. H. (2024). *Process evaluation of an individual music intervention for adults with intellectual disabilities in the context of a randomised controlled trial* [Manuscript submitted for publication].

Abstract

Introduction: Outcomes of randomised controlled trials (RCTs) for people with intellectual disabilities (ID) are important to report, but reporting the practical experiences of conducting RCTs is also important. This study presents a process evaluation of a cluster-RCT on an active music intervention for adults with mild to moderate ID and challenging behaviour.

Method: This evaluation focused on the process of recruitment and participation in the RCT, the implementation of the intervention, the experiences of clients and other disciplines participating in the music intervention and the presumed sustainability of the intervention. Participants, caregivers and music workers were involved in this process evaluation. Various measures were used: evaluation forms, follow-up questionnaires, semi-structured interviews, log reports and RCT recruitment and data collection. A qualitative content analysis was conducted.

Results: Recruitment of enough participants for the RCT was challenging, but during the RCT the level of adherence and participants' retention rates were high. The intervention was implemented successfully in the RCT since the intervention was tailored to the participants' individual needs, resulting in strong fidelity. The intervention was well accepted, feasible, and applicable, with promising indications for sustainability, however, caregiver workload and little perceived support were presented as challenges for usability of the intervention after the RCT.

Discussion: This study highlights that an individual music intervention for adults with ID can be feasible and sustainable, though factors such as caregiver workload and support are critical for long-term success. Further research is needed to address these barriers in order to optimize intervention sustainability.

Introduction

Conducting clinical trials in the field of adults with intellectual disabilities (ID) and challenging behaviour (CB) is known to be challenging, for example because of informed consent issues, recruitment problems and staff turnover (Cleaver & Sakar, 2010; Mulhall et al., 2018). For this reason, in many trials people with ID are excluded (Feldman et al., 2014; Meijer et al., 2021) and the evidence base of interventions regarding people with ID significantly lags behind mental health interventions in general (Hastings, 2013). It is therefore important to strengthen this evidence base in the field of ID not only by incorporating the outcomes of the randomised controlled trials (RCTs), but also by sharing practical experiences. This 'experience base' is a valuable addition to scientific knowledge and can ultimately help reduce the health inequities faced by people with ID, which often arise due to inadequate or absent provision of services and care (Mulhall et al., 2018). Practical experiences of clinical trials can be reported in process evaluations (Moore et al., 2015).

Process evaluations are used to monitor and document intervention implementation or implementation failure, and can aid in understanding the relationship between the intervention characteristics and factors such as fidelity and sustainability of the intervention (Moore et al., 2015). For a process evaluation, a full description of the intervention is necessary. Unfortunately, most reports on RCTs dealing with social and behavioural interventions do not provide insight into the intervention manual (Grant et al., 2013). Due to this lack, it is difficult to generate consistent outcomes for participants, identify active ingredients of the intervention and successfully expand, replicate or adapt the intervention to new populations or settings (Maxwell, 2019). To better understand the fidelity and the quality of the intervention implementation, mechanisms of impact, and contextual factors which shape the outcomes, it is important to fully describe interventions and perform process evaluations, especially for complex interventions, which are characterized by several interacting components (Moore et al., 2015).

An active music intervention can be categorized as a complex intervention, particularly when applied to people with ID, due to its inherent complexities in intervention design, as described above, and study implementation (Randell et al., 2022). Although process evaluations are essential for understanding how, especially complex, interventions work, no studies have yet conducted a comprehensive process evaluation of music interventions for adults with ID. This gap is not surprising, given the overall scarcity of research on music interventions in a controlled experimental setting, preferably an RCT, in this population. However, existing evidence supports the positive effect

of music interventions on mental health, CB and well-being in people with ID. This evidence in people with ID can be found in two scoping reviews, of which one included 12 reviews and 62 empirical studies (Després et al., 2024) and the other covered 44 studies including 3 RCTs (Mino-Roy et al., 2021), an uncontrolled study (Kalgotra & Warwal, 2017) and a cross-sectional study (Jaschke & Scherder, 2015). In children and other clinical populations, such as people with autism spectrum disorder and people with dementia, stronger evidence can be found that music making can have a significant impact on a broad range of mental health, behaviour and well-being domains (Degé & Frischen, 2022; Geretsegger et al., 2022; Pedersen et al., 2017; Van der Steen et al., 2018). In sum, while existing evidence highlights the potential benefits of music interventions for individuals with ID, there is a need for more controlled studies and comprehensive process evaluations.

The aim of this paper is to describe the results of a process evaluation of a cluster-RCT design concerning an active music intervention that was developed for adults with mild to moderate ID and CB. Research questions were:

1. How was the recruitment and participation (of clients and caregivers) in the RCT?
2. To what extent was the intervention implemented as planned and performed in compliance with the programme guidelines that were developed in the RCT?
3. What were the experiences of multiple stakeholders with the intervention?
4. To what extent was music making sustained after the RCT ended?

Method

To answer each research question, the associated process evaluation components had to be specified. These components are based on the Medical Research Council (MRC) Guidance (Moore et al., 2015), the framework by Steckler and Linnan (2002) and the RE-AIM Framework (Glasgow et al., 1999). These components were all recently incorporated in research regarding people with ID (Bosco et al., 2019; Lai et al., 2019; Matthews et al., 2016), since there is no specific or adapted process evaluation framework that can be used for research with people with ID (Randell et al., 2022). Participation in the RCT is defined as the level of enrolment and engagement of participants in the RCT (recruitment, reach and RCT data collection). The implementation of the intervention relates to the delivery of the intervention—whether it was delivered as designed (dose, fidelity, adaptation). Experiences with the intervention help to understand the qualitative experiences of participants, caregivers as well as music workers (acceptability). Sustainability implies the extent to which the intervention was maintained over time, specifically looking at whether participants continued making

music after the intervention ended (maintenance). More information about these components can be found in Table 1.

Table 1

Research aims, components, questions and data sources from the process evaluation

Research aim	Process evaluation component	Question	Data source
1. Recruitment and participation in the RCT	Recruitment	Which procedures and strategies were used to approach stakeholders and identify people with ID to enrol in the study? What were reasons for not participating?	Semi-structured Interviews RCT recruitment
	Reach	Who actually participated in the RCT? To what extent were they representative of the intended target audience?	Evaluation forms music workers Semi-structured Interviews Demographic information
	RCT data collection	What are the response rates of primary outcome data? Were tests and questionnaires administered according to protocol?	RCT data collection Dates of administration of all RCT outcome measures Assessor of proxy questionnaires RCT
2. Intervention implementation	Dose	How much intervention (time-wise) was delivered? What reasons are associated with this dose?	Evaluation forms music workers and participants Semi-structured interviews Log reports music sessions RCT Data collection
	Fidelity	To what extent was the music intervention implemented as planned during the intervention period and performed in compliance with the training manual?	Log reports music sessions RCT data collection
	Adaptation	What variations in delivery of the intervention were made deliberately by the music worker?	Evaluation forms music worker Log reports music sessions RCT data collection
3. Experiences with the intervention	Acceptability	To what extent was the intervention feasible and applicable according to the participants, caregivers and music workers?	Evaluation forms music workers and participants Semi-structured interviews Log reports music sessions RCT data collection
4. Sustainability	Maintenance	Was music making sustained over time and what factors are associated with it?	Follow-up questionnaires participants and caregivers Semi-structured interviews Log reports music sessions

Music intervention – cluster-RCT

In the cluster-RCT, participants with mild or moderate ID and CB, whose age ranged between 21 and 78, were randomised into an individual music intervention group (n=44) that consisted of 16 active music sessions over a maximum of 10 weeks or a care-as-usual (CAU) group (n=53). Participants in the CAU group received a musical budget after follow-up. The music intervention was guided by music workers who worked with a training manual for this purpose. RCT measurements were carried out at baseline, during intervention, post-intervention, and at eight-week follow-up and included self-reports/tests as well as proxy measurements. Figure 1 shows the process for inclusion and cluster-RCT design. The protocol and main findings from this RCT have been published or submitted elsewhere (Smeets et al., 2024; Smeets, Van Rest, et al., 2024; Smeets, Volkers, et al., 2024) and the appendix contains a description of the music intervention.

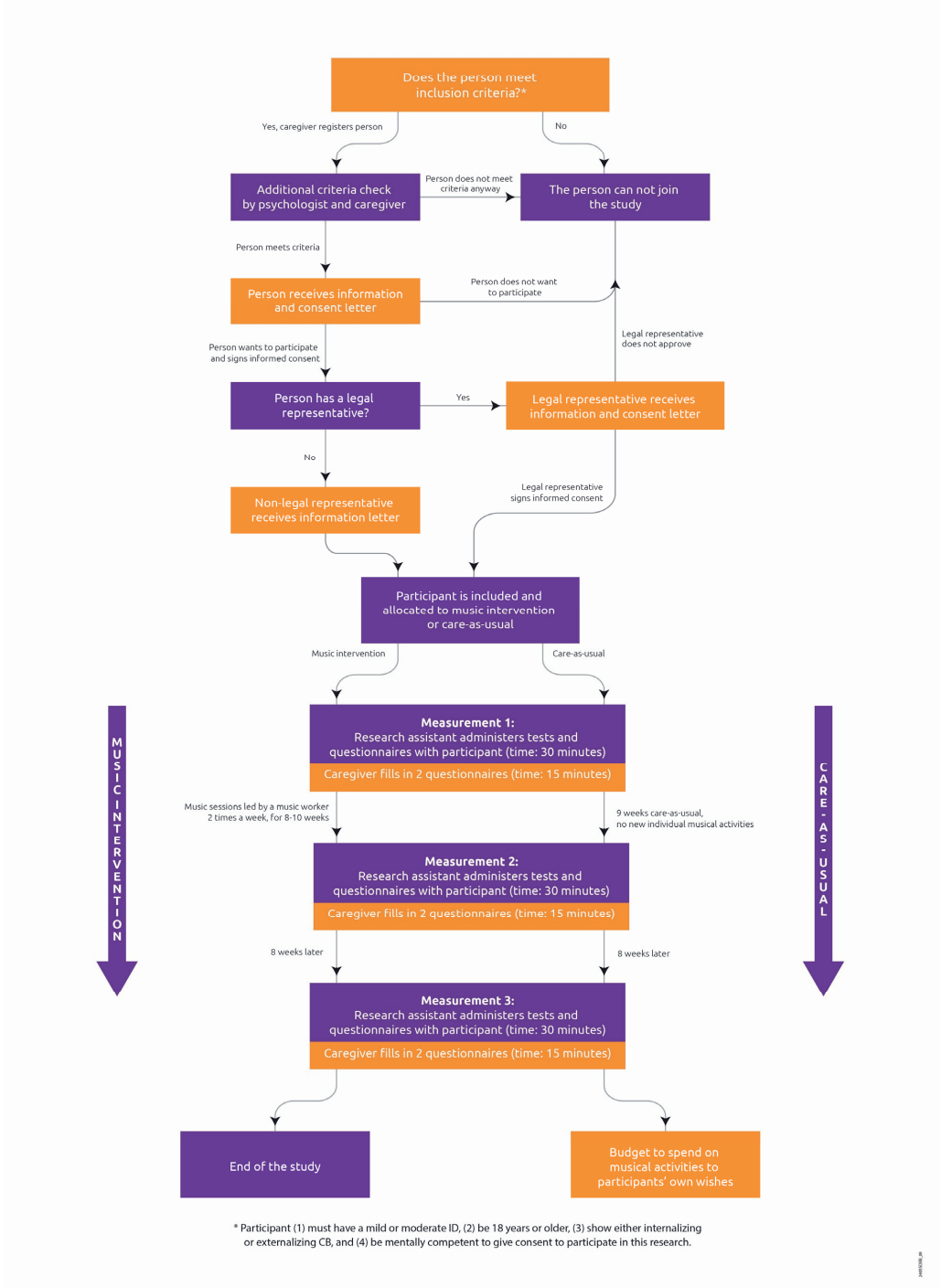
Table 2

Participant characteristics per group

		Intervention group	Care-as-usual group
N		44	53
Age (mean years (SD))		51.6 (14.2)	43.0 (15.4)
Sex (n (%) male)		25 (55%)	29 (55%)
Level of ID (n (%))			
	Mild	25 (57%)	37 (70%)
	Moderate	19 (43%)	16 (30%)

Figure 1

Process for inclusion and RCT design



* Participant (1) must have a mild or moderate ID, (2) be 18 years or older, (3) show either internalizing or externalizing CB, and (4) be mentally competent to give consent to participate in this research.

Participants process evaluation

Besides analysing quantitative data in the RCT evaluation, in this process evaluation qualitative data were analysed from three perspectives. First, information was analysed from the 97 participants with ID (see Table 2). Second, information from the 27 music workers who led the music sessions was analysed. These 15 women and 12 men had different backgrounds, such as music educators ('muziekagogen' in Dutch), music therapists, graduates from music academies, and health care providers with a musical affinity from inside and outside the Philadelphia Care Foundation. Finally, information from the caregivers of the participants was analysed. Caregivers filled in the evaluation questionnaires and follow-up questionnaires and 13 caregivers also participated in semi-structured interviews. These interviewees were 85% female, on average 45 years old (SD=11.8), had on average 13.5 years (SD=12.3) of working experience in ID care, of which on average 5.4 years (SD=4.1) were with the participant they reported on.

Procedure

Ethical approval for this research was given by the review board of the Faculty of Behavioural and Movement Sciences of the Vrije Universiteit Amsterdam (protocol #VCWE-2021-081), prior to commencing the research project. We obtained informed consent from all participants with ID and where applicable from their legal representatives (n=52), and from all music workers who completed the evaluation questionnaires, and from all caregivers who participated in the semi-structured interviews.

Measurements process evaluation

The following process outcome measures were collected: (1) evaluation forms from participants and music workers; (2) follow-up questionnaires from participants and caregivers; (3) semi-structured interviews with caregivers; (4) log reports from music workers for each music session and (5) RCT recruitment and data collection.

Evaluation forms

After the last music session, an evaluation form was completed by participants, to gain their personal views about the music sessions and their (dis)satisfaction with specific elements of it. The music workers completed an evaluation form about their perceived effects regarding behavioural, cognitive and emotional changes in the participant, impact of COVID-19 measures and their most beautiful moment in the intervention.

Follow-up questionnaires

Whether participants continued to participate in musical activities since the ending of the intervention was assessed 2 times. First, participants themselves reported this at follow-up, second this was reported six months after the ending of the intervention by the caregivers of the participants of the music group.

Semi-structured interviews with caregivers

All caregivers working with clients participating in the music intervention (and who did not drop out the RCT), were asked to participate in semi-structured interviews. 13 caregivers agreed to participate. The questions used were the result of a triangulation of available literature with the aims of this study. The purpose of the semi-structured interview was twofold: 1) to find out what perceived outcomes caregivers observed in participants and 2) to assess their experiences with the intervention and the conditions that were required by the RCT design. The semi-structured interviews were conducted online using MS Teams by a research assistant or the primary researcher (GS).

Log reports

Music workers completed a log report after each music session. This log report contained questions about every session: whether the session was performed according to the manual, how much time was spent on active music making and there was room for additional remarks.

RCT recruitment and data collection

Information about the recruitment process was analysed, for example how participants were identified, approached, and enrolled. During the RCT data collection all deviations from the protocol when administering tests and questionnaires were noted and subsequently analysed, for example missing tests and specific challenges and adaptations for participants.

Data analysis

With the respondent's permission, the interviews were recorded and transcribed verbatim. All respondents read the transcribed interview to check whether they agreed with its wording as a way of member validation (Green & Thorogood, 2018). Afterwards, all identifying information was removed and data were anonymized before they were analysed in Atlas.ti 23. Analysis of the data was mostly deductive using codes that corresponded with the components of the process evaluation.

Subsequently, if relevant information fell outside the components, an inductive approach was used to develop additional codes.

To guarantee a robust analysis framework, a preliminary codebook was developed. Next, data of two documents from each data source was coded separately by two research assistants and one researcher (GS). The coded data were compared, discussed and a fourth researcher (KV) was consulted when needed. This procedure led to a second version of the codebook. Next, the two research assistants separately coded five additional documents from each data source. Their codes were compared and discussed with GS, which resulted in a third version of the codebook. Finally, the two research assistants separately coded all documents and subsequently analysed, compared and discussed their codes. This thorough and iterative process ensured consistency in the coding, leading to a robust analysis framework.

Results

The results are narratively reported by every of the four research questions and their components. Quotations are chosen to illustrate these components.

