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The effect of MultiSensory Music Drama on the interactive engagement of students with severe/profound intellectual and multiple disabilities

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Abstract

Background: Interactive engagement, specifically attention and initiation, are considered important skills for facilitating development and learning in students with severe/profound intellectual and multiple disabilities. Prior research has suggested that music therapy and multisensory storytelling are two promising interventions for supporting interactive engagement in this population. However, there are no known/published studies that have investigated the effect of combining relevant elements from musical interaction and multisensory story-telling for this group of students. This study aims to determine the effect of a novel educational approach—MultiSensory Music Drama (MSMD)—on the interactive engagement of students with severe/profound intellectual and multiple disabilities.

Methods: A single-case experimental design, specifically an ABAB design across three participants, was used to describe the effect of MSMD on interactive engagement when compared with a control activity. School staff members' perceptions of student interactive engagement and social validity were obtained.

Findings: Coded video observations showed significantly higher levels of interactive engagement during MSMD for all three participants when compared to the control activity. Furthermore, the ratings on the interactive engagement from school staff members improved for two of the three participants. Finally, school staff members scored the social validity and perceived educational usefulness of the MSMD as very high.

Conclusions: The findings provide initial evidence that MSMD may be effective in supporting the interactive engagement of some students with severe/profound intellectual and multiple disabilities.

KEYWORDS

interactive engagement, music, severe/profound intellectual and multiple disabilities, special education

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Accessible summary

- Engagement in activities is important for development and learning.
- Many people enjoy music activities and taking part in storytelling.
- In this study, a combination of music, drama and sensory stimulation was tested with three students with intellectual and multiple disabilities.
- The activity was engaging for the students and was positively viewed by their teachers.

1 | INTRODUCTION

Persons with severe/profound intellectual and multiple disabilities are dependent on others in relation to daily living activities. They require supportive relationships to express their abilities, their will and their personalities (Granlund et al., 2013; Nakken & Vlaskamp, 2007). Besides severe/profound intellectual and sensory-motor impairments, they commonly also have medical comorbidities (Nakken & Vlaskamp, 2007). Still, this group of people is a heterogeneous group in aetiology, functioning and behaviours (ibid). Their communication is often nonverbal, idiosyncratic and expressed by bodily movements, gestures and sounds (Griffiths & Smith, 2016).

The role of the interaction partner is considered vital for communication development in children with severe/profound intellectual and multiple disabilities (Van Keer et al., 2017). More specifically, responsive partner strategies are important which include waiting for the person to take initiatives, providing time for responses, interpreting sounds, movements and gestures as intentional and meaningful contributions to the communication. and responding accordingly (Wandin et al., 2021). It has been argued that if a person has severe/profound intellectual and multiple disabilities, then patterns of social interaction dynamics are affected (Munde & Zentel, 2020; Wilder, 2008). The complexity of their disabilities may result in a limited repertoire of communicative expressions, as well as physically weaker, slower and more subtle communicative signals from the person (Griffiths & Smith, 2016). Importantly, Van keer et al. (2020) found that responsive partner strategies are significantly related to the interactive engagement of children with severe/profound intellectual and multiple disabilities. In addition to a focus on responsive interaction, Munde and Zentel (2020) highlight the importance of providing choice-making opportunities in meaningful activities such as sensory stimulation and music activities. In addition, individually tailored support was seen as paramount in educational work with students with severe/profound intellectual and multiple disabilities: 'all teaching activities need to be designed according to the strength and needs of each individual learner' (Munde & Zentel, 2020, p. 1).

Interactive engagement behaviours—for example, attention and initiation—are pivotal behaviours for development and learning (Mahoney et al., 2007; Van keer et al., 2019, 2020), that is, central to the development of wide areas of functioning. Hence, a positive change in a pivotal behaviour will potentially produce improvements in a number of other outcomes (Van keer et al., 2019, 2020). Attention is one such pivotal skill central to learning and refers to the selection process that helps us focus on certain aspects of the world while filtering out others (Dehaene, 2020). Initiation can be defined as introducing or starting an interaction or activity (Munde & Vlaskamp, 2015). Persons with severe/profound intellectual and multiple disabilities have been shown to fluctuate in their attention, which limits possibilities for engagement, initiation and learning (Munde & Zentel, 2020).

To support attention and initiation for students with severe/ profound intellectual and multiple disabilities, it is important to build interventions around motivating activities (Munde & Zentel, 2020). Yet, much remains to be learned about how such educational activities should be constituted and organised for these students (Hardesty-Jaynes, 2021). Since the late 1960s, music therapy with people with severe/profound intellectual and multiple disabilities has aimed at supporting nonmusical goals, such as nonverbal communication, social relatedness and engagement (Johnels et al., 2021; Wheeler, 2013). Indeed, music therapeutic approaches involving direct interaction between student and teacher/therapist (i.e., musical interaction) are often integrated into education for these students (McFerran & Shoemark, 2013). Musical interaction approaches are presumed to make use of our innate musicality. Preverbal music activities include using mirroring and taking turns during singing, vocalising or playing and playfully tuning into the child's engagement. These activities not only support musical development but may also potentially improve general development concerning communication skills (Johnels et al., 2021; Rushton & Kossyvaki, 2022). According to a scoping review, musical interaction with children and youths with severe/profound intellectual and multiple disabilities was indeed found to be a promising way of supporting several of the interactive engagement behaviours discussed above, such as engagement, (joint) attention and initiatives (Johnels et al., 2021). In addition, several promising components of musical interaction were identified. These included responsive partner strategies, where the interaction partner listens, tunes in and follows the child's lead. In addition, a flexible use of musical routines within a predictable structure was seen to enable anticipation and promote student initiation. The use of technology-mediated and multisensory music activities was reported to engage the children to explore and participate, and it

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provided participants with large physical limitations new opportunities to express themselves musically.