Recruitment and participation in the RCT

Recruitment

Participants were recruited indirectly through staff members of a large Dutch organisation in the care of people with ID, Philadelphia Care Foundation. Initially, the researchers reached out through e-mail to managers of different residential facilities and sent them information about the study. Additional recruitment strategies, including newsletters, internal news messages and a recruitment video, were used to inform staff about the study, but all initiatives were insufficient in generating sufficient participant enrolment within the original timeframe. As a response to this low enrolment, other stakeholders were asked to assist with the recruitment, such as music coaches from Philadelphia Care Foundation, caregivers and psychologists. Increased emphasis on the importance of sufficient participation from higher-level management (the board of directors) also encouraged caregivers and psychologists to prioritize recruitment. Caregivers recruited most participants, followed by psychologists, while other staff members played a smaller role in the recruitment process.

We are unable to give an absolute number of people with ID who were informed about the study, since different professionals (i.e., caregivers, psychologists) were approached and served as gatekeepers. Gatekeepers reported a lack of interest from potential participants on 15 occasions,

cited unmet inclusion criteria for one or more participants 44 times, and considered the intervention too intense in four instances. Also, 15 people with ID were not found eligible after the psychologist did an additional check applying the inclusion criteria. In addition, there were facility-related reasons for not participating (e.g., a lot of new residents, staff shortage or busy and chaotic times) (n=10). And in some cases, facilities were simply not interested in participating in the study. In facilities with eligible participants, caregivers did not always refer additional potential participants for the study, despite the likelihood of other suitable residents. Some caregivers were unaware they could refer multiple residents from the same facility, while others selectively referred those they deemed most appropriate.

> *Yes, for me they were the most obvious. It wouldn't take away that we would have other clients as well where this could definitely apply (caregiver).*

Reach

For reach we looked at the demographic characteristics of the participants to understand the diversity and representativeness of participants. The participants included in the study were living across the Netherlands and all received professional care from the same organisation. All participants were interested in music and 19% of them had a psychiatric diagnosis (e.g., autism spectrum disorder, mood disorder). Despite inclusion criteria and the additional check by the psychologist, there were some participants included with limited CB or with dementia.

> *We also looked at which clients already have something to do with music. And that was PPN20 and PPN22. Plus the bit of attention they experience as very nice and which makes them feel very special (caregiver).*

RCT data collection

Most participants who chose to discontinue their participation in the RCT, still took part in the study's primary outcome measurements. This resulted in a high response rate of at least 91% for both self-reports and proxy measures across both groups (music group and CAU group). Additionally, some participants selectively opted out of providing data at specific measurement points without formally withdrawing their consent to participate in the study. As a result, while the overall sample size was maintained, these instances introduced some missing data within the dataset.

Of all proxy questionnaires completed, 71% were filled in by the same caregiver at all three measurement times as was requested. Of all other cases, 85% were filled in by the same caregiver at

two of the measurement times. Reasons for not adhering to the request of having the same caregiver complete all questionnaires were the inability to reach a specific caregiver or the reassignment of a caregiver to a different position.

Of all measurements completed, not all data were received 'on time'. For the music group intervention, only one week after the last music session the post-intervention measurement was scored as on time, while this was two weeks (8-10 weeks after baseline measurement) for the CAU group. In addition, the appointment for post-intervention measurement with the participant was made after the last music session, while this was already planned at baseline in the control group. This resulted in a higher on time post-intervention rate for self-reports in the control group (92% self-report, 50% proxy) compared to the intervention group (66% self-report, 53% proxy). For the follow-up measurement, both groups already made their appointment at post-intervention within a two-week time frame (7-9 weeks after post-intervention measurement), which resulted in a 94% self-reports and 77% proxy reports score on time.

Intervention implementation

Dose

The total duration of per session active music making was 39 minutes on average, which was quite close to the targeted duration of 40 minutes in the manual (or 45 minutes, depending on the type of cooling down activity). This average time of active music making was also influenced by the fact that some participants received more than the protocolized intervention dose. Per participant, the mean time of active music making per session ranged from 14 to 55 minutes. In the log reports music workers noted that sometimes it was necessary to perform other non-musical activities during a session, for example clean the desk of the participant because this was too messy, or first to celebrate the birthday of another resident. In addition, sometimes participants got distracted by the presence of other people (e.g., spouse, daughter). Sometimes participants said to the music worker that they preferred to talk a bit longer or they only wanted to listen that session and not play music. Some sessions were terminated earlier, for example because the participant was too tired, whereas some sessions continued longer than one hour.

> *Because PPN35 was doing so well, I went on a bit longer so he had a bit more distraction. (music worker)*

Fidelity

Of all 44 participants in the music intervention, seven stopped the intervention after 0-8 sessions. Reasons mentioned were: sessions were too much after all (n=3), unable to schedule sessions twice a week/within the timeframe (n=2), pregnancy (n=1) or not wanting to continue (n=1). 36 Participants (81%) received all 16 music sessions of which 94% managed to do so within the maximum time frame of 10 weeks. Reasons for not receiving all sessions were that some sessions were skipped and apparently not caught up, because the participant did not show up or was ill. Sometimes there was a larger than planned timespan between two sessions, due to illness, changes in day care or quarantine related to COVID-19. In 60% of all music sessions, the fixed order of activities was adhered to according to the training manual.

Adaptation

Music workers deliberately adapted some intervention sessions deviating from the training manual. A few times, major adaptations were made; two sessions were combined to 'one' session of two hours, combining it with the sessions of two other participants. In one instance a session was changed into a completely other activity, namely a dinner. Furthermore, music workers often tailored sessions with minor adaptations to meet participants' preferences, moods, needs or interests (e.g., with dancing), to better connect with the participant. Music workers stated that sometimes participants mainly wanted to talk and it was needed to reserve time for this need, in order to move on with the session. Music workers tried to adjust to the participants' musical skills, to their emotions or their attention span by shortening or constantly changing the musical exercises. Activities like the welcoming song or listening exercise were often skipped due to time constraints or because they felt it was unsuitable or childish. Overall, adaptations were made mainly for the younger half of participants (below the median age of 50), and for participants with mild ID.

Experiences with the intervention

Acceptability of participants

Most participants (85%) found the music sessions very enjoyable, 12% found them enjoyable and 2% did not enjoy them. What most participants liked best was playing the music instruments, making music (together) and singing. They also enjoyed making their own music or song and performed it when the intervention ended. Most participants did not want the music sessions to end. When asked if the participant would recommend the music sessions to another resident from the facility, 73% stated they would recommend the music sessions, 10% would not recommend the music sessions,

for example because they found the music sessions not matching their needs and 17% were unsure whether they would recommend the music sessions.

When asked what they wanted to change, 66% wanted more possibilities to make music per week versus 7% who wanted less music per week, 7% wanted the music sessions at a different day or time and 7% would want to be able to choose a different music worker.

Acceptability of caregivers

All caregivers stated that they would recommend the music sessions to their colleagues. They observed that participants enjoyed the music sessions and were happy and enthusiastic about it. Caregivers evidenced this joy in the participants' laughter, in the things participants said about the music sessions and in their behaviour (e.g., everything went more 'smooth'). Caregivers observed that participants really looked forward to the sessions. The changes in behaviour they saw occurred primarily during the sessions, though this changes occasionally persisted for a short time afterwards. When this happened, the changes continued throughout the day of the sessions but generally dissipated the following day. Most caregivers noted participants got more relaxed and calm. Other participants got really excited, cheerful and experienced a lot of positive tension. Caregivers observed a connection between the music worker and the participant and noticed that some participants opened up, got more loose and talked more. Sometimes these observed benefits carried over to the participants' daily routines, positively affecting their interactions with caregivers. Some caregivers mentioned they observed a decrease in psychiatric symptoms, for example in tics or being less occupied with voices in one's head. Some caregivers also noted a decrease in CB. They also mentioned that the participant's regulation of emotions got better, that they could better talk about their feelings or listen/make music when they felt down or needed to relax. Most caregivers observed an improvement in self-esteem as sessions progressed, however, some others did not report this. Caregivers did not notice long-term effects. In contrast to participants, caregivers did not want to change the frequency of the music sessions (2 times per week), although their first response to the frequency was that this was quite intense and hard to fit in the schedule of the participants. For future music activities though, they preferred once a week, albeit they thought that some participants would want to make music every day. Caregivers did, however, reckon an hour was perhaps too long for some participants, for example for participants with a short attention span or participants who are easily distracted.

Yes, I really liked the fact that they could participate and I did get really excited about it myself, because they liked it and they also really enjoyed it. And that really did come first for me, that they really enjoyed it. The moment they were attending a music session, you also noticed that there were other residents who really wanted that too (caregiver).

Acceptability of music workers

Music workers also observed that participants looked forward to the sessions, enjoyed the sessions, got more relaxed and calm, and saw a decrease in psychiatric symptoms. They felt a strong mutual connection. Most music workers did see an improvement in self-esteem as sessions progressed. In line with the caregivers, they found an hour too long for some participants. On the other hand, music workers also mentioned that most participants were able to concentrate and focus on the music during the sessions. When asked what they thought of as the most beautiful moment during the intervention, 30% answered the contact with the participant and the connection between them, while 21% of the music workers also enjoyed seeing the personal development and growth of the participants. Another 16% mentioned they found it beautiful to see the emotions of the participants, them being touched by music and enjoying music. Additionally, 9% mentioned they loved to see that the participant showed self-pride, 9% loved seeing the improvisation during the sessions, and another 9% valued seeing participants relax and maintain their focus during the session. Finally, 7% found participants' enthusiasm particularly beautiful.

I am told by more and more caregivers that the music sessions are doing PPN96 very well. She likes it, is a bit happier during the day, and gradually during the hour she also makes more and more jokes, and joins in incredibly well. Wonderful to see. She didn't want to go to day care after class today at first, but eventually went. As if through the music she crawled out of her negative spiral and became happier again. Nice work! (music worker)

Sustainability

Maintenance

When participants were asked if they had continued music making after the last music session, 53% gave a positive answer. Examples of what they did were: having weekly sessions with the music worker, playing guitar at work and singing songs. There were also several musical activities mentioned by caregivers that continued after the intervention, for example turning on the radio, singing songs together, and organizing a music bingo or a karaoke event. In three cases the music worker kept on coming after the last session and in one case this was turned into a group music

activity. It was also mentioned that a volunteer was coaching a band at one facility. In addition, one participant became a member of an established music band, while another began taking music classes, and another participant acquired a musical instrument (handpan).

Several reasons were mentioned by caregivers for not continuing music making. Caregivers did not know what was expected from them regarding the continuation of music making after the intervention stopped and said they needed more knowledge and guidance. It was often mentioned, not only by caregivers, but also by participants and music workers, that the participant needed another person to stimulate music making. In line with this, some participants did not like making music on their own. The interest in music and music sessions diminished once the music sessions ended. In addition, the caregivers stated that they experienced high workload and stated that they had too little time to make music as a priority. As a consequence, the box with music instruments which was offered to every participating facility was often kept unused. Furthermore, the inability to provide adequate support, coupled with a lack of motivation or a failure to recognize the potential benefits of providing musical support also hindered caregivers. Some caregivers found that other team members perceived the study as their personal project and thus did not get involved, resulting in the activity of music making not being carried out by the entire team of caregivers. Other reasons for not continuing music making were a busy schedule of the participants and a relocation.

Discussion

This is the first process evaluation of an active music intervention, addressed from the point of view of participants, caregivers, as well as music workers. This was done by exploring the participation in the RCT, the implementation of the intervention, the experiences with the intervention and the sustainability of the intervention. It was shown that an active music intervention for people with ID is feasible and attractive, although some challenges require further exploration.

Recruitment and participation in the RCT

With respect to the trial design, one of the main challenges was the initial low number of participants referred to the study by professionals. In our study, we developed a new strategy as the study progressed (i.e., engaging other stakeholders and capturing the attention of management) which eventually led to a sufficient recruitment number. These recruitment challenges are often seen in RCTs with people with ID (Lally et al., 2022; Mulhall et al., 2018). Recruitment numbers might be higher when researchers contact possible participants directly, rather than relying on professionals

to introduce the study and seek consent (Cleaver & Sakar, 2010). In addition, providing more direct access to participants could shorten recruitment time and enable a more engaging, participatory consent process for people with ID (Lennox et al., 2005). The intended sample size of people with ID was eventually reached. We chose a smooth procedure of screening for inclusion and exclusion, by which we involved psychologists to assess the required level of CB and the absence of dementia resulting in some participants who had signs of dementia or too little CB. CB is associated with psychiatric disorders among people with mild or moderate ID (Peña-Salazar et al., 2020) and the prevalence of a psychiatric disorder in our sample (18.6%) was even lower than a sample of people with mild to moderate ID without CB (29%) (Mazza et al., 2020) and much lower than a sample of people with ID and CB (69%) (Myrbakk & Tetzchner, 2008). It is therefore doubtful whether we actually reached participants with high levels of CB in our study sample. If strict criteria are relevant for a sample, implementing a preliminary assessment prior to the study commencement is essential, although this would be seen as too much work and potentially decrease the recruitment rate further.

Retention was similar to, though slightly higher than in, other intervention studies involving individuals with primarily mild or moderate ID (Lally et al., 2022; Matthews et al., 2016; Van Schijndel-Speet et al., 2017). What probably contributed to this high level of adherence and high retention rates were the one-to-one delivery (just as in Lally et al., 2022), less invasive data collection measures (Matthews et al., 2016; Van Schijndel-Speet et al., 2017) and the domiciliary setting of the music intervention (just as in Van Schijndel-Speet et al., 2017). In addition, the musical budget that participants in the control group received after the last measurement was completed may have contributed to the high retention rate in this group. The limited amount of missing data was not a significant concern due to the multilevel analysis conducted on the primary outcome data (Twisk et al., 2018). Inter-rater bias emerged in the proxy questionnaires, likely due to staff turnover and high caregiver burden—a common challenge in ID-RCTs (Mulhall et al., 2018). This also contributed to proxy measures being submitted less promptly than self-report measures. Additionally, self-reports in the control group were more frequently on time at post-intervention compared to those in the music intervention group, because they had a longer data collection timeframe and their assessment was scheduled in advance, when the researcher was at the facility for the baseline measurement. To have a high ‘on time’ score for self-reports, it is recommended to schedule all assessments at baseline, and to reschedule only if the intervention has not yet been completed then.

Intervention implementation

The realised dose of active music making delivered to participants was in accordance with the allocated dose as outlined in the manual and the fidelity with regard to the number of music sessions within the allocated time frame was quite high. Adherence to the study protocol by participants, staff, and researchers is essential in an RCT to ensure consistency and validity of the results (Mulhall et al., 2018). However, minor adaptations in the delivery of the intervention were needed to be made by the music worker, for example in the order of activities or in the activities themselves. These adaptations were mainly done in order to adjust to the participants' preferences or needs and may have contributed to the intervention's success in terms of retention and acceptability. Personalizing or tailoring an intervention to take into account, for example, communication and cognitive difficulties is important in interventions for people with ID (Lally et al., 2022; Royston et al., 2023). However, some major adaptations, such as combining sessions or changing sessions into a dinner, reflect poor fidelity and possibly compromised the intention of the intervention. Overall, it is interesting that the adaptations were made mainly for younger participants (below the median age of 50), and for participants with mild ID. Perhaps these participants are more capable communicators and are more precise to formulate their needs. Furthermore, they are more keen to be engaged in a conversation, which was often mentioned by music workers as an adaptation. This observation highlights the need for further research to explore this.

Experiences with the intervention

Overall, participants, caregivers, as well as music workers were enthusiastic about the music intervention, as indicated by the high participation rates and qualitative feedback. Participants looked forward to the sessions and did not want the intervention to end. Caregivers observed improvements in self-esteem and emotion regulation and a decrease in psychiatric symptoms and CB. Sometimes observed benefits carried over to the participants' daily routines, positively affecting their interactions with caregivers. The mutual interaction and the strong connection were also an important factor for music workers. This interpersonal relationship is considered crucial in music programmes (Murphy & McFerran, 2017) and is possibly more important than the activity itself (Andring, 2015). However, the significance of this relationship and the increased level of attention in an individual music intervention requires further research.

Sustainability

More than half of the participants continued music making after the intervention ended. This is a promising finding, given that it is hard to sustain change in people with ID (Heller et al., 2011).