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A parallel approach to music therapy is multisensory storytelling, which involves direct interactions to engage attention, listening and interpretation using sensory stimuli to support storytelling (ten Brug, 2015). To a large extent, this is an individualised activity, where the form and content of the story are adjusted to the individual with severe/profound intellectual and multiple disabilities (Penne et al., 2012). To optimise alertness and attention in the individual, multisensory storytelling emphasises the importance of performing the story in a consistent way (e.g., using both verbal text and sensory stimuli) and providing the listener with opportunities to manipulate the multisensory objects (ten Brug, 2015). Research has shown that multisensory storytelling can potentially enhance social interaction, engagement and attention in people with severe/ profound intellectual and multiple disabilities (ten Brug et al., 2016; Young et al., 2011).

Interestingly, outside the context of students with severe/ profound intellectual and multiple disabilities, adding music during interactive storytelling has been shown to improve engagement and learning. In classroom-based research with students with autism, Carnahan and colleagues demonstrated that combining music and storytelling elements increased engagement in several students (Carnahan et al., 2009). Similarly, Fornefeld (2013) specifically highlighted the use of music and prosody of language in multisensory storytelling with persons with disabilities, including those with severe/profound intellectual and multiple disabilities. This use of musical elements is specifically described to support a process of sensory-aesthetical comprehension. These findings might suggest that the combination of multisensory storytelling, dramatised interaction and musical interaction might confer an added value compared with any of these activities and interventions considered alone. However, there is a paucity of research regarding the systematic joint use of dramatised and multisensory storytelling and musical interaction in supporting interactive engagement in persons with severe/profound intellectual and multiple disabilities specifically.

There is a growing emphasis in educational practices and in policy contexts on identifying scientifically supported interventions and approaches when it comes to pedagogical work with people with severe/profound intellectual and multiple disabilities (Munde & Zentel, 2020), as well as in special education more generally (Odom et al., 2003). However, there is a welldocumented research practice gap in many research and practice fields, including in special education (Getenet, 2019; Greenwood & Abbott, 2001). Greenwood and Abbot discuss several reasons for this gap, especially as the relevance of much educational research is commonly considered low by school staff, as there is a perceived failure to produce interventions that are feasible and useful in classroom settings. To address the question of social validity and implementation of intervention research in school contexts, it is vital to solicit educators' input on the interventions (Komesidou et al., 2022). In the context of students with severe/profound intellectual and multiple disabilities, the inclusion of stakeholder

perspectives seems particularly relevant considering that current scientific knowledge regarding educational approaches for these students is highly limited (Hardesty-Jaynes, 2021; Munde & Zentel, 2020).

In the current study, we aim to evaluate the effectiveness of a custom-developed educational approach called MultiSensory Music Drama (MSMD) that systematically combines musical interaction with dramatised multisensory stimulation (Johnels, 2022). A more detailed description will be provided in the Section 2, but in essence, MSMD is an individualised approach consisting of a short narrative/drama including sensory stimulation and music activities. It is created to fit the individual's preferences and abilities. The drama includes opportunities for student agency, such as choice-making opportunities, exploration of favourite props and dancing/movement. In addition, goals related to the educational syllabus are considered in the drama. The goal of the approach is to support interactive engagement in the individual and it is informed by research and practical experience regarding beneficial educational approaches for students with severe/ profound intellectual and multiple disabilities (Johnels et al., 2021: McFerran & Shoemark, 2013; Munde & Zentel, 2020; Nind, 2007; ten Brug, 2015). The responsivity of the interaction partner, communication support and repetition within a set structure is emphasised in the approach to enable student anticipation and agency. An MSMD lesson is recommended to last ~15 min.

The following research questions were addressed:

- What is the effect of MSMD on the interactive engagement behaviours in students with severe/profound intellectual and multiple disabilities?
- To what extent is the MSMD approach perceived as useful and socially valid by the participating school staff members?

2 | METHODS

2.1 | Setting, recruitment and ethics

In Sweden, where this study took place, there are two adapted forms of education eligible for students with intellectual disabilities. School for students with intellectual disabilities, where students with mildto-moderate intellectual disabilities receive their education and where the school subjects are similar to the general Swedish compulsory school but with greater flexibility in progress and learning goals; and school for students with more severe-profound intellectual disabilities where the curricula are more related to daily living skills than to academic achievement, mainly in relation to art activities, communication, motor abilities, activities of everyday living (aiming at developing students' skills in relation to everyday life routines) and practical applications of technology and science. All students participating in the current study received the latter curriculum. Due to the complex needs of students with severe/ profound intellectual and multiple disabilities, special schools are commonly organised in small classes with a high staff ratio (Östlund, 2015).