However, it also became clear that attention needs to be given to the needs of caregivers in order to maintain the benefits of the intervention. They may have limited knowledge of music interventions and require information and support to effectively encourage people with ID to continue music making. Caregivers may see this as an addition to their personal workload on their already full schedule, especially given the limited support from other team members, and consequently this may become a low priority. Therefore, to enable caregivers to better support people with ID, it may be necessary to have regular meetings with caregivers to help address any challenges they are facing and provide positive reinforcement for any music-making activities they are promoting or, even better, engaging in. It is also necessary to (financially) compensate them for attending these meetings or it should be considered part of their working hours, in addition to core duties (Kersten et al., 2023). Furthermore, teamwork plays a critical role in knowledge sharing and application, with support and assistance among team members acting as key facilitators (Kersten et al., 2018).

Strengths and limitations

A strength of this study is the triangulation of data: participants, caregivers and music workers were involved in the process evaluation and various data sources were used. The execution of each music session has been monitored during the entire intervention period, ensuring that detailed and valid information about the implementation was available. Furthermore, a number of important findings emerged from the qualitative data which would not have been evident based on the quantitative analyses such as the importance of tailoring the intervention to individual needs and the importance of the mutual connection between music workers and participants.

Some limitations should also be mentioned. The measures used in this process evaluation were self-constructed and therefore not previously validated for this population. Further, the researchers did not carry out direct observations in the music sessions and consequently had to rely on the log reports, which were completed by the music workers themselves. This might have caused more positive reporting of the intervention's implementation. However, the music workers noted numerous adaptations that were made, thus indicating their ability to assess their own work and their answers were interpreted alongside the participants' and caregivers' questionnaires. Another limitation is that no interviews with participants or music workers were carried out, which could have enriched the perceptions of the implementation and acceptability of the music intervention. Last, there is no information available on people who declined participation or on facilities that chose not to participate, nor on the factors that could have encouraged their involvement.

Conclusion

This is the first process evaluation exploring the circumstances and results of an individual music intervention for adults with ID. The findings suggest that once participants are recruited, interventions of this nature can proceed with minimal attrition. The intervention was successfully implemented in terms of fidelity and adaptations were made in order to tailor the intervention to every person's needs. The intervention was well accepted, feasible and applicable. Furthermore, our study highlights that for supporting staff having enough time to involve clients in music activities and working in a stimulating context (i.e., to avoid high workload, low morale) is of utmost importance. These factors can contribute to the sustainability of interventions concerning music making.

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Appendix

Description of the music intervention, based on Robb et al. (2011)

Overall

The music intervention consisted of 16 individual music sessions held twice a week within 8 to 10 weeks. The sessions took place in a quiet setting, typically in the participants' apartments or another room within the facility that ensured a disturbance-free environment. The music intervention was guided by a training manual for the music workers, in which the frequency, average time per (elements of the) session, and the content of every music session were outlined (see below). The manual was designed based on the input of different advisory groups with professional music educators, caregivers, and people with ID. In general, during the sessions, the music worker adapted the genre or specific pieces of music to the wishes and preferences of the participant. In the manual, no guidelines were provided regarding the acceptable decibel levels or who should be responsible for controlling the volume. After each session, the music worker completed a log report containing a series of questions about each session: whether the session was carried out in accordance with the manual, how much time was used for active music making, as well as any other additional remarks they wished to make.

Music worker

The music sessions were conducted by a music worker, who could be anyone who is able to play an instrument and is enthusiastic and capable of conducting musical sessions with a person with ID. In practice, most often music workers were music therapists, music therapy students, musicians, music educators (‘muziekagoog’ in Dutch) and caregivers.

Music sessions

Contents of the music box

In this study, a music box was provided for the music sessions. The music box contained a variety of instruments including a jingle stick, hand drums (consisting of one small and one large drum), bongos, a triangle, a percussion egg, a rainmaker, chimes, djembe drums (consisting of one small and one large drum), a woodblock, boom whackers, a cajon, maracas, a hand guiro, a microphone with Bluetooth capability and a speaker, a small keyboard, and a guitar.

Preliminary meeting

The preliminary meeting, with a duration of 30 minutes, was conducted to finalize the planning and logistical arrangements for the music sessions, which included determining the location and the methods of communication. Additionally, the meeting served to gather relevant information about the participant. The participant, caregiver, and the music worker attended this meeting.

Structure of the music sessions

The music sessions were standardized and had a duration of approximately 60 minutes. The sessions followed a structured format. The structure of the music sessions can be found in Table 1.

Table 1

Structure of the music sessions

Activity	Duration	Goal
Entry	2 minutes	The music worker asks the participant how he or she feels at that moment (happy, sad, scared, angry or tired) and the intensity of this feeling (little, normal, intense) and writes down the answer in the log report.
Welcome song	2 minutes	The welcome song may either be an existing song, or a personalized song adapted specifically for the participant and is accompanied by an instrument.
Listening exercise	5 minutes	The primary objective of the music listening task is to focus and maintain attention. In this phase, a diverse range of musical pieces is played, covering different styles and genres. Following the musical piece, a question is presented to the participant, inquiring about their perception and interpretation of the music, such as identifying specific elements heard, evaluating the emotional tone of the music, or recognizing narrative themes in the music.
Warming up	5 minutes	The warming-up activity is intended to stimulate the body and/or vocal cords of the participant. The specific exercises employed during this phase are determined by the preferences of both the music worker and the participant. Examples of exercises may include imitating a specific rhythm or dancing, followed by a stop when the music comes to a stop.
Active music-making	35 minutes	In this section, the participant actively engages with the musical instruments from the music box. Throughout each session, a specific theme is emphasized, which is further elaborated below.
Cooling-down	5 minutes	The cooling-down is intended to facilitate a peaceful and relaxed transition out of the active music-making phase of the session. Various techniques may be employed to support this aim.
Farewell song	2 minutes	During the last activity, the music worker and participant play a familiar, predetermined song. This song remains consistent across sessions, providing a comforting and familiar routine for the participant.
End	2 minutes	The music worker asks the participant again how he or she feels at that moment (happy, sad, scared, angry or tired) and the intensity of this feeling (little, normal, intense) and writes down the answer in the log report.

Area of focus per music session

The contents of each music session were tailored towards a specific area of focus, which is shown in Table 2. Music workers had their own freedom in designing appropriate exercises, but several examples could be found in the manual. A diploma was awarded to the participant at the final session as a completion certificate.

Table 2

Content of each music session

Number of music session	Area of focus	Content
1	Making contact / getting acquainted	The welcome song is utilized as a tool to facilitate self-introduction. The 'listening exercise' and 'warming-up' parts are used for self-introduction by using a music fragment chosen by the participant, which is then discussed through a series of questions. Following this, the purpose of the sessions is outlined, and the instruments are introduced.
2-6	Emotions	These sessions are focused on different emotions and the influence of music on them. The sessions are subsequently structured around the following variables: enjoyable music vs. unenjoyable music, levels of tension versus relaxation, happiness, sadness, and alertness.
7-12	Executive functions	These sessions are focused on executive functioning, specifically focusing on inhibition, working memory, and cognitive flexibility. The sessions include exercises, such as recalling sounds (working memory), playing the opposite of the music worker's actions (cognitive flexibility), and halting movement when the music stops (inhibition).
13-16	Making the final song	The objective of this part is to produce a final song, which may either be instrumental or vocal. The final song incorporates the participant's hobbies, interests, or traits. Additionally, the participant's preference for inviting guests to the final song presentation and whether to record the final song or not are explored.

Final meeting

The final meeting involved evaluating the participant's overall satisfaction with the music sessions and providing recommendations on how the participant may continue pursuing music if they so desire. The final meeting was attended by the participant, music worker, and caregiver.

CHAPTER 9

9

Chapter 9

General discussion

The main objective of this thesis was to investigate the effects and feasibility of a music intervention for people with intellectual disabilities (ID) who live at residential facilities in the Netherlands. Part 1 is about a pilot cluster-RCT, in which the effectiveness and feasibility of a weekly music group intervention was compared to leisure group activities (**chapters 2 and 3**). Part 2 describes a partially online music intervention during COVID-19 to examine the feasibility, experiences, and short-term outcomes in challenging behaviour (CB) and well-being of adults with ID (**chapter 4**). In part 3 we performed a cluster-randomised controlled trial (RCT), with the protocol detailed in **chapter 5**. During the cluster-RCT we tested the effectiveness of an individual music intervention on CB, mental health, self-esteem and well-being (**chapter 6**), executive functioning (EF) (**chapter 7**) and evaluated the cluster-RCT and music intervention in a process evaluation (**chapter 8**).

In this closing chapter an overview of the main findings will be provided. Next, four main key findings are provided, followed by an overview of the strengths and limitations of this thesis, its implications for clinical practice and suggestions for future research are discussed.

Overview of main findings

Part 1: Pilot cluster-RCT

In a pilot cluster-RCT with 29 adults with ID, no evidence was found that caregiver-assisted active music making in a group improves well-being, self-esteem, social acceptance, CB and EF more than leisure activities within 16 weekly sessions. In fact, participants in the leisure activities group intervention showed significantly higher inhibitory control and significantly less internalizing behaviour at post-intervention compared to participants attending the music group intervention (**chapter 2**). The feasibility study showed that the music group intervention seemed suitable in practice. People with ID gave positive feedback about the music intervention as did music providers as well as caregivers, who reported greater happiness, calmness, and social bonding between the participants in the music intervention compared to those participating in the leisure intervention. They also reported that the leisure intervention was not feasible due to practical issues resulting in a lack of appeal (**chapter 3**). The overall design of this study was found unfeasible due to issues, such as the high demands of the tasks (i.e., homework assignments, completing forms, questionnaires, and tests) and the excessive workload as perceived by caregivers. It was concluded that three major adaptations were needed to perform a large-scale RCT: (1) an individual intervention tailored to different cognitive abilities; (2) care-as-usual (CAU) as a control condition; and (3) improve

communication over the goal and purpose of the intervention and clarification of the roles of all actors in the intervention, resulting in a project group per facility.

Part 2: COVID study

Since COVID-19 appeared in 2020, we had to postpone the intended cluster-RCT and decided to conduct another pilot study (**chapter 4**). In this mixed-methods study we included 10 participants with mild or moderate ID who received 16 one-hour individual music sessions in 10 weeks, either face-to-face or online. Participants and the providers of the music intervention generally reported positive experiences concerning taking part in the music intervention, and the overall satisfaction of the participants was high. The face-to-face music sessions were preferred to online music sessions. Adjustments to tailor an online music intervention include beginning with face-to-face sessions, adding shorter online sessions and using adequate digital platforms. Furthermore, though CB of these participants showed improvements, the results concerning well-being were not conclusive.

Part 3: Cluster-RCT

Based on the findings in the aforementioned studies, the design of the cluster-RCT was adjusted to reduce the demand and reliance on caregivers by making use of more appropriate tests and questionnaires, a higher intensity and individualised music intervention, replacing the active control group with a CAU control group and working with a broad range of music providers, instead of only music educators ('muziekagoog' in Dutch). These modifications were taken into account in the study protocol for the cluster-RCT (**chapter 5**). In this cluster-RCT people who wanted to participate in the study were assigned to one of two groups: 1) the intervention group received 16 individual music sessions in a maximum of 10 weeks and 2) the control group received CAU. Compared to participants in the control group, participants in the individual music intervention showed significant improvements in CB at post-intervention and follow-up, and reductions in anxiety and depression at follow-up. The music sessions also helped to stimulate positive emotionality. Compared to participants in the control group, no significant changes were found in well-being and self-esteem (**chapter 6**) or EF (**chapter 7**) in participants in the individual music intervention. Unfortunately, the measures used for EF were found to be too difficult for some participants, especially for those with moderate ID. In the process evaluation of this cluster-RCT it was found that recruitment for the RCT proved challenging, though response rates for the self-report and proxy measures of the study's primary outcome data and music intervention retention rates were high (**chapter 8**). The intervention was implemented successfully with high fidelity, and adaptations were made to tailor it to individual needs. The music intervention was well accepted by participants, caregivers and music

workers, feasible and applicable in practice, with promising indications for sustainability. However, caregiver's workload and little perceived support presented challenges for the intervention's future usability.

Reflection on the findings

Reflecting on the results of this thesis, four main key findings emerge:

1) ***An individual music intervention is effective in reducing CB and promotes the emotional well-being and mental health of people with ID***

This thesis showed that an individual active music intervention is an effective and sustainable way of improving CB in adults with mild or moderate ID (**chapter 6**), even when the music intervention is partially online (**chapter 4**). Additional analyses showed that internalizing behaviour as well as externalizing behaviour significantly improved more in the music intervention condition compared to the control condition (**chapter 6**), which is in line with results of previous studies in children and adolescents with ID (Kalgotra & Warwal, 2017) or without ID (Geipel et al., 2018; Ye et al., 2021). That music as a non-biological and non-psychotherapeutic intervention is effective for CB is an important finding, since around 10 to 18% of people with ID display CB (Bowring et al., 2017; Lowe et al., 2007) and most other interventions for people with ID are biological interventions (i.e., pharmacological) (Heyvaert et al., 2010) with the risk of adverse side-effects (Deb et al., 2007). Since CB is negatively linked to well-being (Myrbakk & Von Tetzchner, 2008), a music intervention that reduces CB might also impact the quality of life (QoL) of people with ID (Friedman, 2018).

Besides the positive effect of an active music intervention on CB in people with ID themselves, it is also promising for the well-being of caregivers. When people with ID show CB, caregivers experience less job satisfaction (Deveau & McGill, 2019), more stress (Raczka, 2005; Smyth et al., 2015) and are more likely seeking to leave their employment (Deveau & McGill, 2019; Olivier-Pijpers et al., 2020; Robertson et al., 2005). In addition, working with people with CB is correlated with higher rates of burnout among caregivers (Deveau & McGill, 2019; Nevill et al., 2022; Shead et al., 2016). Given the high staff turnover in facilities (De Bles et al., 2022; Hithersay et al., 2014; Lennox et al., 2007; Morisse et al., 2013), employee commitment may increase when CB is reduced (Smyth et al., 2015).

In addition, this thesis found evidence for the fact that a music intervention can help to stimulate positive feelings (**chapter 6**). People with ID and CB often exhibit an inability to properly cope with emotions and it is known that music can elicit and help regulate already existing positive feelings

(Cook et al., 2019; Moore, 2013; Sakamoto et al., 2013; Van Goethem & Sloboda, 2011). Even though most of these positive effects on feelings were short-term findings (Cooke et al., 2010; Cooke et al., 2010), in this thesis a decrease of anxiety and depression was found in participants with ID at follow-up (**chapter 6**). This might imply a long-term (delayed) effect, or could be due to continued musical engagement after the last planned music session, since more than half of the participants in the music intervention continued music making after the last session (**chapter 8**). In sum, music interventions stimulate positive feelings, and reduce anxiety and depression, which increases the emotional well-being and mental health of adults with ID.

This thesis did not find a significant effect of a music intervention on EF, self-esteem or directly on well-being as an outcome measure. It is possible that the amount of cognitive engagement during the music sessions was not enough to stimulate EF (**chapter 7**), but it is also possible that interventions are mainly effective on strengthening EF in younger adults and adults with mild ID, because even leisure activities in a group activity showed a positive effect on inhibition, i.e., an EF variable, in this subgroup (**chapter 2**). This probably can be attributed to differences in brain plasticity, either due to age-related decline, given that inhibition decreases from the age of 35 (Ferguson et al., 2021) or to low levels of brain plasticity in people with a more severe ID, which leads to smaller intervention effects (Söderqvist et al., 2012). Perhaps, people with moderate ID require alternative intervention strategies when targeting EF, such as altered durations or more tailored tasks (Kirk et al., 2015; Söderqvist et al., 2012). On the other side, EF is difficult to measure properly, especially in people with moderate ID (**chapters 2 and 7**). This warrants further research to better understand the effect of interventions on EF in this subgroup of people with ID. This thesis also did not find a significant positive effect on self-esteem as outcome measure, assessed by a self-report questionnaire (i.e., subscale Global Self-Worth of the Self-Perception Profile for Adolescents) (**chapter 6**). However, caregivers and music workers observed an improved self-esteem in most participants (**chapter 8**) and it was also found that the self-esteem of participants in the music group improved in the pilot cluster-RCT, however not in comparison to those participating in the leisure activities group (**chapter 2**). Finally, this thesis did not find a direct positive effect on well-being as outcome measure which might be due to a ‘ subjective well-being homeostasis’ (i.e., subjective well-being is fairly constant in diverse living conditions) (Cummins, 2005; McGillivray et al., 2009) or a limited intra-individual variability, which could have negatively influenced the sensitivity to detech change (Coster, 2013) in our sample. This limited intra-individual variability is probably caused by the fact that there were only three response options in the questionnaire. This is however recommended in Likert scales for people with mild to moderate ID (Kooijmans et al., 2022). In conclusion, while this

thesis did not identify a significant effect of a music intervention on EF, self-esteem, or well-being as outcome measures, it highlights potential factors influencing these findings.