Ethical approval was obtained from the ethical review board in Uppsala, Sweden (Dnr: 2019-05328). Contact was made with special education teachers and principals working at schools for students with severe/profound intellectual and multiple disabilities in Gothenburg, Sweden, via e-mail. One of the contacted schools expressed interest and written permission was obtained from the school principal and staff to participate in the study. The first author met with all special education teachers working at the school through a video conference and the study was explained, as well as the participant selection criteria. Three of the special education teachers (from three of the school's four classes) volunteered to participate and each suggested one potential student from their class who matched the inclusion criteria (see below). Also, the teachers suggested one teacher assistant each from their class for participation. The teachers contacted the caregivers of the potential students and provided them with contact information for the researchers and information about the study. The caregivers of all three students provided their informed written consent for their children to participate in the study. Given the students' severe/profound level of intellectual disability and complex communication needs, it was not possible to elicit assent from the students. Participating school staff members and the first author were very attentive to any signs of discomfort in the students. If the students showed signs of overarousal (i.e., getting too excited or upset) or were too tired to participate, the lesson was stopped, which happened on two occasions out of a total of 48 lessons.

2.2 | Participants

The inclusion criteria for participants were that they (i) studied a curriculum on daily living skills for students with severe/profound intellectual disabilities; (ii) were between the ages of 7 and 12 years old; (iii) had severe/profound intellectual and multiple disabilities, and no or minimal verbal language. Three students participated in the

study. Table 1 summarises the participant descriptions reported by parents and school staff members. (No medical records or school records were available to the researchers).

Two school staff members per student (one special education teacher and one teacher assistant) participated in the study. All teachers were fully qualified special education teachers. The teacher assistants had a secondary educational background. Hereafter, the term 'school staff members' is used for all of the participating school staff (i.e., two members per student).

2.3 | Study design

The study adhered to principles for single-case experimental design (SCED) studies (Kratochwill et al., 2013) to identify a possible effect of MSMD (the independent variable) versus a control activity (joint picture book reading) on student interactive engagement (the dependent variable). The independent variable was systematically altered in an ABAB design in which the 'A' phases were the joint reading (control activity) and the 'B' phases were the MSMD lessons. Each phase in the study consisted of four lessons. Thus, in all, 16 lessons were carried out individually with each student. Our study included three attempts to demonstrate an intervention effect, that is, between conditions A1 and B1. B1 and A2. A2 and B2. To avoid order effects, the order of the conditions for each participant was randomised using an online resource: http://slump.nu. Students 1 and 2 started with the joint reading and Student 3 started with MSMD. The length of the lessons ranged between 9.51 and 22.12 min. The average length of the lessons was 13.8 min (see Table 3).

2.4 | Materials and procedures

2.4.1 | Measures

1. The Child Behaviour Rating Scales, revised–(CBRS; Mahoney, 1998) is an observation-based rating instrument examining

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Student	Age	Sex	Additional diagnoses	Communication and motor ability	Interests
1	9 years old	Male	Cerebral palsy, microcephaly	A few words. Eye contact, sounds, facial expressions, using hands and mouth to explore objects. Non-ambulatory.	Socially interested. Playing with water and soap bubbles, exploring different materials, music and children's songs and being outside.
2	8 years old	Female	Autism, epilepsy	No verbal language. Sounds, eye contact, facial expressions, using mouth to explore objects. Ambulatory with support.	Music, drumming and playing the flute, likes physical contact, massage and movement, playing with water and joint reading.
3	12 years old	Male	Epilepsy	No verbal language. Eye gaze, sounds, facial expressions, uses hands and mouth to explore objects. Nonambulatory.	Socially interested, likes physical contact. Likes to be outside. Likes joint reading and children's songs.

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TABLE 2 Content of individualised MSMD.

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	Sensory stimulation	Songs/music
Student 1	 Plants from the forest: fir tree, willow, moss (touch, smell, taste) Recordable single-message devices with sounds of an owl and a forest creek (sound) Toy snail (touch) Toy bird with sound (touch, sound) Water (touch, sound) Scarf (sight, touch) 	 Starting song (singing names and taking turns) Song with playful anticipation element (playing instrument, and 'being caught') Water song (playing with water) Instrument song (playing together) Goodbye song (signal for finish)
Student 2	Recordable single-message device with sound from a spaceship (sound) Ice (touch, taste) String of beads (touch, sight, sound) Hot wheat pillows (touch) Scarfs (sight, touch) Drum (touch, sound) Toy animals (touch, sound, sight)	 Taking off (drum-playing and sound-making to illustrate the launch of the spaceship) Ice planet song (playing instruments together on iPad application or percussion instruments) Sun and fire song (song with playful anticipation element; drumplaying, waving with scarfs) Going home (drum-playing and sound-making to illustrate the launch of the spaceship) Ocean song (playing an ocean drum) Goodbye song (signal for finish)
Student 3	 Plants from the forest: fir tree, willow, moss (touch, smell, taste) Recordable single-message devices (sounds from an owl, and a forest creek) Toy snail (touch) Toy bird with sound (touch, sound) Water (touch, sound, taste) Scarf (sight, touch) 	 Starting song (singing our names) Song with playful anticipation element (playing a musical instrument and 'being caught') Water song (playing with water) A lullaby (playing musical instruments) Goodbye song (signal for finish)

Abbreviation: MSMD, MultiSensory Music Drama.

aspects of interactive engagement of persons with severe/ profound intellectual and multiple disabilities (Hostyn et al., 2011; Van keer et al., 2017, 2020). In CBRS, 10 min of video recordings of interactions are coded on a five-point Likert scale. The CBRS consists of two overarching constructs based on a total of seven subscales: attention (attention to activity, persistence, involvement, compliance/cooperation) and initiation (initiation: activities, initiations: adult, affect). The CBRS has shown adequate reliability and validity in previous research studies with the target group (Hostyn et al., 2011; Van keer et al., 2017, 2020, but see Rensfeldt Flink et al., 2022). The coding procedure was conducted for each 2-minute interval, that is, in total, five ratings per video. (If the video recording exceeded 10 min, only the first 10 min were scored).

- 2. As an additional index of the dependent variables, the Engagement Model (Carpenter et al., 2015) was used. In this assessment, the two participating school staff members working with each student independently rated five indicators of engagement of the student following each lesson (control activity and MSMD): exploration, realisation, anticipation, initiation and persistence on a four-point Likert scale. This resulted in a total engagement score for each lesson.
- 3. 'Teacher Post-Intervention Acceptability and Importance of Effects Survey' (Lane & Beebe-Frankenberger, 2004) was completed by the two participating school staff members working with each of the three students respectively regarding the perceived usefulness and social validity of the MSMD approach

(cf., Carnahan et al., 2009). The survey consisted of 10 questions related to the social validity of the approach in the pedagogical context as well as the suitability in engaging and developing abilities in the students. Each question could be rated on a Likert scale ranging from 1 to 7, where 1 corresponded to 'do not agree at all' and 7 corresponded to 'strongly agree'.

2.4.2 | MSMD

Before the start of the data collection, the first author had an online meeting and a lecture with all special education teachers working at the school, where background information on MSMD was provided. Also, the first author had a separate on-site meeting with each student's two participating school staff members to discuss and summarise interests, strengths and ways to communicate for each student. The first author presented three examples of MSMDs for the school staff members (i.e., an adventure in the forest, a travel to outer space and a day at the beach). Each student's two school staff members choose an MSMD for the student and thereafter the first author individually adapted the activities in the MSMDs for each student based on the information provided by parents and school staff.

Students 1 and 3 had similar interests (i.e., being outdoors, exploring plants and animals) and the MSMD about an adventure to the forest was chosen. The students were able to explore different plants and toy animals, water and different musical

instruments (i.e., drums, percussion instruments, an autoharp, an iPad Pro with the music application 'Bloom') and make choices about the continuation or ending of the drama via recordable single-message devices and visual support. The students' individual differences in temperament and preferences of songs resulted in slightly different MSMDs as can be seen in Table 2 (i.e., Student 3 preferred a calm lullaby, whereas Student 1 was offered a more up-tempo instrument song). For Student 2, the fantasy drama about a trip to outer space was chosen. Her school staff suggested this drama since they thought that the different musical activities and sensory stimulation provided on the different planets would be engaging for the student. In the drama, the student helped start the space rocket by playing on a big drum, she was able to explore different temperatures and props on different planets (e.g., ice on the ice planet, warm wheat pillows and red scarfs on the sun, as well as playing with water, exploring plants and toy animals when returning to planet Earth). She also was able to play different drums, percussion instruments and iPad applications with music. Similar to the other students, she was able to make choices about the continuation or ending of the drama via recordable singlemessage devices and visual support. An illustration and a description of the application of MSMD are summarised in Figure 1.

A booklet for visual communication support was made for each MSMD. Table 2 describes in detail the content of each student's individualised MSMD.

2.4.3 | Control activity

Joint reading was used as a control activity. Responsive partner strategies (waiting for the student to take initiative and to provide responses, interpret sounds, movements and gestures as intentional and meaningful) were used during both activities as was visual support (in the joint reading activity, this came in the form of the picture book). A narrative element was also integral to both activities. However, only in the MSMD did we use musical activities and propsbased sensory stimulation as described above. See Figure 2 for a visual illustration of the materials used for Student 2 in the MSMD, as well as the control activity.

2.5 | Data collection

The data collection period lasted over two consecutive months (November and December, 2021); however, due to the Christmas holidays, one of the students received the last lesson in January 2022. The MSMD and control activity lessons were carried out in the school during regular school hours by the first author, who is also a music educator and music therapist. The lessons were carried out individually in a room within the school. The school staff members assisting each student were present during the lessons to interpret communicative signals or detect signs of discomfort in the students. The lessons were carried out once per day, three or four times a



FIGURE 1 The interaction partner (to the right) is singing a song about a bird, in an MultiSensory Music Drama (MSMD) about the forest. Simultaneously, she is helping the student to hold a soft toy bird in her hand. The interaction partner supports the student to explore the bird by stroking the feathers and squeezing it (to make sounds). On the table in front of the student, a booklet covering the storyline of the MSMD is placed. Also, it entails visual communication support which is used as augmented input to the spoken language. The interaction partner is constantly attentive to detect any signals from the student. The interaction partner interprets and confirms the student's reactions by pointing at the symbols in the visual communication support, and through imitating and taking turns with the student in the activity. A big drum, which is one of the student's favourite instruments, is placed next to her wheelchair. In the upcoming music activity, the student and the interaction partner are taking turns in a playful music activity including singing and doing an 'l-caught-you-game'.