To round off this key finding that focused on the effect of interventions for people with ID who already have CB, it is also important to identify and focus on methods for preventing CB to become established in an individual's repertoire (Lloyd & Kennedy, 2014). This can be done through the use of prevention methods that target individuals at risk or that make use of early interventions (Allen et al., 2013), with the selection of effective, evidence-based interventions being crucial. Effective interventions that target CB in adults with ID need to focus on emotion regulation, provide meaningful activities, are individualized and tailored to individual needs (i.e., adaptations in intervention content, session order, pace, or duration and delivery format) (Royston et al., 2023). It is evident that a one-size-fits-all generalised approach to address CB is inappropriate for people with ID. While interventions such as music may hold promise, it is essential to continue exploring and evaluating a range of individualized, evidence-based approaches to both prevent and address CB in people with ID. This raises the research question to be asked: are music activities the most effective intervention, or are there other interventions that might also target CB effectively?

2) Unclear whether music is beneficial over other enjoyable activities

Making music can be an effective intervention, but perhaps a music intervention is not more effective than other enjoyable and engaging interventions (Degé & Frischen, 2022). In previous studies in typically developing children and people with dementia, group-based music activities were not more effective than other structured, enjoyable activities such as cooking, reading, dance lessons and art classes, when it comes to behavioural functioning (Samson et al., 2015; Vink et al., 2014), emotional state (Samson et al., 2015), QoL, depression and self-esteem (Cooke et al., 2010), agitation and anxiety (Cooke et al., 2010) and different components of EF (Bowmer et al., 2018; D'Souza & Wiseheart, 2018; Kosokabe et al., 2021). Only two studies found that musical activities which lasted a long time, specifically 8 months (Frischen et al., 2021) and 2.5 years (Jaschke et al., 2018), were found to be more effective than other activities on some components of EF. In **chapter 2** we compared music activities with leisure activities and found that leisure activities were more effective when it comes to internalizing behaviour and inhibition. To determine what intervention is most effective for which person with ID, it is necessary to look at other factors rather than a "one-size-fits-all" approach.

Other factors that determine 'which intervention is most effective for whom', depends also on what the person likes to do most and the connection this person has with the person who provides the intervention (Andring, 2015; Murphy & McFerran, 2017). A strong personal connection is possibly more important than the activity itself (Andring, 2015) and is a core element in multiple approaches (Tournier et al., 2023), which was also confirmed in our studies by participants, music workers and caregivers (**chapters 4 and 8**). For a real connection, a joint engagement in a meaningful activity, a safe atmosphere and alignment to the needs of people with ID are important (Tournier et al., 2023). All components were present in the music intervention. Besides this connection, the person should have the possibility to choose what he or she likes to do most and that gives an intrinsic reward and achievement (Campbell, 2021). In our studies, participants were able to choose the type of music, musical instruments (**chapters 2, 4, 6 and 7**) and what leisure activity to do in each session (**chapter 2**). This is possible the reason that our participants enjoyed the music sessions very much (**chapters 3, 4 and 8**). The combination of an enjoyable music activity with a strong connection with the music worker resulted in our studies in a significant decrease in CB, anxiety and depression, and caregivers observed improvements in self-esteem and emotion regulation. They also observed calmer and more relaxed participants which resulted in more pleasant behaviour in the group directly after the music session (**chapter 8**). Sometimes these observed benefits carried over to the participants' daily routines, positively affecting their interactions with caregivers as well. In sum, while music may not outperform other enjoyable activities on certain outcome measures, people with ID enjoy making music, and when there is a good connection with the music worker, it results in multiple positive behavioural and emotional effects.

3) The music intervention is feasible with high acceptability

This thesis showed that the participants responded favourably to the music intervention, which was well-received and effectively engaged adults with ID (**chapters 3, 5 and 8**). Participants looked forward to the music sessions and did not want the sessions to stop. Caregivers noticed that participants enjoyed the music sessions and were happy and enthusiastic about the music sessions, which was also reported by the music workers. Besides this enjoyment and the good connection with the music worker, as mentioned above, this thesis also showed that it was feasible to implement the music intervention, with high dose and fidelity scores. Furthermore, in the music intervention adaptations were mainly made in order to adjust to participants' preferences or needs. These adaptations may have contributed to the intervention's success in terms of retention and acceptability. However, when activities, e.g., music sessions or answering questions in proxy

questionnaires, depend on caregivers, they are found to be less feasible (**chapters 3 and 8**), which is recognized as a major challenge when training caregivers (Beeber et al., 2010). Due to the staff shortage in the ID sector, it is not surprising that in all our studies of this thesis the caregiver burden was perceived as high. Despite this finding, all caregivers that participated in the semi-structured interviews (**chapter 8**) recommended to their colleagues that residents should participate in the cluster-RCT.

4) Characteristics of music intervention important for success

Besides the enjoyment and connection with the music worker, this thesis shows that four other characteristics of the music intervention seem to be necessary to be an effective intervention. First, **individual** music sessions are preferred over group sessions, which is also in line with previous research (Pelletier, 2004; Vasionyte & Madison, 2013) and the fact that we did not observe an effect on CB in a group-based music intervention (**chapter 2**), but did find such an effect in an individual music intervention (**chapter 6**). Individual sessions can be tailored to the specific needs and ability levels of the participant. This is especially important for people with ID, due to their diversity in musical preference, cognitive functions, communicative and social skills (Mulhall et al., 2021; Ptomey et al., 2017). In addition, in individual sessions the level of attention is higher and people with ID have the opportunity to use the sessions for what they need most at that moment, for example to speak with the music provider about personal issues instead of making music (**chapters 5 and 8**). Talking about personal issues suggests that the participant was able to connect and establish a trusting bond with this professional, something that is easier to build up in individual rather than group sessions, but which is very important for the well-being of people with ID (Van Asselt-Goverts et al., 2015; Van Asselt-Goverts et al., 2015).

Second, the **duration, intensity and frequency** of the music sessions is important to consider. In our cluster-RCT we found that intensity and frequency of the music intervention did not have a significant impact on the outcome measures (**chapters 6 and 7**). Perhaps the additional benefits of increasing the frequency of individual music sessions (3 times a week) or the amount of music making (in attendance and minutes of active music making) become marginal after a certain dosage is reached (2 times a week, 12 sessions or 480 minutes of active music making) (**chapter 6**). While a weekly music intervention did not show significantly greater effects on measures compared to leisure activities (**chapter 2**), significant within-group improvements in self-esteem were observed in the group of people with ID that received the music intervention. Previous studies have also reported significant outcomes following weekly music interventions, but often lack comparison with a control

group (Hillier et al., 2011) or demonstrate comparable benefits in EF and depression across both a music group and another arts-based control group, along with significant within-group increases in self-esteem for the music group (Park et al., 2015). Compared to intensity and frequency, the duration and total amount of music sessions might be more important. In a scoping review about music and young people with ID, it was found that most interventions consisted of only 10 sessions, concluding that this small number of sessions may not provide the optimal conditions for the participants to get used to the settings or the intervention (Després et al., 2024). This is also supported by two studies demonstrating that extended musical activities, lasting 8 months (Frischen et al., 2021) or 2.5 years (Jaschke et al., 2018), significantly improved EF in typically developing children. Our finding in the cluster-RCT that anxiety and depression significantly improved after 4 months, is further support for a longer duration of a music intervention. So, perhaps a music intervention performed once a week over a period longer than 16 sessions is needed to establish measurable effects on for example self-esteem, EF, anxiety, and depression in adults with ID.

Third, **face-to-face** music sessions are preferred to online music sessions. Online sessions were found to be impersonal and lacking togetherness (**chapter 4**), which makes it more difficult to build the important connection with one another, as achieved through face-to-face music sessions. Additionally, when online sessions are necessary (such as during an epidemic), beginning with face-to-face sessions before transitioning to online formats was found to be beneficial for fostering this connection.

Fourth, music interventions need to be **well-structured**, with **clear objectives** that align with behavioural improvement goals. Music interventions with a recognizable structure, recurring elements and fixed sequence, with a series of positive actions that the participant enjoys, are important elements to stimulate behavioural change (Jackson, 2007; Pater et al., 2019). Our music intervention followed a structured, consistent format, with each session centred on a particular goal. However, if this goal is to improve EF, it is crucial to allocate more sessions specifically to this objective, as dedicating only 6 out of 16 sessions to this goal proved to be insufficient (**chapter 7**). Thus, tailoring the duration and structure of individual and face-to-face music interventions to align with specific goals is essential for maximizing their effectiveness.

Strengths and limitations

This thesis has several strengths. First, we studied the effects of a music intervention on various outcome measures for adults with ID. Second, we used multiple designs (cluster-RCT, feasibility study, process evaluation) and different methods of data collection (self-report questionnaires and

tests, proxy questionnaires, semi- structured interviews). Third, we developed an intervention manual which allowed music workers to provide the intervention in a uniform manner. This made the results more measurable and comparable. Finally, through the implementation and testing of various active music interventions, including a caregiver-assisted music group intervention, a partially online individual music intervention and a face-to-face individual music intervention, the research offered a deeper understanding of how different formats can be tailored to address the diverse needs of adults with ID. This thesis adds to the knowledge base of effective interventions for people with ID, since we found significant effects in an RCT, which is the gold standard by which causal inferences can be made (Draper, 2004).

Despite these strengths, the findings presented in this thesis also need to be interpreted within the context of the limitations of the studies conducted. A first limitation is the scope of the target population of these studies. We focused on people with mild to moderate ID, with one exception in **chapters 2 and 3**, where four people with borderline intelligence were included. Therefore, the results cannot be generalized to other ID target populations such as people with severe ID. Second, only persons with ID with a positive predisposition toward music were included, which limits the generalizability. As a result, it remains uncertain whether people with little or no prior interest in music would experience similar benefits. Third, blinding of, for example, participants, intervention providers or executors of the measurements could not be preserved at any stage, neither in the pilot cluster-RCT nor in the cluster-RCT. This could have led to for example performance or detection bias, positively influencing the outcomes of the music intervention group, which are threats to validity. Fourth, in the cluster-RCT we only compared the music intervention to CAU. Although this is preferable to engaging no control group at all, it raises questions about whether the observed improvements are the result of the music intervention itself, or are simply the result of increased levels of attention and contact with the music provider (Vink et al., 2013). Fifth, since different caregivers filled in the questionnaires at different time points about one person, this may have introduced inter-rater bias, as was mentioned in **chapter 8**. A sixth limitation relates to the lack of clarity regarding the active elements in the music intervention and how these elements impact the outcomes. Although the multicomponent nature of our music intervention likely contributed to its efficacy, it also precluded making inferences about the contribution of each component (e.g., making music, singing, listening to music, cooperation with the music worker) on the outcomes. This lack of clarity around component efficacy limits the ability to refine and target future interventions to maximize benefits. Seventh, EF was found to be difficult to measure in the pilot cluster-RCT. Although we decided to adjust the EF test measures in the cluster-RCT, the various tests used

remained sometimes too challenging, particularly for people with moderate ID. This could have led to a 'floor effect,' which is common in studies on EF in this population (Willner et al., 2010). As a result, efforts to reduce invalid or unreliable data in order to enhance the overall quality of the dataset also reduced the sample size of the pilot cluster-RCT as well as the cluster-RCT. Despite several limitations, associated with necessary decisions for feasibility or the ever-changing nature of clinical practice, this thesis underscores the value of a comprehensive approach and rigorous methodology. It contributes to strengthening the field of evidence-based interventions for adults with ID and highlights the promising potential of music interventions.

Implications for clinical practice

The insights provided by this thesis have important implications for people with ID and their caregivers in clinical practice. This is an important finding, given that in the Netherlands music is not often offered in a structured, intentional way to improve the lives of people with ID living in residential facilities (Jaschke & Scherder, 2015). Music programs are sometimes treated as recreational activities, rather than a way of intentionally and structurally targeting CB or emotional regulation. As highlighted earlier, a weekly, well-structured individual music intervention with a long duration (i.e., exceeding 16 sessions) and clear objectives is recommended. A weekly intervention is also in concordance with the wishes of caregivers (**chapter 8**). However, the insights from this thesis must be effectively communicated to and integrated into daily care practices, as they are often overlooked by professionals in the field.

Transferral of knowledge to caregivers

Even though there is an increasing shortage of well-trained caregivers in the Netherlands, the knowledge what a structured music intervention can positively contribute to the lives of people with ID needs to be transferred to caregivers who provide daily care. There is a gap between scientific knowledge and its use in everyday practice (Garretsen et al., 2007). To bring this knowledge more to practice this thesis showed that a successful music intervention that improves CB and mental health can be provided by any person, following a detailed manual, who enjoys music, plays a music instrument or can sing and is able to make a connection with the person with ID. This should not be done incidentally and on a random basis, but it is important to provide music in a structured way (Savarimuthu & Bunnell, 2002) and in an encouraging environment (Pitts, 2009). When caregivers have the aforementioned skills or are willing and able to develop them, the positive effects of music for people with ID can be more easily integrated directly into the daily care (Gotell et al., 2000). In addition, it seems that carer involvement is crucial to facilitate and sustain change (Royston et al.,

2023). Unfortunately, caregivers may still not be fully aware of how music can be used in a structured, evidence-based way. As was shown in **chapter 8**, in some facilities, music was offered sporadically or not at all after the intervention stopped, which does not create the consistency needed to make a lasting impact on the lives of people with ID. Caregivers experience time constraints and need more guidance in sustaining musical activities. Additionally, caregivers often experience "change fatigue" from the overwhelming influx of new practices to implement. Consequently, they struggle with a difficult choice: dedicating time to long-term quality improvement (i.e., a music intervention) versus focusing on providing immediate quality of care by regularly working shifts. Having to make this choice everyday can be a source of stress (Bisschops et al., 2024). To help with the transferral to daily practice, communities of practice could be established, which aim at facilitating knowledge sharing, but could also help to translate this knowledge into practical changes (Barbour et al., 2018). When setting up a community of practice, working with motivated and engaged professionals (for example the most enthusiastic caregivers) and making a tailored implementation plan can help (Bisschops et al., 2024). The support from other staff members when working with people with ID and CB is also found to be important for the well-being of caregivers (Lokman et al., 2023), thereby indicating the implementation needs to be imbedded in a team-wide approach. Teamwork is important when sharing and applying knowledge, especially the provision of support and assistance in a team are facilitating factors (Kersten et al., 2018). Furthermore, providing preconditional resources such as time, space, and budget along with a stimulating environment marked by an open, safe climate and supportive frameworks, such as multidisciplinary consultations are essential to stimulate the application of new knowledge in ID-care (Kersten et al., 2023). In line with this, for a successful implementation of the music intervention among caregivers, it is important that they receive supervision from professional music workers, have protected time to learn and practice these skills and can share responsibilities with other team members (Royston et al., 2023).This could lead to more engagement and embedment of these skills in routine practice, allowing for a more sustained change. When the music intervention is provided by a caregiver, a side benefit is that caregivers are already familiar with the persons with ID, are employed within their residential facilities, and probably already have a good connection with the person with ID. In the event of a pandemic, the intervention can continue online if necessary, as the recommended initial in-person contact to establish a bond of sense and trust (**chapter 4**) has already been achieved. However, it is crucial to enhance caregivers' capacity and technical proficiency with digital systems to effectively facilitate these online sessions.

Besides the transfer of knowledge about what music can do for people with ID, caregivers should get more acquainted with the importance of research, especially with all aspects of research designs as RCTs. Conducting RCTs is essential, as they are widely regarded as the "gold standard" for assessing the effectiveness of interventions. However, conducting RCTs is often complex, especially when it involves participants with ID and their caregivers. Many caregivers and clinical staff do not understand the requirements of an RCT (Mulhall et al., 2020). In our studies (**chapters 3 and 8**) we found that especially the recruitment through gatekeepers, data collection difficulties (i.e., timely and by the same caregiver completion of questionnaires), caregiver burden and low perceived support by caregivers were barriers to complete the intended research. These barriers are common in ID-RCTs (Hassiotis et al., 2018; Mulhall et al., 2018) and possibly lead to inadequate delivery of an intervention (Matthews et al., 2016). They also withhold to build an expansive evidence-base of effective interventions in ID care.

Potential strategies that can help to facilitate and encourage the development of RCTs in the field of ID might include directly recruiting people with ID (Cleaver & Sakar, 2010) and seeking innovative and accessible ways to increase the understanding of RCT concepts to clinical staff (Robotham et al., 2011). In addition, researchers should emphasise how the extra work for caregivers involved in making an RCT a success, will fit alongside their core duties (Robotham et al., 2011) and the organisation should provide conditional resources such as time, space and budget to stimulate caregivers in taking part in RCTs (Kersten et al., 2023). Last, the provision of support and assistance from management and other team members seems crucial when sharing and applying knowledge (Kersten et al., 2018). In conclusion, it is important to equip caregivers with a better understanding of RCT research designs, so that they can contribute effectively to the development of a robust evidence base for interventions in the ID field.

Target audience

Besides the transfer of knowledge to caregivers, it is recommended to adapt this type of music intervention especially to the needs of people with ID and CB, anxiety, depression or emotion regulation problems. There is substantial evidence that music can have beneficial effects on these outcome measures. Not only is this supported by the results in this thesis (**chapter 6**), but numerous studies report on the effects of music on CB (Pedersen et al., 2017; Van der Steen et al., 2018; Ye et al., 2021), anxiety (Lu et al., 2021), depression (Tang et al., 2020) and emotion regulation (Moore, 2013). By prioritizing people with these specific problems, the intervention's effectiveness and impact are expected to be maximized.

Future directions for research

Besides the implication for practice, this thesis has implications for future research in ID-care. First, future studies could investigate when and for who music or other leisure activities interventions are most effective, which would also fill a gap in the current leisure research base for people with ID (Campbell, 2021). By this means the unique value of an individual music intervention as opposed to special attention otherwise provided for a person can be studied. When designing such studies, it will be important to account for participant characteristics, such as age and level of ID, as these factors appear to influence the relationship between leisure activities and internalizing behaviour, and between leisure activities and inhibition (chapter 2). However, given the findings in chapters 3 and 8, there are some practical issues to be considered, given that this places an even higher amount of burden upon the various stakeholders. Therefore, alternative research designs, such as a series of n-of-1 trials (Hawksworth et al., 2024) could be used to evaluate individualized interventions while minimizing logistical challenges. Third, although the use of an intervention manual allowed music workers to provide the music sessions in a uniform manner, which made the results more measurable and comparable, the results from the process evaluation (chapter 8) showed that minor and major adaptations were made. In future research, it would be good to incorporate direct observations or video analysis of a random sample of sessions, to create a better understanding of what happens during music sessions. In line with this, it would also help to unravel the multicomponent nature of the music intervention and to make inferences about the contribution of each component on the outcomes, in order to help to identify the "most active ingredients". Additionally, physiological measures (e.g., levels of salivary α-amylase, heart rate, respiratory rate) could be valuable tools for pinpointing these components, offering objective data on the intervention's effects on participants (Leubner & Hinterberger, 2017; Nilsson, 2008; Poquérusse et al., 2018). Fourth, when examining outcomes like self-esteem and well-being, the quantitative results might differ from the qualitative findings. Detailed case reports with qualitative evaluations could provide valuable insights into the impact of the music intervention on these variables (Clift, 2012), helping bridge the gap between quantitative and qualitative perspectives. Overall, it would be interesting to further explore music interventions with different research designs, in order to strengthen our understanding of their benefits for adults with ID.

This thesis highlights the potential of an individualised music intervention for adults with mild or moderate ID and CB. However, questions about the role of attention and the added value of a music intervention compared to other effective interventions remain unanswered. Music as an intervention should be integrated more into the daily care routines of people with ID living in residential facilities,

and caregivers should be equipped and supported to incorporate it effectively. Given time and funding limitations, it is essential to prioritize music interventions as a valuable tool for enhancing the behaviour, mental health and well-being of people with ID and their caregivers.

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APPENDICES



Appendices

Summary
Samenvatting
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APPENDICES



Summary

General introduction

Chapter 1

The general introduction (chapter 1) provides an overview of the literature to better understand the challenges faced by people with intellectual disabilities (ID) and examines the potential of music interventions as a strategy to address these challenges.

One of these challenges is challenging behaviour (CB). In the Netherlands, approximately 1.45% of the population are people with ID, of which 18.1% exhibit CB. Challenging behaviour can be defined as behaviour that is harmful or distressing to the individual or their environment. It occurs more frequently in people with ID than in the general population and is associated with factors such as trauma, limited communication abilities, and social exclusion. CB negatively affects quality of life (QoL) for both individuals and caregivers, contributing to increased stress and caregiver burnout. People with ID also tend to have deficits in executive functioning (EF), including inhibition, cognitive flexibility, working memory, and attention, which are further impaired when CB is present. Additionally, individuals with ID are more likely to experience mental health issues, social isolation, chronic stress, and low self-esteem, all of which further reduce their QoL and psychological well-being.

A promising strategy to address these concerns is a music intervention, which has been shown to enhance social interaction, improve mental health, and reduce stress. Existing research in general and clinical populations (e.g., dementia, autism spectrum disorder, typically developing children) supports the beneficial effects of music interventions on well-being, mood, EF, CB, and social functioning. In people with ID, particularly those with mild to moderate ID, non-experimental studies report positive associations with improved QoL, mood, social engagement, and reduced CB. However, experimental studies are limited, often small-scale, and methodologically constrained (e.g., lack of control groups, short follow-up).

Despite these limitations, the research to date indicates promising potential for music interventions in supporting individuals with ID. To address the current evidence gaps, the present thesis investigates the effects and feasibility of active music interventions for adults with ID living in residential facilities in the Netherlands. Using a mixed-methods approach, the research explores group and individual formats, in-person and digital music interventions, and their impact on CB, EF, mental health, self-esteem, and well-being. The thesis is structured in three parts, each addressing a specific research question:

Part 1: Pilot cluster-RCT

What are the effects and feasibility of a weekly music group intervention compared to leisure group activities (chapters 2 and 3)?

Part 2: COVID study

What are the feasibility, experiences and short-term outcomes of a partly digital music intervention twice a week (chapter 4)?

Part 3: Cluster-RCT

What are the effects and feasibility of an individual music intervention twice a week compared to care-as-usual (CAU) (chapters 5, 6, 7 and 8)?

Part 1: Pilot cluster-RCT

Chapter 2

Chapter 2 presents a pilot study that assessed the effects of two interventions, active music making and leisure activities, in adults with ID. In previous research participation in leisure activities for individuals with ID is associated with improved well-being, enhanced social skills, and a reduction in emotional and behavioural problems. Active music making also appears to enhance well-being, social acceptance, and reduce CB. The study compared the efficacy of a weekly caregiver-assisted active music group intervention with a weekly caregiver-assisted leisure activities group intervention, using pre-intervention, post-intervention, and follow-up measurements. Outcome measures were well-being, self-esteem, social acceptance, CB, and three components of EF (inhibitory control, cognitive flexibility, and visual working memory).

This pilot cluster-RCT involved 29 adults with mild ID, moderate ID, or borderline intellectual functioning across four residential facilities. The music intervention was provided by trained music workers, following a structured routine to ensure consistency and replicability. Sessions included preparation with a coordinating caregiver, a fixed sequence of musical activities, and active participation from both participants and multiple caregivers. The leisure activities intervention consisted of interactive non-musical activities (e.g., gaming, darts, cards, baking, puzzling) and matched the music intervention in terms of group interaction and attention.

While within-group improvements were observed in both interventions, no overall superiority of the music intervention was found. Notably, individuals participating in leisure activities showed greater

reductions in internalizing behaviour and improvements in inhibitory control post-intervention than individuals participating in the music intervention, particularly among younger individuals with mild ID. Participants in the music intervention reported higher self-esteem and lower need frustration afterwards, but these effects were not significantly stronger than those involved in leisure activities.

The findings suggest that general leisure activities may be effective in enhancing certain aspects of well-being and EF, especially for younger participants with mild ID. Engagement in music activities may require longer duration, greater frequency, or more individualized approaches to achieve significant outcomes. Methodological limitations included small sample size, measurement difficulties in moderate ID participants, and intervention heterogeneity. Future research should consider longer-term, tailored interventions and use more inclusive and appropriate assessment tools to better evaluate outcomes in this diverse population.

Chapter 3

Conducting pilot studies and feasibility testing have proven to be valuable strategies in preparing for the implementation of large-scale RCTs. Therefore, in chapter 3 the feasibility of the pilot cluster-RCT comparing a music group intervention with a leisure activities group intervention among adults with ID was assessed. This study focused on six key areas of feasibility: acceptability, demand, implementation, practicality, adaptation, and limited efficacy of the pilot. The experiences and perspectives of both the people who received (one of) the interventions (adults with ID) and the people who provided (one of) the interventions (professional caregivers, music workers and a research assistant) were assessed. A mixed-methods approach was employed, in which log reports, evaluation questionnaires, and RCT outcome measures (proxy questionnaires, self-reports, and EF tests) were analysed.

Engagement in the music group intervention was generally well-received by adults with ID, who reported enjoyment, emotional engagement, and a sense of group connection. Caregivers and music workers observed some positive effects, though caregiver involvement was inconsistent, and homework tasks were often too demanding. Implementation was challenged by logistical issues and varying levels of cognitive ability among participants. In contrast, engagement in the leisure activities group intervention was found less appealing and added little value to daily routines. The RCT design itself proved overly demanding for both caregivers and participants. Caregivers struggled with the workload, unclear communication, and low motivation to complete research tasks. Some assessments, particularly EF testing, were found too difficult for participants with more severe forms of ID.

Despite limitations such as a small sample size and the sole reliance on self-report and log data, the study highlights critical considerations for future large-scale RCTs in ID research. Recommendations include simplifying intervention protocols, using CAU as a comparison, tailoring RCT designs and materials to participant abilities, and improving communication through designated project teams at each facility. The findings emphasize the importance of feasibility testing to optimize both interventions and research methodologies for vulnerable populations.

Part 2: COVID study

Chapter 4

The onset of COVID-19 in 2020 necessitated the postponement of the intended cluster-RCT, leading to the implementation of an additional pilot study, which is presented in chapter 4. The COVID-19 pandemic had a particularly severe impact on vulnerable populations, such as people with ID. Face-to-face music interventions were disrupted by restrictions due to social distancing, prompting the need for alternative approaches, such as online interventions. This pilot study evaluated the feasibility, experiences, and short-term outcomes of a partially online music intervention for adults with ID during the COVID-19 pandemic. Ten participants received 16 individual music sessions over 10 weeks, delivered either face-to-face or online. The decision about the format was made collaboratively by the participants and their assigned music workers before each session. Using a concurrent mixed-methods design feasibility, participant and provider experiences, and changes in CB and well-being were assessed.

Key findings of this research showed that the intervention was generally feasible, although technical issues, caregiver availability, and individual impairments (e.g., poor vision) limited online participation. Participants and music workers reported high levels of appreciation, emotional engagement, and improved musical ability, with sessions perceived as relaxing and enjoyable. Personalisation of the music sessions and strong interpersonal bonds between participants and music workers were vital to the intervention's success.

However, online sessions were mostly rated negatively and viewed as impersonal, difficult to manage, and technically problematic. Face-to-face sessions were consistently preferred. Adjustments to tailor an online music intervention include beginning with face-to-face sessions, adding shorter online sessions and using adequate digital platforms.

Quantitative results indicated a reduction in CB, even among those receiving more online sessions. No significant change in well-being was observed, likely due to the already high baseline levels and due to limitations of the assessment tool that was used.

Part 3: Cluster-RCT

Chapter 5

Chapter 5 presents the design and rationale for the cluster-RCT assessing the effectiveness of an individual active music intervention for adults with mild to moderate ID showing CB. Based on the findings in the aforementioned studies, the design of the cluster-RCT was adjusted to reduce the demand and reliance on caregivers by using more appropriate assessment methods and questionnaires, a higher intensity of music sessions and applying individual music sessions, replacing the active control group with a CAU control group and providing music sessions by a broad range of music enthusiasts (music workers) rather than only relying on music educators ('muziekagoog' in Dutch).

The primary objective of the study was to assess the effect of the music sessions on CB, well-being, depression, anxiety, self-esteem and EF. A secondary goal was to evaluate the implementation and impact of the intervention based on the experiences of the participants with ID, the caregivers, and the music workers. Participants were randomly assigned, at the facility level, to the intervention group or the control group. The intervention consisted of 16 individual one-hour music sessions delivered twice weekly over a period of 8 to 10 weeks by one of the music workers. Each session followed a structured yet flexible format, allowing for personalization based on participant preferences and needs. Sessions involved active music-making, including singing, playing instruments, and engaging in exercises designed to target emotional expression and elements of EF. Participants in the control group continued with their usual care but were offered a music-related budget after the study would be ended to ensure equity and engagement.

The study aimed to include 96 participants, based on the results of a power analysis. Data collection included both quantitative and qualitative components. Quantitative measures assessed CB, well-being, depression, anxiety, self-esteem, and four EF domains using caregiver reports, self-report questionnaires, and computer-based tasks adapted for individuals with ID. Qualitative data from interviews, session logs, and evaluations provided further insight into the acceptability, feasibility, and perceived impact of the intervention.

The study design incorporated a strong methodological framework, including cluster randomization, pre- and post-intervention assessments, an 8-week follow-up, and a comprehensive process evaluation based on established frameworks. Analyses included both intention-to-treat and per-protocol approaches, with linear mixed models used to assess changes over time. Ultimately, this study aimed to contribute to the development of evidence-based practices in ID care by evaluating whether individual active music participation could serve as a meaningful, accessible, and effective intervention for reducing CB and enhancing psychological and cognitive well-being in people with ID and CB.

Chapter 6

In chapter 6 the outcomes of the cluster-RCT on CB, mental health and well-being in adults with mild or moderate ID and CB are presented. The intervention consisted of 16 individual sessions over 8 to 10 weeks, conducted twice weekly by a music worker. Participants engaged in active music-making, including playing various instruments and singing, with the sessions tailored to their preferences and abilities. The primary aim of the study was to evaluate the intervention's effects on CB, well-being, anxiety, depression, and self-esteem. Secondary aims included exploring whether treatment effects were influenced by baseline levels of CB or level of ID, and whether additional music sessions yielded greater benefits.

A total of 97 participants were recruited, 44 participating in the intervention group and 53 in the control group. Outcome measures included caregiver-rated and self-reported assessments of CB (using the Aberrant Behaviour Checklist), well-being (using the Personal Well-being Index – ID), anxiety and depression (via the Anxiety, Depression, and Mood Scale), and self-esteem (via the subscale Global Self-Worth of the Self-Perception Profile for Adolescents). In addition, participants were asked how they felt before and after each music session. Data were collected at baseline, post-intervention, and eight weeks after the intervention. Analyses were conducted using linear mixed models, with both intention-to-treat and per-protocol approaches.

The results demonstrated that individuals participating in the music intervention showed significantly greater reductions in total CB than those participating in the control group (CAU group), both immediately after the intervention and at follow-up. These effects extended to both internalizing and externalizing subtypes of CB. Moreover, significant reductions in anxiety and depression scores were observed at follow-up in individuals participating in the music intervention, with these effects more pronounced among participants with higher CB at baseline. No overall effects were found on well-being or self-esteem.

Self-reported emotional states also improved: participants reported significantly more pleasant feelings after music sessions compared to before, and those who already felt happy reported an increase in the intensity of their positive feelings. However, increasing the number of music sessions beyond the standard 16 did not result in additional significant benefits, suggesting a potential saturation point for intervention frequency.

The study offers several strengths, including a well-powered RCT design, a multi-site implementation, and the use of both proxy and self-report measures. The intervention was designed to be feasible and scalable, relying on music workers rather than licensed music therapists, which increases its applicability in real-world settings. Nonetheless, limitations include the inability to blind participants or researchers, the lack of an active control condition, and challenges related to the measurement tools' sensitivity, particularly self-report scales in participants with moderate ID.

Chapter 7

Executive functioning, including attention, working memory, inhibition, and cognitive flexibility, is essential for daily living and are often impaired in individuals with ID. In chapter 7 the impact of an individual music intervention on EF compared to CAU and the feasibility of EF measurements for adults with ID are presented.

Executive functioning was measured using three tablet-based games designed for children and adolescents with mild ID, covering cognitive flexibility, attention, inhibition, and working memory. Research assistants noted participant motivation and distraction during each test, as well as deviations from the test administration protocol in a log report. A principal component analysis (PCA) was performed to combine variables into one measure, and inter-group changes over time were analysed using linear mixed models. Feasibility was assessed with inductive thematic analysis.