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FIGURE 2 Materials used in the MultiSensory Music Drama (left) and control activity (right).

TABLE 3Data collection activities.

		Student 1 (date) duration in minutes	Student 2 (date) duration in minutes	Student 3 (date) duration in minutes
Lesson number	1	Reading (8/11) 14:36	Reading (8/11) 13:52	MSMD (8/11) 21:50
	2	Reading (10/11) 10:51	Reading (10/11) 11:46	MSMD (10/11) 19:26
	3	Reading (11/11) 11:20	Reading (11/11) 10:57	MSMD (11/11) 15:43
	4	Reading (15/11) 10:14	Reading (15/11) 12:05	MSMD (15/11) 17:55
	5	MSMD (17/11) 13:30	MSMD (17/11) 13:06	Reading (17/11) 13:11
	6	MSMD (18/11) 15:16	MSMD (18/11) 13:04	Reading (18/11) 11:21
	7	MSMD (19/11) 14:58	MSMD (19/11) 13:31	Reading (19/11) 13:05
	8	MSMD (22/11) 14:06	MSMD (25/11) 13:31	Reading (22/11) 13:26
	9	Reading (25/11) 11:55	Reading (26/11) 11:22	MSMD (25/11) 15:47
	10	Reading (26/11) 9:51	Reading (29/11) 10:18	MSMD (26/11) 17:20
	11	Reading (29/11) 10:50	Reading (1/12) 11:56	MSMD (29/11)18:27
	12	Reading (1/12) 10:31	Reading (2/12) 15:14	MSMD (1/12) 22:12
	13	MSMD (13/12) 18:18	MSMD (3/12) 14:48	Reading (3/12) 17:19
	14	MSMD (15/12) 14:05	MSMD (6/12) 10:34	Reading (20/12) 12:19
	15	MSMD (15/12) 14:22	MSMD (16/12) 14:19	Reading (21/12) 13:38
	16	MSMD (17/1)16:31	MSMD (17/12) 13:49	Reading (21/12) 14:56

Abbreviation: MSMD, MultiSensory Music Drama.

week. Due to sick leave, two of the students had two lessons in 1 day on one occasion. See Table 3 for details regarding data collection activities.

All lessons were filmed using two Sony Handycam video cameras (model HDR-PJ650) on the stands. The video recordings were set up to capture the student as well as the overall interaction.

2.6 | Data analyses

Quantitative rating scores (CBRS and teacher ratings on the Engagement Model) were graphed and visually analysed within and

across phases. The visual analysis followed the procedure described by Lane and Gast (2014) as recommended by Vannest et al. (2018). The percentage of non-overlapping data (PND) was calculated as an effect size indicator. PND scores of >90 were interpreted as highly effective, 70%–90% as fairly effective and <50% as unreliable or ineffective (Scruggs & Mastropieri, 2013). Finally, effect sizes were also calculated using Tau-U; this method controls for possible positive baseline trends. A Tau-U score of 0–0.20 is considered to indicate a small effect, 0.21–0.60 is considered a moderate effect and 0.61–1.0 is considered a large effect (Vannest & Ninci, 2015). The Tau-U calculator from a website—singlecaseresearch.org—was used to calculate Tau-U scores.

2.7 | Inter-rater reliability

The video recordings of CBRS were performed by two coders—the second author, who coded all video recordings, and an independent coder, who coded a random 33% (N = 16) of the video recordings—from both the A and the B phases to evaluate inter-rater reliability (IRR). To calculate IRR, intraclass correlations (ICCs) with absolute agreement and values for a single measure were used. The two subscales that constitute the main outcome measures, that is, the attention and initiation subscales, had ICCs of .57 and .61, respectively. According to criteria in Cicchetti (1994), the obtained ICCs are judged as 'good' (>0.6) and 'fair' (0.4–0.6). Considering the well-known difficulty of making these types of ratings with the current target group (e.g., Rensfeldt Flink et al., 2022), we consider the obtained ICCs as satisfactory.

3 | FINDINGS

3.1 | Interactive engagement of students

As shown in Figures 3–5, there is a difference in favour of the MSMD lessons for all three students according to visual analysis of all coded interactive behaviours from CBRS, although visual inspection reveals this is less clear for student 3. The mean and median scores during the MSMD phases were higher for all three students (see online Supporting Information for details). For Students 1 and 2, visual inspection demonstrates a clear effect of MSMD on interactive behaviours, although the differences were larger for the attention scores than for the initiation scores. The PND (percent of non-overlapping data points) for Students 1 and 2 was 100% and there was an immediate increase in scores when the MSMD lessons started. For Student 3, the PND was 75% for attention and 63% for initiation. There was also no clear immediate change in scores between the different phases. The control phase (baseline) was stable for all three students.