The findings showed no significant differences in EF between the group of participants engaged in music and those in the CAU group at either post-intervention or follow-up. This was true regardless of the participant's baseline level of EF, level of ID, or level of participation in the intervention. A principal component analysis on the EF measures revealed that they primarily reflected a single factor, best interpreted as focused attention. Despite efforts to use EF tests previously validated in related populations, many participants struggled with the assessments. Common issues included difficulty understanding instructions, maintaining attention, using the tablet correctly, and staying motivated. These challenges resulted in a high proportion of invalid data, particularly among participants with moderate ID.

Results of the study showed that the music intervention, though carefully designed and implemented, may not have been sufficiently intensive, long, or targeted to EF to produce measurable change. It also underscores the difficulty of accurately measuring EF in this group. Future research should consider using a broader range of EF measures, including proxy reports and adaptive assessments, to capture functioning more reliably. It may also be necessary to develop longer or more targeted interventions that are specifically designed to stimulate EF domains. Additionally, the cognitive diversity within the ID population, as well as factors such as age, motor skills, and potential comorbidities, must be taken into account when designing both interventions and assessment tools.

Chapter 8

Reporting the practical experiences of conducting RCTs is as important as reporting outcomes. Chapter 8 presents a process evaluation of the cluster-RCT. This evaluation focused on the process of recruitment and participation in the RCT, the implementation of the intervention, the experiences of clients and professionals participating in the music intervention and the presumed sustainability of the intervention. Data were collected using evaluation forms, follow-up questionnaires, semi-structured interviews, log reports, and recruitment data. Participants, caregivers and music workers were involved in this process evaluation. Qualitative data were analysed using a systematic coding process to ensure consistency and depth of understanding.

Participants were recruited through staff of a Dutch care organization, though initial recruitment was slow and required additional efforts. The study eventually reached its targeted sample size, but inclusion of some participants with low levels of CB or dementia suggests limitations in the process of screening. Retention was high, with over 91% response rates for self- and proxy reports, despite some missing data due to scheduling delays and staff turnover. The intervention was performed largely according to protocol, with participants receiving nearly the full intended amount of music sessions. Minor adaptations were common and tailored to participants' preferences and needs, especially for younger individuals and those with mild ID. Most participants completed all sessions and reported high enjoyment, with caregivers and music workers observing improvements in mood, behaviour, emotional regulation, and self-esteem. However, these effects were often short-lived. More than half of the participants engaged in the music intervention continued music activities post-intervention, but sustainability was hindered by lack of caregiver time, support, and clarity about their role.

This process evaluation showed that individual active music interventions for people with ID are feasible, well-received, and capable of delivering short-term benefits in behaviour and emotional well-being. Recruitment was challenging; however, the implementation of revised recruitment strategies ultimately resulted in sufficient participant enrolment. High retention and adherence were supported by one-on-one delivery and by a familiar setting. Tailoring the intervention enhanced engagement but occasionally compromised fidelity. The interpersonal connection between participant and music worker appeared to represent a key facilitating factor. While post-intervention music-making continued for some, sustainability requires clearer caregiver guidance, reduced workload, and stronger team collaboration. To maintain long-term benefits, organizational structures must better support caregivers in facilitating ongoing musical engagement.

General discussion

Chapter 9

Chapter 9 presents a general discussion of the findings from this thesis. It begins with a summary of the key findings, followed by a reflection on their significance. Next, the studies' strengths and limitations, implications for clinical practice, and suggestions for future research are discussed.

Reflecting on the results of this thesis, four key findings emerge. First, the research presented in this thesis demonstrated that an individual active music intervention is an effective and sustainable way to improve CB in adults with mild or moderate ID, even when the intervention is partially online. The intervention not only positively affected the individuals with ID, but also showed promising results on the well-being of their caregivers. Additionally, it was found that music interventions could help reduce anxiety and depression, thereby improving emotional well-being and mental health for adults with ID. Second, it remains unclear whether music is more beneficial than other enjoyable activities. While making music can be an effective intervention, it may not outperform other engaging activities. Thus, continued exploration of various individualized, evidence-based approaches to address and prevent CB in adults with ID is essential. Third, the participants responded positively to the music intervention, which was well-received and effectively engaged adults with ID. The intervention was also feasible to implement, achieving high dose and fidelity scores. Finally, the research presented in this thesis identified four key characteristics that appear necessary for a music intervention to be effective. First, individual music sessions are preferred over group sessions. Second, the duration, intensity, and frequency of the sessions are crucial to consider. Third, face-to-face sessions are preferred to online sessions. Fourth, the intervention must be well-structured with clear objectives that align with behavior improvement goals.

The studies incorporated in this thesis have several strengths. They explore the effects of a music intervention for adults with ID, utilizing multiple research designs (cluster-RCT, feasibility study, process evaluation) and data collection methods (questionnaires, tests, interviews). They offer valuable insights into different intervention formats and highlight the importance of a structured intervention manual. Despite these strengths, the included studies in this thesis also have limitations, including targeting not all individuals with ID and CB (only those with mild to moderate ID), inclusion of only participants with a positive interest in music, lack of blinding in the studies, and uncertainty regarding the specific active components of the intervention. Additionally, assessing EF in individuals with ID proved to be challenging, especially for those with moderate ID. Overall, the research presented in this thesis underscores the value of rigorous methodologies and contributes to the field of evidence-based interventions for adults with ID, particularly in the promising area of music interventions.

Important implications for clinical practice, especially for caregivers of adults with ID, include the need for structured music interventions to improve emotional regulation and CB. The findings suggest that caregivers should be trained to present these interventions in a consistent, evidence-based manner, with proper supervision and support from professional music workers. Additionally, caregivers need more resources and enough time to integrate music interventions into daily care routines. The research also emphasizes the importance of equipping caregivers with knowledge about research, particularly RCTs, to build a robust evidence base for ID interventions. Furthermore, it is recommended to adapt music interventions to the specific needs of individuals with CB, anxiety, depression, or emotion regulation issues to maximize effectiveness.

Based on the results presented in this thesis, several directions for future research in ID care can be suggested. Future studies could investigate the differential effect of music interventions compared to leisure activities interventions, considering factors like age and level of ID. Research designs such as n-of-1 trials could help address logistical challenges while evaluating individualized interventions. It is also recommended to use direct observations or video analysis of music sessions to better understand the dynamics and to identify the most active components of the intervention. Additionally, integrating physiological measures could provide more objective data on the intervention's effects. Future research should also explore both quantitative and qualitative outcomes, such as self-esteem and well-being, using detailed case reports. In this thesis the importance of integrating music interventions into daily care routines is emphasized and caregivers

should be supported to effectively incorporate these interventions, prioritizing music as a valuable tool to improve the well-being of people with ID.

APPENDICES



Samenvatting

Algemene inleiding

Hoofdstuk 1

Hoofdstuk 1 biedt een overzicht van de relevante literatuur om beter inzicht te krijgen in de uitdagingen waarmee mensen met een verstandelijke beperking (VB) te maken hebben en verkent de potentie van muziekinterventies als strategie om deze uitdagingen te verlichten.

In Nederland heeft circa 1,45% van de bevolking een VB, waarvan 18,1% tevens moeilijk verstaanbaar gedrag (MVG) vertoont. Moeilijk verstaanbaar gedrag wordt gedefinieerd als gedrag dat schadelijk of belastend is voor het individu zelf en/of diens omgeving. Dit gedrag komt vaker voor bij mensen met een VB dan in de algemene bevolking en wordt in verband gebracht met factoren zoals trauma, beperkte communicatieve vaardigheden en sociale uitsluiting. MVG heeft een negatieve invloed op de kwaliteit van leven (KvL) van zowel de persoon zelf als van diens begeleiders, en draagt bij aan verhoogde stress en burn-out onder zorgverleners. Mensen met een VB hebben daarnaast vaak verminderde executieve functies (EF), waaronder inhibitie, cognitieve flexibiliteit, werkgeheugen en aandacht. Deze functies worden verder beperkt wanneer er tevens sprake is van MVG. Daarnaast hebben mensen met een VB een grotere kans op psychische problemen, sociale isolatie, chronische stress en een laag zelfbeeld, factoren die hun KvL en psychisch welzijn verder verminderen.

Een veelbelovende strategie om deze problemen te verminderen is een muziekinterventie, waarvan is aangetoond dat deze sociale interactie bevordert, de mentale gezondheid verbetert en stress vermindert. Bestaand onderzoek bij zowel de algemene bevolking als bij klinische doelgroepen (zoals mensen met dementie of een autismespectrumstoornis of kinderen) bevestigt de positieve effecten van muziekinterventies op welzijn, stemming, EF, MVG en sociaal functioneren. Bij mensen met een VB, met name bij een lichte tot matige VB, tonen niet-experimentele studies positieve verbanden met een verbeterde KvL, stemming, sociale betrokkenheid en een vermindering van MVG. Experimenteel onderzoek is echter schaars, doorgaans kleinschalig en methodologisch beperkt (bijvoorbeeld door het ontbreken van controlegroepen of korte follow-upperioden).

Ondanks deze beperkingen laten de onderzoeken tot nu toe veelbelovende resultaten zien betreffende muziekinterventies voor mensen met een VB. Om de ontbrekende kennis aan te vullen, onderzoeken de studies in dit proefschrift de effecten en haalbaarheid van actieve muziekinterventies bij volwassenen met een VB die in residentiële voorzieningen in Nederland wonen. Met een combinatie van verschillende onderzoeksmethoden wordt het effect van fysieke en

digitale muziekinterventies op individueel en op groepsniveau onderzocht op MVG, EF, mentale gezondheid, zelfvertrouwen en welzijn. Het proefschrift is daarom opgebouwd uit drie delen, die elk een specifieke onderzoeksvraag behandelen:

Deel 1: Pilot cluster-RCT

Wat zijn de effecten en de haalbaarheid van een wekelijkse muziekinterventie in groepsverband vergeleken met recreatieve groepsactiviteiten (hoofdstukken 2 en 3)?

Deel 2: COVID-studie

Wat zijn de haalbaarheid, ervaringen en kortetermijneffecten van een gedeeltelijk digitale muziekinterventie die tweemaal per week plaatsvindt (hoofdstuk 4)?

Deel 3: Cluster-RCT

Wat zijn de effecten en de haalbaarheid van een individuele muziekinterventie die tweemaal per week plaatsvindt, vergeleken met standaardzorg (hoofdstukken 5, 6, 7 en 8)?

<u>Deel 1: Pilot cluster-RCT</u>

Hoofdstuk 2

Hoofdstuk 2 presenteert een pilotstudie waarin de effecten werden onderzocht van twee interventies: actieve muziekparticipatie en recreatieve activiteiten, bij volwassenen met een VB. Recreatieve activiteiten worden bij mensen met een VB geassocieerd met verbeterd welzijn, sterkere sociale vaardigheden en minder emotionele en gedragsproblemen. Ook actieve muziekparticipatie lijkt het welzijn en de sociale acceptatie te bevorderen en MVG te verminderen. Deze studie vergeleek de effectiviteit van wekelijkse, door begeleiding ondersteunde, groepssessies met actieve muziekparticipatie versus recreatieve activiteiten. Metingen werden gedaan voor en na de interventie en na een follow-upperiode. Uitkomstmaten waren welzijn, zelfbeeld, sociale acceptatie, MVG en drie componenten van EF (inhibitie, cognitieve flexibiliteit en visueel werkgeheugen).

Aan deze pilot cluster-RCT namen 29 volwassenen deel met een lichte VB, matige VB of functionerend op zwakbegaafd niveau, verdeeld over vier woonvoorzieningen. De muziekinterventie werd geleid door muziekbegeleiders die een vaste routine volgden om consistentie en reproduceerbaarheid te waarborgen. De sessies bestonden uit een voorbereiding met een coördinerende begeleider, een vaste volgorde van muzikale activiteiten en actieve deelname van deelnemers en begeleiders. De interventie met recreatieve activiteiten bestond uit interactieve niet-

muzikale activiteiten (zoals spelletjes, darten, kaarten, bakken en puzzelen) en was qua groepsinteractie en aandacht vergelijkbaar met de muziekinterventie.

Hoewel binnen beide groepen verbeteringen werden gezien, bleek de muziekinterventie niet effectiever dan de interventie met recreatieve activiteiten. Opmerkelijk was dat deelnemers aan de recreatieve activiteiten, en vooral jongere personen met een lichte VB, een grotere afname in internaliserend gedrag en verbeteringen in inhibitie lieten zien na de interventie dan deelnemers aan de muziekactiviteiten. De deelnemers in de muziekgroep rapporteerden een beter zelfbeeld en minder frustratie van behoeften, maar deze effecten waren niet significant sterker dan bij de deelnemers in de andere groep.

De resultaten laten zien dat recreatieve activiteiten effectief kunnen zijn in het verbeteren van bepaalde aspecten van welzijn en EF, vooral bij jongere deelnemers met een lichte VB. Het is mogelijk dat de muziekinterventie pas significante effecten oplevert wanneer deze langer duurt, een hogere frequentie heeft of beter is afgestemd op de deelnemer. Methodologische beperkingen waren onder andere de kleine steekproefomvang, meetproblemen bij deelnemers met een matige VB en heterogeniteit in de interventies. Toekomstig onderzoek zou zich moeten richten op langduriger, meer op maat gemaakte interventies en het gebruik van meer inclusieve en geschikte meetinstrumenten om de uitkomsten bij deze diverse populatie beter in kaart te brengen.

Hoofdstuk 3

Het uitvoeren van pilotstudies en haalbaarheidsonderzoek is een waardevolle strategie gebleken ter voorbereiding op de implementatie van grootschalige RCT's. Hoofdstuk 3 beschrijft de haalbaarheid van de pilot cluster-RCT waarin een groepsinterventie met muziek werd vergeleken met een groepsinterventie met recreatieve activiteiten bij volwassenen met een VB. Deze studie richtte zich op zes kernaspecten van haalbaarheid: acceptatie, behoefte, implementatie, praktische uitvoerbaarheid, aanpassing en beperkte effectiviteit van de pilot. De ervaringen en perspectieven van zowel de ontvangers van (één van) de interventies (volwassenen met een VB) als van degenen die de interventies uitvoerden (groepsbegeleiders, muziekbegeleiders en een onderzoeksassistent) werden in kaart gebracht. Er werd een mixed-methods benadering gehanteerd, waarbij logboeken, evaluatievragenlijsten en RCT-uitkomstmaten (vragenlijsten via begeleiding, zelfrapportages en EF testen) werden geanalyseerd.

De muziekinterventie werd over het algemeen positief ontvangen door de deelnemers. Zij rapporteerden plezier, emotionele betrokkenheid en een gevoel van verbondenheid met elkaar.

Groepsbegeleiders en muziekbegeleiders zagen enkele positieve effecten, hoewel de betrokkenheid van de groepsbegeleiders wisselend was en de huiswerktaken vaak als te belastend werden ervaren. De uitvoering werd bemoeilijkt door logistieke uitdagingen en verschillen in cognitief niveau tussen de deelnemers. De recreatieve activiteiten bleken daarentegen minder stimulerend en boden weinig toegevoegde waarde. Zowel deelnemers als groepsbegeleiders uitten hun ontevredenheid en noemden een gebrek aan variatie en relevantie. Het RCT-ontwerp bleek te belastend voor zowel deelnemers als groepsbegeleiders. Groepsbegeleiders hadden moeite met de werklast, onduidelijke communicatie en er was een lage motivatie om onderzoekstaken uit te voeren. Sommige meetinstrumenten, met name de EF-testen, bleken te moeilijk voor deelnemers met een lagere VB.

Ondanks beperkingen zoals een kleine steekproefomvang en het gebruik van zelfrapportagelijsten en logboeken, benadrukt deze studie enkele cruciale aandachtspunten voor toekomstige grootschalige RCT's binnen het VB-onderzoek. Aanbevolen wordt om interventieprotocollen te vereenvoudigen, standaardzorg als controleconditie te gebruiken, studiedesign en -materialen beter af te stemmen op de doelgroep, en de communicatie te verbeteren via een projectteam per locatie. De bevindingen onderstrepen het belang van haalbaarheidsonderzoek om zowel interventies als onderzoeksmethoden te optimaliseren voor kwetsbare doelgroepen.

Deel 2: COVID-studie

Hoofdstuk 4

Door de COVID-19 pandemie in 2020 werd de geplande cluster-RCT uitgesteld en werd een nieuwe pilotstudie uitgevoerd (hoofdstuk 4). De COVID-19-pandemie had een bijzonder ernstige impact op kwetsbare groepen, zoals mensen met een VB. Fysieke muziekinterventies vielen stil door sociale restricties en andere maatregelen, waardoor alternatieve vormen, zoals online interventies, noodzakelijk waren. Deze pilotstudie onderzocht de haalbaarheid, ervaringen en kortetermijneffecten van een deels digitale muziekinterventie voor volwassenen met een VB tijdens de COVID-19-pandemie. Tien deelnemers kregen 16 individuele muzieksessies verspreid over 10 weken, fysiek of online. Het format werd per sessie gezamenlijk bepaald door de deelnemer en muziekbegeleider. Een mixed-methods onderzoeksdesign evalueerde de haalbaarheid, ervaringen van deelnemers en muziekbegeleiders, en veranderingen in MVG en welzijn.

De interventie was over het algemeen haalbaar, hoewel technische problemen, geringe beschikbaarheid van begeleiding en individuele beperkingen (zoals slecht zicht) deelname aan online sessies bemoeilijkten. Deelnemers en muziekbegeleiders rapporteerden een hoge mate van

waardering, emotionele betrokkenheid en verbeterde muzikale vaardigheden. De sessies werden als rustgevend en plezierig ervaren. Personalisatie van de muzieksessies en een sterke band tussen deelnemer en muziekbegeleider waren essentieel voor het succes van de interventie.

De online sessies werden overwegend negatief beoordeeld: onpersoonlijk, lastig uitvoerbaar en technisch problematisch. Fysieke sessies kregen consequent de voorkeur. Online muziekinterventies kunnen beter worden afgestemd op de doelgroep door te starten met fysieke sessies, kortere online sessies aan te bieden en geschikte digitale platforms te gebruiken.

De kwantitatieve resultaten toonden een afname in MVG, zelfs bij deelnemers die meer online sessies volgden. Er werd geen significante verandering in welzijn gevonden, waarschijnlijk door een hoog baselineniveau en beperkingen van het meetinstrument.

Deel 3: Cluster-RCT

Hoofdstuk 5

In hoofdstuk 5 komt het ontwerp van en de rationale voor de cluster-RCT aan bod. De cluster-RCT onderzoekt de effectiviteit van een individuele actieve muziekinterventie voor volwassenen met een lichte tot matige VB en MVG. Het onderzoeksdesign werd op basis van bevindingen uit de eerder beschreven studies aangepast om de belasting voor en afhankelijkheid van begeleiding te verminderen. Dit werd bereikt door geschiktere tests en vragenlijsten te gebruiken, een intensievere individuele muziekinterventie toe te passen, standaardzorg als controlegroep in te zetten en samen te werken met diverse muziekbegeleiders in plaats van alleen muziekagogen.

Het primaire doel van deze studie was het beoordelen van het effect van de muzieksessies op MVG, welzijn, depressie, angst, zelfbeeld en EF. Een secundair doel was het evalueren van de implementatie en impact van de interventie, met input van deelnemers, groepsbegeleiders en muziekbegeleiders. Deelnemers werden op locatieniveau willekeurig toegewezen aan de interventiegroep of de controlegroep. De interventie bestond uit 16 individuele muzieksessies van één uur, tweemaal per week, gedurende een periode van 8 tot 10 weken, uitgevoerd door één van de muziekbegeleiders. Elke sessie had een vaste structuur met voldoende flexibiliteit om aan voorkeuren en behoeften van de deelnemer aan te passen. Sessies bestonden uit actieve muziekparticipatie, zoals zingen, instrumenten bespelen en oefeningen gericht op emotionele expressie en componenten van EF. Deelnemers in de controlegroep ontvingen standaardzorg, maar

kregen na afloop van het onderzoek een bepaald budget om te besteden aan muzikale activiteiten, als compensatie en om betrokkenheid te waarborgen.

De studie had als doel om 96 deelnemers te includeren, op basis van de resultaten van een poweranalyse. Dataverzameling was zowel kwantitatief als kwalitatief. Kwantitatieve metingen richtten zich op MVG, welzijn, depressie, angst, zelfbeeld en vier componenten van EF, via vragenlijsten voor begeleiding, zelfrapportages en computertaken aangepast voor mensen met een VB. Kwalitatieve gegevens werden verzameld via interviews, logboeken van de muzieksessies en evaluaties, om meer inzicht te krijgen in de acceptatie, haalbaarheid en impact van de interventie.

Het onderzoeksdesign kende een sterke methodologische opzet, met cluster-randomisatie, metingen vóór en na de interventie, een follow-up na 8 weken, en een procesevaluatie op basis van bestaande kaders. Analyses, zowel op basis van intention-to-treat- als per-protocolbenaderingen, werden uitgevoerd met behulp van lineaire mixed models om veranderingen over de tijd te analyseren. Uiteindelijk beoogde deze studie bij te dragen aan de verdere ontwikkeling van evidence-based interventies in de zorg voor mensen met een VB. Dit werd onderzocht door na te gaan of individuele actieve muziekparticipatie een zinvolle, toegankelijke en effectieve interventie kan zijn voor het verminderen van MVG en het bevorderen van psychisch en cognitief welzijn bij mensen met een VB en MVG.

Hoofdstuk 6

Hoofdstuk 6 beschrijft de resultaten van de cluster-RCT betreffende MVG, mentale gezondheid en welzijn bij volwassenen met een lichte tot matige VB en MVG. De interventie bestond uit 16 individuele sessies in 8 tot 10 weken, twee keer per week, uitgevoerd door muziekbegeleiders. Deelnemers deden actief mee aan muziekactiviteiten, waaronder het bespelen van diverse instrumenten en zingen, waarbij de sessies waren afgestemd op hun voorkeuren en mogelijkheden.

Het primaire doel van de studie was het effect van de muzieksessies te beoordelen op MVG, welzijn, angst, depressie en zelfbeeld. Secundaire doelen waren de invloeden van baseline-levels van MVG en het VB-niveau op de behandeleffecten en of aanvullende muzieksessies extra voordelen zouden opleveren.

In totaal werden 97 deelnemers geïncludeerd: 44 in de interventiegroep en 53 in de controlegroep. De uitkomstmaten waren MVG (gemeten met de Afwijkende Gedrag Schaal), welzijn (gemeten met de Persoonlijk Welzijn Index-VB), angst en depressie (gemeten met Angst, Depressie en Stemming

Schaal) en zelfbeeld (gemeten met de subschaal Gevoel van Eigenwaarde van de Competentiebelevingsschaal voor adolescenten). Ook werd de deelnemers gevraagd hoe ze zich voelden voorafgaand en na afloop van elke muzieksessie. Gegevens werden verzameld bij baseline, direct na de interventie en acht weken later als follow-up. Analyses werden uitgevoerd met lineaire mixed models volgens zowel intention-to-treat als per-protocol benaderingen.

De resultaten toonden aan dat de deelnemers in de muziekinterventiegroep een significant sterkere afname in MVG lieten zien dan de deelnemers in de controlegroep, zowel direct na de interventie als bij follow-up. Deze effecten waren zichtbaar voor zowel internaliserend als externaliserend gedrag. Daarnaast werd bij follow-up een significante afname in angst en depressie geconstateerd bij de deelnemers aan de muziekinterventie, met name bij degenen met een hogere baseline-MVG. Er werden geen significante effecten gevonden voor welzijn of zelfbeeld.

De zelf-gerapporteerde gevoelens verbeterden eveneens: deelnemers rapporteerden significant meer plezierige gevoelens na een muzieksessie in vergelijking met ervoor, en deelnemers die zich al gelukkig voelden, meldden een toename in de intensiteit van deze positieve gevoelens. Meer dan 16 sessies leverde geen extra significante voordelen, wat wijst op een mogelijk verzadigingspunt.

De studie kent verschillende sterke punten: het ontwerp was een goed uitgeruste RCT, uitgevoerd op meerdere locaties en met gebruik van zowel proxy- als zelfrapportage-instrumenten. De interventie was ontworpen om haalbaar en opschaalbaar te zijn, doordat gebruik werd gemaakt van muziekbegeleiders in plaats van muziektherapeuten. Desalniettemin zijn er beperkingen, waaronder het ontbreken van blindering, geen actieve controlegroep en de beperkte gevoeligheid van de meetinstrumenten, vooral de zelfrapportages bij deelnemers met een matige VB.

Hoofdstuk 7

Executieve functies, waaronder aandacht, werkgeheugen, inhibitie en cognitieve flexibiliteit, zijn essentieel voor het dagelijks functioneren en blijken vaak beperkt te zijn bij mensen met een VB. In hoofdstuk 7 wordt het effect van een individuele muziekinterventie op EF vergeleken met standaardzorg, en wordt de haalbaarheid van EF-metingen bij volwassenen met een VB onderzocht.

Executieve functies werden gemeten met drie tabletspellen ontwikkeld voor kinderen en adolescenten met een lichte VB. Deze spellen waren gericht op cognitieve flexibiliteit, aandacht, inhibitie en werkgeheugen. Onderzoeksassistenten noteerden motivatie, concentratie en eventuele afwijkingen van het testprotocol in logboeken. Een principal component analysis (PCA) werd uitgevoerd om de meetgegevens zo mogelijk tot één factor te herleiden. Vervolgens werd met

lineaire mixed models geanalyseerd of er veranderingen optraden tussen de groepen. De haalbaarheid van de metingen werd geëvalueerd met behulp van inductieve thematische analyse.

De resultaten toonden aan dat er geen significante verschillen in EF waren tussen de deelnemers aan de muziekinterventie en de deelnemers die standaardzorg ontvingen, noch direct na de interventie, noch bij de follow-up. Dit uitblijvende resultaat werd vastgesteld ongeacht het baselineniveau van EF, het VB-niveau of de intensiteit van interventiedeelname. De PCA toonde aan dat de EF-metingen voornamelijk één factor representeerden, die gerichte aandacht representeerde. Ondanks het gebruik van EF-tests die eerder gevalideerd waren in verwante doelgroepen, hadden veel deelnemers moeite met het uitvoeren van de taken. Vaak bleek het lastig om instructies te begrijpen, de aandacht erbij te houden, de tablet goed te bedienen of gemotiveerd te blijven. Hierdoor was er een grote hoeveelheid ongeldige data, met name bij deelnemers met een matige VB.

De resultaten van deze studie tonen aan dat de muziekinterventie, ondanks de zorgvuldige opzet en uitvoering, mogelijk niet voldoende intensief, langdurig of genoeg toegespitst was op EF om tot meetbare effecten te leiden. Tegelijkertijd onderstrepen de resultaten hoe moeilijk het is om EF op een valide manier te meten bij deze doelgroep. Voor toekomstig onderzoek is het aan te raden om gebruik te maken van een breder scala aan meetinstrumenten, waaronder observaties door begeleiding of adaptieve tests, om het functioneren betrouwbaarder in kaart te brengen. Daarnaast zijn langere of EF-gerichte interventies en passende meetinstrumenten nodig, rekening houdend met cognitieve diversiteit, leeftijd, motorische vaardigheden en bijkomende problematiek.

Hoofdstuk 8

Het rapporteren van praktische ervaringen bij het uitvoeren van RCT's is minstens zo belangrijk als het beschrijven van de onderzoeksresultaten. Daarom wordt in hoofdstuk 8 een procesevaluatie gepresenteerd van de cluster-RCT. Deze evaluatie richtte zich op het wervings- en participatieproces, de uitvoering van de interventie, de ervaringen van cliënten en betrokken professionals bij de muzikale interventie, en de verwachte duurzaamheid ervan. De gegevens zijn verzameld via evaluatieformulieren, follow-upvragenlijsten, semigestructureerde interviews, logboeken en registraties van het wervingsproces. De procesevaluatie betrok zowel deelnemers als groepsbegeleiders en muziekbegeleiders. De kwalitatieve data werden systematisch gecodeerd om consistentie en diepgang te waarborgen.

Deelnemers werden geworven via medewerkers van een Nederlandse zorgorganisatie. De werving verliep aanvankelijk traag en vergde extra inspanningen. Hoewel de beoogde steekproefgrootte

uiteindelijk werd gehaald, bleek uit de deelname van enkele cliënten met weinig MVG of met dementie dat het screeningsproces niet altijd optimaal was. De respons was hoog: meer dan 91% van de zelfrapportages en proxyvragenlijsten werd ingevuld, ondanks enige uitval als gevolg van planningsproblemen en personeelswisselingen. De interventie werd grotendeels volgens protocol uitgevoerd. De meeste deelnemers ontvingen vrijwel alle geplande sessies. Kleine aanpassingen in de sessies kwamen regelmatig voor en waren afgestemd op individuele wensen en behoeften, vooral bij jongeren en deelnemers met een lichte VB. De overgrote meerderheid rondde het volledige programma af en gaf aan veel plezier te hebben beleefd. Zorgverleners en muziekbegeleiders rapporteerden verbeteringen in stemming, gedrag, emotieregulatie en zelfbeeld, al waren deze effecten vaak tijdelijk van aard. Meer dan de helft van de deelnemers aan de muziekinterventie bleef na afloop muziekactiviteiten voortzetten. Toch werd de structurele inzet van muziek binnen de dagelijkse zorg bemoeilijkt door een gebrek aan tijd, onvoldoende ondersteuning en onduidelijkheid over de rol van groepsbegeleiders.

De procesevaluatie liet zien dat individuele, actieve muziekinterventies bij mensen met een VB goed uitvoerbaar zijn, positief worden ontvangen, en op korte termijn gunstige effecten kunnen hebben op gedrag en emotioneel welzijn. De werving kende uitdagingen, maar het toepassen van nieuwe wervingsstrategieën hielp om voldoende deelname te bereiken. De hoge mate van betrokkenheid en deelname werd versterkt door het individuele format en een vertrouwde omgeving. Het individueel afstemmen van de interventie verhoogde de betrokkenheid, maar leidde soms tot afwijkingen van het oorspronkelijke protocol. De relatie tussen deelnemer en muziekbegeleider bleek van groot belang. Hoewel een aantal deelnemers na afloop doorging met muziekactiviteiten, is blijvende toepassing alleen mogelijk wanneer groepsbegeleiders beter worden begeleid, hun werkdruk vermindert en de samenwerking binnen het team wordt versterkt. Hiervoor is een organisatorische structuur nodig om groepsbegeleiders actief te ondersteunen bij het continueren van muziekactiviteiten.

Algemene discussie

Hoofdstuk 9

Hoofdstuk 9 bevat een algemene discussie over de bevindingen uit dit proefschrift. Het begint met een samenvatting van de belangrijkste resultaten, gevolgd door een beschouwing wat deze resultaten betekenen. Vervolgens worden de sterke punten en beperkingen van de studies, implicaties voor de praktijk en aanbevelingen voor toekomstig onderzoek behandeld.

Op basis van de uitkomsten van dit proefschrift, komen vier belangrijke inzichten naar voren. Ten eerste tonen de studies in dit proefschrift aan dat een individuele actieve muziekinterventie een effectieve en duurzame manier kan zijn om MVG te verminderen bij volwassenen met een lichte tot matige VB, zelfs bij gedeeltelijke online uitvoering. De interventie was gunstig voor zowel de deelnemers als het welzijn van de begeleiding. Daarnaast bleek dat muziekinterventies angst en depressie kunnen verminderen, wat bijdraagt aan een beter emotioneel welzijn en mentale gezondheid bij deze doelgroep. Ten tweede blijft onduidelijk of muziek effectiever is dan andere recreatieve activiteiten. Muziek maken is effectief, maar niet noodzakelijk effectiever dan andere plezierige activiteiten. Dat onderstreept de noodzaak om verschillende, op maat gemaakte en onderbouwde benaderingen te blijven verkennen om MVG bij volwassenen met een VB te voorkomen en te behandelen. Ten derde werden de muziekinterventies over het algemeen positief ontvangen. Deelnemers reageerden enthousiast en betrokken, en de interventie was goed uitvoerbaar in de praktijk. Zowel de mate waarin deelnemers alle sessies volgden als de trouw aan het protocol waren hoog. Tot slot werden vier kenmerken geïdentificeerd die cruciaal lijken voor het succes van een muziekinterventie. Ten eerste geven deelnemers de voorkeur aan individuele sessies boven groepssessies. Ten tweede zijn de duur, intensiteit en frequentie van de sessies van groot belang. Ten derde blijken fysieke sessies effectiever dan online sessies. En tenslotte dient de interventie goed gestructureerd te zijn, met duidelijke doelstellingen gericht op gedragsverbetering.