Ratings of aspects of engagement, made by the participating school staff members, were analysed using the Engagement Model (Carpenter et al., 2015). The results are also visually graphed, as shown in Figures 6-8. For students 1 and 2, the mean and median scores were higher during the MSMD phase compared with the control phase (see online Supporting Information for details). For Student 1, the control phase was variable but decelerating. There was no immediate effect for Student 1 when the first MSMD phase started. As can be seen in Figure 6, the scores then increased rapidly from the second lesson in which MSMD was used. There was an immediate drop when the control phase was initiated a second time and an immediate increase when MSMD was introduced again. PND was 88%, thus demonstrating a fairly good effect of the MSMD. For Student 2, there was an immediate effect when MSMD was initiated and the scores dropped immediately when the control activity was introduced a second time. However, the baseline trend during the second control phase accelerated rather steeply. As can be seen in

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Figure 7, it is not impossible that the higher scores during the last MSMD phase mirror this positive trend. Nevertheless, PND was 100%, thus indicating that the MSMD phase was highly effective. Thus, it is fairly safe to conclude that the MSMD lessons had an impact on Student 2's engagement as rated by school staff members. For student 3, no effect of the MSMD lessons on the staff member ratings of engagement was demonstrated, with the ratings showing a highly variable pattern from day to day with the possible exception of the first MSMD phase.

MSMD and control activity were also compared using Tau-U (see Table 4). Tau-U showed significant levels on all coded interactive behaviour as coded by CBRS. Regarding the ratings of engagement, made by the participating school staff members, there were significant differences in favour of MSMD for Students 1 and 2. For Student 3, there were no significant differences.

3.2 | Perceived usefulness and social validity of MSMD

To address the second research question, the post-intervention survey (adapted from Lane & Beebe-Frankenberger, 2004) was analysed. The participating school staff members' sum score rated the approach very high, ranging from 60 to 65 out of a total of 70 (i.e., 86%–93% out of the maximum score).

All of the six participating school staff members scored the maximum (rating '7') on Item 4 ('was appropriate to use with the student'), as well as on Item 5 ('fit into the school context'). On Statement 3 ('the pedagogical approach was a good way to motivate the student'), all but one (who scored 6) scored 7. This was also the case with Statement 9 ('is a pedagogical approach that I will use in the future'). Statements 7 and 8 received an overall lower rating. Statement 7 ('improved the student's overall performance') received 5 from all but one staff member (who scored 4). Statement 8 ('will have lasting positive effects') received 5 from four of the informants and '4' from two of the informants. The items and the descriptive data are reported in Table 5.

4 | DISCUSSION

This study aimed at assessing the effects of the MSMD approach on the interactive engagement of students with severe/profound intellectual and multiple disabilities, as well as evaluating the perceived usefulness of the approach in the school setting, as reported by school staff members. The goal of MSMD is to support interactive engagement in the student. It is informed by research and practical experience regarding beneficial educational approaches for students with severe/profound intellectual and multiple disabilities (e.g., Johnels et al., 2021; Munde & Zentel, 2020; Nind, 2007), in which musical interaction and sensory stimulation are used conjointly with responsive partner strategies in a narrative activity. Thus, MSMD brings together



FIGURE 3 (a) Student 1: Child Behaviour Rating Scales (CBRS) attention score. (b) Student 1: CBRS initiation score. MSMD, MultiSensory Music Drama.

approaches suggested to be motivating and helpful in and of themselves. First, musical interaction is a key component and has previously been shown to provide a motivating framework for interaction (both for the interaction partner and the student with severe/profound intellectual and multiple disabilities), where the music engagement supports a playful and sustained flow in the interaction, despite students' fluctuating levels of attentiveness and rare initiations (Johnels et al., 2021; Perry, 2003). In addition,



FIGURE 4 (a) Student 2: Child Behaviour Rating Scales (CBRS) attention score. (b) Student 2: CBRS initiation score. MSMD, MultiSensory Music Drama.

repetition, a set structure in songs, a responsive interaction partner, and multisensory and technology-mediated music activities were seen in a prior scoping review on musical interaction to support initiation, engagement and anticipation in children and youths with severe/profound intellectual and multiple disabilities (Johnels et al., 2021). Second, multisensory storytelling has been evaluated in several studies including in school contexts for students with severe/profound intellectual and multiple disabilities (Fornefeld, 2013; Preece & Zhao, 2015; ten Brug, 2015). In this approach, the students get to take part in

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FIGURE 5 (a) Student 3: Child Behaviour Rating Scales (CBRS) attention score. (b) Student 3: CBRS initiation score. MSMD, MultiSensory Music Drama.

an individually adapted narrative by exploring props and it has been shown to support attentiveness and engagement in interaction in people with severe/profound intellectual and multiple disabilities (Penne et al., 2012; ten Brug, 2015). MSMD is particularly inspired by research suggesting that combining music interaction and storytelling elements might confer added pedagogical value compared with each approach considered separately (Carnahan et al., 2009; Fornefeld, 2013). In addition,



FIGURE 6 Student 1: School staff member rating scores of engagement. MSMD, MultiSensory Music Drama.



FIGURE 7 Student 2: School staff member rating scores of engagement. MSMD, MultiSensory Music Drama.



FIGURE 8 Student 3: School staff member rating scores of engagement. MSMD, MultiSensory Music Drama.

Participant	Measure of engagement	Tau-U	p	CI 85%
Student 1	CBRS, attention	1.00	0.0011	0.5591 < > 1
	CBRS, initiation	1.00	0.0011	0.5591 < > 1
	School staff member ratings	0.875	0.0043	0.434 < > 1
Student 2	CBRS, attention	1.00	0.0011	0.5591 < > 1
	CBRS, initiation	1.19	0.0001	0.7466 < > 1
	School staff member ratings	1.00	0.0011	0.559 < > 1
Student 3	CBRS, attention	0.75	0.0143	0.3091 < > 1
	CBRS, initiation	0.8437	0.0059	0.4028 < > 1
	School staff member ratings	0.1875	0.5403	-0.253 < > 0.628

TABLE 4Tau-U across all phases.

Note: Benchmarks for effectiveness: small < 0.20; moderate = 0.21–0.60; large = 0.61–0.1.0.

Abbreviations: CBRS, Child Behaviour Rating Scale; CI, confidence interval.

responsive partner strategies (Van keer et al., 2020; Wandin et al., 2021) are considered highly important in the MSMD approach, as is the general idea that learning in students with severe/profound intellectual and multiple disabilities is best

TABLE 5 Postintervention survey ratings of the perceived

 usefulness and social validity of MSMD by school staff members.

Item	Median	Min-Max
1. MSMD fit into the school schedule	6.5	4-7
2. MSMD did not take up too much time	7	6-7
3. MSMD was a good way to motivate the student	7	6-7
4. MSMD was appropriate to use with the student	7	7-7
5. MSMD fit into the school context	7	7-7
6. MSMD showed the student's abilities in the moment.	6	5-6
7. MSMD improved the student's overall performance	5	5-6
8. MSMD will have lasting positive effects	5	4-5
9. MSMD is a pedagogical approach I will use in the future.	7	6-7
10. MSMD is a pedagogical approach that I will recommend to others.	7	6-7

Note: The highest possible score per item is 7.

Abbreviation: MSMD, MultiSensory Music Drama.

fostered when the interaction is carried out in a nurturing and warm social climate (cf., e.g., Nind, 2007).

Overall, this study brings initial evidence that MSMD is indeed a potentially effective and pedagogically useful approach when

working with students with severe/profound intellectual and multiple disabilities. Specifically, the video observations, coded with the CBRS (Mahoney, 1998), showed a significant increase in students' interactive engagement during MSMD compared with joint reading, although the results for Student 3 were less clear. The coded assessment was largely supported by the ratings of the school staff members of student engagement, as these ratings showed a significantly so for Student 3. In addition, staff members reported a high or very high potential pedagogical value for the interventional set-up, that is, perceived usefulness and high social validity in the school context (c.f., Munde & Zentel, 2020). All in all, the approach of combining musical interaction with dramatised multisensory stimula-tion seems very promising for engaging these students.

As described above, research about students with severe/ profound intellectual and multiple disabilities is scarce, and there is specifically a need for more research evaluating the usefulness and effectiveness of interventions used in educational settings (Munde & Zentel, 2020). Hence, we argue that the current study adds important new knowledge to the research field.

4.1 | Methodological considerations and limitations

Given our encouraging results, it is important to highlight a number of critical features of the study design, including limitations, as well as chart directions for future research.

First, as this is the first study evaluating MSMD involving only three participants over a period of ~2 months, the study needs to be replicated to test the generalisability of the effects.

Second, it is important to highlight that we mainly focus on the student's interactive engagement during the activities studied, that is, in the moment. Although the focus on engagement in the 'here and now' is a key perspective for many professionals, as well as parents, interacting with this group of children (Granlund et al., 2013), it might nevertheless be interesting to examine whether a long-term intervention with MSMD, with detailed follow-up, might reveal more generalised effects, including transferable abilities, on students' social interactive abilities. Indeed, the concept of pivotal behaviours indicates that attention and engagement are central to the development of wide areas of functioning, such that a positive change in a pivotal behaviour will potentially produce improvements in a number of other behaviours (Van keer et al., 2019, 2020). Notably, however, in the postintervention survey (adapted from Lane & Beebe-Frankenberger, 2004), the item with the lowest rating was whether the MSMD approach was expected to have lasting positive effects on the students. Thus, at this stage, it is far from certain that any of the immediate positive effects observed here will lead to more generalised developmental effects. However, several school staff members also reported information suggesting that a continuation

of the MSMD lessons was expected to yield such improvement, which is interesting and clearly worthwhile to follow up on. A controlled follow-up study with outcome measures of general social interactive ability and functioning would constitute a rigorous evaluation of this possibility. To date, controlled studies focussing on educational outcomes in students with severe/ profound intellectual and multiple disabilities are lacking (Munde & Zentel, 2020).

Third, in the SCED, we selected the control activity of joint reading. This choice was arguably well-motivated since it is a common pedagogic activity in schools for the current group of students, including the three participants in our study. Additionally, the same control activity was used in the study by Carnahan et al., (2009), which focused on the effects of interactive reading materials using visual cues and music with children with autism (without severe/profound intellectual and multiple disabilities). That being said, it is important to note that another control activity might have yielded a different pattern of results than those presented here. In future research, it might be particularly interesting to explore whether there are certain aspects of the rather complex MSMD approach that yield particularly strong effects (e.g., musical interaction vs. multisensory stimulation vs. dramatised storytelling) or if it is the case-as we hypothesise-that the combined approach yields effects greater than the sum of its constituent parts.