De studies in dit proefschrift hebben verschillende sterke punten. Zo worden de effecten van muziekinterventies bij volwassenen met een VB onderzocht en wordt hierbij gebruik gemaakt van verschillende onderzoeksopzetten, zoals een cluster-RCT, een haalbaarheidsstudie en een procesevaluatie, evenals diverse methoden om data te verzamelen (vragenlijsten, tests, interviews). Daarnaast bieden de studies belangrijke inzichten in verschillende interventievormen en onderstrepen ze het belang van een duidelijk protocol. Er zijn echter ook beperkingen: de studies richtten zich enkel op mensen met een lichte tot matige VB; alleen deelnemers met positieve belangstelling voor muziek werden geselecteerd; blindering was niet mogelijk; en het is onduidelijk welke onderdelen van de interventie cruciaal waren. Ook bleek het meten van EF lastig, vooral bij deelnemers met een matige VB. Samengevat onderstrepen de studies in dit proefschrift het belang van een gedegen methodologische aanpak en dragen ze bij aan evidence-based interventies voor volwassenen met een VB, met name op het gebied van muziekinterventies.

Voor de dagelijkse begeleidingspraktijk wordt aanbevolen een structurele muziekinterventie in te zetten om emotieregulatie en MVG te verbeteren. Groepsbegeleiders zouden gericht opgeleid moeten worden om deze interventies op een consequente en onderbouwde manier uit te voeren,

met de juiste begeleiding van professionele muziekbegeleiders. Daarnaast hebben zij meer middelen en tijd nodig om muziekactiviteiten duurzaam in de dagelijkse zorg te integreren. De studies in dit proefschrift laten ook zien hoe belangrijk het is dat groepsbegeleiders kennis opdoen over RCT-onderzoek, zodat er meer evidence-based interventies binnen de VB-zorg tot stand kunnen komen. Bovendien wordt geadviseerd om muziekinterventies aan te passen aan de specifieke behoeften van mensen met een VB die mogelijk te maken hebben met MVG, angst, depressie of emotieregulatieproblemen, om de effectiviteit te maximaliseren.

Op basis van de gepresenteerde resultaten in dit proefschrift worden verschillende richtingen voor toekomstig onderzoek binnen de zorg voor mensen met een VB voorgesteld. Zo wordt aanbevolen om het verschil in effect tussen muziekinterventies en andere recreatieve activiteiten verder te onderzoeken, waarbij rekening wordt gehouden met factoren zoals leeftijd en het niveau van de VB. Kleinschalige onderzoeksopzetten, zoals n=1-studies, kunnen helpen om beter in te spelen op praktische uitdagingen en bieden mogelijkheden om effecten op individueel niveau te evalueren. Daarnaast wordt het aanbevolen om observaties en video-analyses van muzieksessies in te zetten. Deze kunnen meer inzicht geven in de manier waarop muziek werkt en welke onderdelen van de interventie daadwerkelijk bijdragen aan het effect. Ook het toevoegen van fysiologische metingen kan waardevolle, objectieve informatie opleveren over hoe muziekinterventies invloed hebben op het lichaam en gedrag. Verder wordt aangeraden om in toekomstig onderzoek zowel kwantitatieve als kwalitatieve uitkomsten te onderzoeken, zoals veranderingen in zelfbeeld of welzijn, bijvoorbeeld in casestudies. Tot slot benadrukken de bevindingen in dit proefschrift het belang van structurele inbedding van muziekinterventies in de dagelijkse zorgpraktijk. Dit vraagt om goede ondersteuning van groepsbegeleiders, zodat zij deze interventies effectief kunnen toepassen. Op die manier kan muziek bijdragen aan het verbeteren van het welzijn van mensen met een VB.

APPENDICES



Makkelijk lezen samenvatting

Deze tekst gaat over een onderzoek.
Dit onderzoek is uitgevoerd bij Philadelphia.
En ging over muziek maken met mensen met een verstandelijke beperking.
Hieronder kun je lezen wat uit dit onderzoek is gekomen.

Begin

Sommige mensen hebben een verstandelijke beperking.
Zij hebben vaak elke dag hulp nodig.
Soms hebben ze moeilijk gedrag.
Moeilijk gedrag is lastig voor mensen met een verstandelijke beperking zelf.
En voor anderen.

Mensen met een verstandelijke beperking vinden onthouden vaak moeilijk.
En ze vinden plannen vaak moeilijk
Ze voelen zich ook vaker alleen of verdrietig.
Hun leven kan daardoor minder prettig zijn.

Muziek kan misschien helpen bij deze problemen.
Muziek maken kan rust geven en vrolijk maken.
Muziek is ook heel fijn om samen te doen.

De onderzoekers wilden meer weten over muziek.
Er is weinig onderzoek naar muziek bij mensen met een verstandelijke beperking.
Daarom deden de onderzoekers hier onderzoek naar.
Het onderzoek ging over muziek maken door volwassenen van Philadelphia.

Muziek maken is soms alleen.
En soms in een groep.
Soms wordt er op de locatie muziek gemaakt.

En soms gebeurt dit met een computer.

Online noemen we dat.

De onderzoekers keken of door muziek maken het moeilijk gedrag minder werd.

Ook keken de onderzoekers hoe tevreden mensen waren met het muziek maken.

En of ze minder angstig of somber werden door het muziek maken.

Het onderzoek bestond uit 3 delen:

- **Deel 1:** Onderzoek met groepen

  Muziek maken werd vergeleken met het doen van andere activiteiten.
- **Deel 2:** Onderzoek tijdens corona

  Muziek maken was soms op locatie.

  En soms online.
- **Deel 3:** Onderzoek met 1-op-1 muzieklessen

  Muziek maken werd vergeleken met gewone zorg.

Deel 1

Deel 1 gaat over 1 klein onderzoek.

Bijna 30 volwassenen van Philadelphia deden mee aan de studie.

1 groep deelnemers maakte muziek.

1 andere groep deelnemers deed activiteiten.

Zoals spelletjes, bakken of puzzelen.

Alle deelnemers kregen hulp van begeleiders.

De deelnemers maakten 1 keer per week muziek.

Of deden activiteiten.

Na 16 keer stopten de deelnemers met muziek maken.

Of met activiteiten doen.

De onderzoekers keken wat er veranderde door de muziek.

En de onderzoekers keken wat er veranderde door de activiteiten.

Het ging beter met alle deelnemers.
Het ging goed met de mensen die muziek maakten.
Maar het ging ook goed met de mensen die activiteiten deden.
Muziek maken en activiteiten doen maakten de deelnemers rustiger.
Ook konden de deelnemers zich beter beheersen.

De onderzoekers vroegen zich af hoe dit kon.
Misschien werkt muziek beter wanneer je langer oefent.
Misschien is 1 keer per week muziek maken te weinig.

De onderzoekers luisterden ook naar de deelnemers en begeleiders.
En vroegen hun wat ze van het onderzoek vonden.
De onderzoekers keken of het onderzoek goed uit te voeren was.

Muziek maken vonden de deelnemers leuk en waardevol.
Zij waren blijer en rustiger.
De deelnemers voelden zich ook meer verbonden met elkaar.
De muziek-activiteit had soms praktische problemen.
Het was bijvoorbeeld lastig om tussen de lessen door huiswerk te maken.
De deelnemers die activiteiten deden vonden dat saai en steeds hetzelfde.

Het onderzoek was lastig uit te voeren.
Er moesten veel formulieren en vragen-lijsten worden ingevuld.
Begeleiders zeiden dat dit veel tijd kostte.
Sommige vragen-lijsten en testen waren te moeilijk voor de deelnemers.

Het onderzoek gaf nuttige lessen voor nieuw onderzoek.
Maak alles eenvoudiger en beter passend was een tip.
Ook is duidelijke uitleg en hulp belangrijk.
Daarom deden de onderzoekers een nieuw onderzoek.

Deel 2

Deel 2 gaat over een onderzoek tijdens de corona tijd.
De corona tijd was heel erg voor mensen met een verstandelijke beperking.
Heel veel mocht niet.
Muziek maken op locatie was lastig.
Muziek maken met elkaar online kon wel.

Deelnemers aan dit onderzoek kregen 16 muzieklessen van 1 uur.
Ze kregen 1-op-1 les van een muziekbegeleider.
Ze kozen samen of de les op de locatie of online was.
10 mensen van Philadelphia deden mee.
De onderzoekers keken of de muziek-activiteit goed werkte.
De onderzoekers keken ook wat de deelnemers ervan vonden.
En of het gedrag van de deelnemers veranderde.
En of zij zich beter voelden.

De deelnemers en de muziekbegeleiders waren blij met de lessen op locatie.
Zij vonden het fijn om samen muziek te maken.
Het moeilijk gedrag van de deelnemers werd minder.
Muziek maken op de locatie was fijner dan online muziek maken.
Ook waren er technische problemen met de computer.
Online muziek maken gaat beter als het kort is.
En als je eerst op locatie begint.

Deel 3

Als laatste hebben de onderzoekers een groot onderzoek uitgevoerd.

In dit grote onderzoek gingen ook weer mensen van Philadelphia muziek maken.

Om te kijken of muziek maken moeilijk gedrag minder maakt.

Ook werd naar andere dingen gekeken.

Bijvoorbeeld of je bang bent.

En of de deelnemers beter kunnen denken als ze muziek maken.

Er deden bijna 100 mensen van Philadelphia mee.

Deze deelnemers werden verdeeld in 2 groepen.

1 groep mensen ging muziek maken.

En de andere groep mensen ging geen muziek maken.

Mensen die muziek maakten deden dat 16 keer.

Elke keer 1 uur.

En 2 keer per week.

Muziek maken ging steeds op dezelfde manier.

Zo dat de deelnemers het fijn vonden.

Zij zongen en speelden instrumenten

Muziekbegeleiders gaven de lessen.

De deelnemers vulden vragen-lijsten in.

En ze deden testjes op een tablet.

De begeleiders van de groep vulden ook vragen-lijsten in.

De vragen-lijsten werden 3 keer ingevuld.

Er werd gevraagd wat iedereen van het muziek maken vond.

En of het moeilijk gedrag van de deelnemers minder werd door muziek te maken.

De onderzoekers wilden ook weten hoe het muziek maken was gegaan.

En of de deelnemers muziek bleven maken.

De mensen die muziek maakten hadden minder vaak moeilijk gedrag.
Ook waren de deelnemers die muziek maakten minder bang.
Zij waren blijer nadat ze muziek gemaakt hadden.
En de mensen die al blij waren werden na het muziek maken nog blijer.
Er was geen verschil in denken tussen de mensen die muziek maakten.
En de mensen die geen muziek maakten.
Het invullen van de vragen-lijsten en maken van de testen was soms lastig.

De deelnemers vonden muziek maken leuk.
En ze deden goed mee.
Begeleiders zagen mensen vrolijker en rustiger worden.
Zelfvertrouwen en minder bang zijn ging soms beter.
Maar dit bleef niet lang.
Een goede band met de muziekbegeleider was erg belangrijk.
Na de 16 keer muziek maken bleven veel mensen door-gaan met muziek.
Begeleiders vonden doorgaan met muziek maken lastig.
Er was te weinig tijd voor de begeleiders.
Duidelijke afspraken en genoeg tijd zijn nodig voor begeleiders.
Zodat muziek maken in de groep blijft.

Einde

We hebben veel geleerd van dit onderzoek.

Muziek helpt wanneer iemand moeilijk gedrag heeft.
Deelnemers werden rustiger.
En voelden zich vaak beter na muziek maken.
Ook begeleiders merkten dit.
De meeste mensen vonden muziek maken leuk.
En ze deden goed mee.
Sommigen gingen door met muziek maken.

Muziek is niet altijd beter dan andere activiteiten.

Deelnemers moeten kiezen wat ze leuk vinden.

1-op-1 muziek maken gaat beter dan in een groep.

Muziek maken op locatie is fijner dan online.

De onderzoekers willen nog meer weten.

Bijvoorbeeld wanneer muziek maken beter is dan andere activiteiten.

En hoe mensen met een verstandelijke beperking het beste vragen kunnen beantwoorden.

Muziek is belangrijk.

Muziek moet vaker op de groep gemaakt worden.

Begeleiders van Philadelphia hebben hulp en tijd nodig.

Zo kan muziek maken mensen met een verstandelijke beperking echt helpen.

APPENDICES



Dankwoord

altied, wat ge ok dut blief 't vertrouwe beware

Rowwen Hèze

Eindelijk, na zóveel jaar, ben ik dan toe aan het dankwoord. Wat heb ik hier lang naar uitgekeken! Het volgen van een promotietraject heb ik ervaren als een proces van *vechte, valle en opstoan*. Ik voelde me vaak niet alleen een onderzoeker, maar ook een planner, financieel expert, logistiek coördinator, privacy officer, administratieve kracht, vormgever, stagebegeleider, archivaris, boekhouder, imposter syndrome-expert en zo veel meer. Ik heb 11 keer gedacht: 'ik stop ermee' en gelukkig 12 keer: '*de rug recht, de schouders d'r onder*'. Ondanks (of dankzij) tegenslagen en vertraging heb ik ontzettend veel geleerd. Over onderzoek doen, maar misschien nog wel meer over mezelf. Ik weet dat ik nooit uitgeleerd ben, maar ik weet inmiddels ook dat er heel veel te zeggen is voor rust, balans en hier-en-nu.

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APPENDICES



Curriculum Vitae

Gerianne Smeets was born on February 27, 1988, in Heerlen, the Netherlands. After completing her pre-university education (gymnasium) at the Bernardinuscollege in Heerlen, she enrolled in the International Business program at Maastricht University in 2006. However, within one year she realized this was not the right fit and she decided to switch to Health Sciences, also at Maastricht University. She obtained her bachelor's degree in 2009, majoring in Mental Health Sciences.

She continued her academic journey at Tilburg University, where she completed the master's program in Psychology & Mental Health, track Clinical Psychology. As part of this program, she completed a clinical internship at the Orbis Medisch Centrum (now Zuyderland Medisch Centrum) in Sittard-Geleen, after which she received her basic psychodiagnostics registration (diagnostiekaantekening).

Following her graduation in 2011, Gerianne worked briefly in various roles, including as a research assistant, before starting as a psychologist at Philadelphia Care Foundation (Stichting Philadelphia Zorg) in 2012. In this role, she worked within residential facilities for adults with primarily mild intellectual disabilities and gained substantial experience in working with adults with autism. In addition to her clinical work, she developed guidelines and provided courses for caregivers on autism and suicide prevention in adults with intellectual disabilities.

In January 2017, she began her PhD as an external candidate at VU Amsterdam. Her research, focused on the effects of music interventions for adults with intellectual disabilities, was supervised by Professor Erik Scherder, Professor Xavier Moonen, and Dr. Karin Volkers. A year later she also enrolled in a post-initial master's program in Epidemiology at VU Amsterdam, which she successfully completed in 2021. Throughout her PhD, Gerianne continued her role as a psychologist at Philadelphia Care Foundation, a position she will retain following the completion of her doctorate.

APPENDICES



List of publications

Published

Smeets, G. J. J., Van Rest, M. M., Volkers, K. M., Moonen, X. M. H., & Scherder, E. J. A. (2025). The effects of an individual music intervention on executive functions in adults with intellectual disabilities. *Acta Psychologica*, *259*, 105356. https://doi.org/10.1016/j.actpsy.2025.105356

Smeets, G. J. J., Volkers, K. M., Scherder, E. J. A., & Moonen, X. M. H. (2023). Active music making and leisure activities for people with intellectual disabilities: A cluster randomized pilot study. *Research in Developmental Disabilities*, *142*, 104606. https://doi.org/10.1016/j.ridd.2023.104606

Smeets, G., Volkers, K., Scherder, E., & Moonen, X. (2024). An Individual Music Intervention for Adults With Intellectual Disabilities and Challenging Behavior: Protocol for a Randomized Controlled Trial. *JMIR Research Protocols*, *13*, e52497. https://doi.org/10.2196/52497

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Under review

Smeets, G. J. J., Volkers, K. M., Hoozemans, M. J. M., Scherder, E. J. A., & Moonen, X. M. H. The impact of a music intervention on challenging behaviour, mental health and well-being in adults with intellectual disabilities: a cluster-randomized controlled study.

Smeets, G. J. J., Volkers, K. M., Scherder, E. J. A., & Moonen, X. M. H. Process evaluation of an individual music intervention for adults with intellectual disabilities in the context of a randomised controlled trial.

Swart, E. A. K., Smeets, G. J. J., Volkers, K. M., Scherder, E. J. A., & Moonen, X. M. H. Feasibility of a pilot cluster RCT with a music and a leisure activities intervention for adults with intellectual disabilities.