Fourth, the outcome measure, interactive engagement, which in keeping with prior research (Van keer et al., 2019, 2020), operationalised with the attention and initiation subscales of CBRS and ratings of the Engagement Model could be discussed. Indeed, these are not the only possible choices of assessment instruments, and we think, in particular, that future research could include additional assessments of attention, covering more aspects of this broad construct, including gaze stability and social attention (Wandin et al., 2020).

Fifth, the intervention was carried out by the first author, who is a trained music educator and music therapist. In Swedish schools for students with severe/profound intellectual and multiple disabilities, the students seldom meet with a trained music teacher or therapist more than once or twice a week (and often more seldom) and typically in small group settings. Thus, exploring to what extent the students' 'ordinary' class teacher/ special educators or support staff can make use of the MSMD approach independently would be a very relevant future research topic. For instance, what level of musical ability is necessary, and what kind of professional development and supportive materials (e.g., prerecorded music and visual communication support) would be needed and perceived as helpful. Framing this research from the perspective of professional teacher knowledge and development might be fruitful in such future research (Van Driel & Berry, 2012) as would the inclusion of implementation science in the development of evidence-based or evidence-informed practices (Komesidou et al., 2022; Munde & Zentel, 2020).

4.2 | Practical implications

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If continued support for the approach is attained, a manual with learning materials needs to be made available to educators, which to date only exists in part and in Swedish (Johnels, 2022). It is our practical experience that teachers, educators and support staff are readily able to implement key aspects of MSMD, although the exact manner in which the pedagogical activity is carried out will always be affected by the individual educator/support staff, the student and the context. We hope the current work can inspire more research on MSMD and more generally on pedagogical approaches for engaging and supporting students with severe/ profound intellectual and multiple disabilities in educational settings.

5 | CONCLUSIONS

Current scientific knowledge regarding educational approaches for students with severe/profound intellectual and multiple disabilities is strikingly scarce. The findings of this study suggest that the combined usage of musical interaction with dramatised multisensory stimulation—MSMD—is effective in supporting the social interactive engagement of these students.

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CONFLICT OF INTEREST STATEMENT

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

Data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

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REFERENCES

Carnahan, C. C., Shobana musti-Rao, M., & Jody Bailey, B. (2009). Promoting active engagement in small group learning experiences for students with autism and significant learning needs. Education and Treatment of Children, 32(1), 37-61.

- Carpenter, B., Egerton, J., Cockbill, B., Bloom, T., Fotheringham, J., Rawson, H., & Thistlethwaite, J. (2015). Engaging learners with complex learning difficulties and disabilities: A resource book for teachers and teaching assistants. Routledge.
- Cicchetti, D. V. (1994). Guidelines, criteria, and rules of thumb for evaluating normed and standardized assessment instruments in psychology. *Psychological Assessment*, *6*, 284–290.
- Dehaene, S. (2020). How we learn: The new science of education and the brain. Penguin.
- Fornefeld, B. (2013). Storytelling with all our senses: Mehr-Sinn Geschichten. In N. Grove, Using storytelling to support children and adults with special needs (pp. 78–85). Routledge.
- Getenet, S. (2019). Using design-based research to bring partnership between researchers and practitioners. *Educational Research*, *61*(4), 482–494.
- Granlund, M., Wilder, J., & Almqvist, L. (2013). Severe multiple disabilities. In M. Wehmeyer, The Oxford handbook of positive psychology and disability (pp. 452–474). Oxford University Press.
- Greenwood, C. R., & Abbott, M. (2001). The research to practice gap in special education. *Teacher Education and Special Education: The Journal of the Teacher Education Division of the Council for Exceptional Children, 24*(4), 276–289.
- Griffiths, C., & Smith, M. (2016). Attuning: A communication process between people with severe and profound intellectual disability and their interaction partners. *Journal of Applied Research in Intellectual Disabilities*, 29, 124–138.
- Hardesty-Jaynes, T. L. (2021). Teaching students with profound intellectual and multiple disabilities [Doctoral dissertation]. Walden University.
- Hostyn, I., Petry, K., Lambrechts, G., & Maes, B. (2011). Evaluating the quality of the interaction between persons with profound intellectual and multiple disabilities and direct support staff: A preliminary application of three observation scales from parent-infant research. *Journal of Applied Research in Intellectual Disabilities*, 24(5), 407–420.
- Johnels, L., Vehmas, S., & Wilder, J. (2021). Musical interaction with children and young people with severe or profound intellectual and multiple disabilities: A scoping review. *International Journal of Developmental Disabilities*, 69(4), 487–504.
- Johnels, L. (2022). Multisensoriskt musikdrama Ett pedagogiskt arbetssätt för personer med flerfunktionsnedsättning. *Socialmedicinsk Tidskrift*, (5–6), 733–743.
- Komesidou, R., Feller, M. J., Wolter, J. A., Ricketts, J., Rasner, M. G., Putman, C. A., & Hogan, T. P. (2022). Educators' perceptions of barriers and facilitators to the implementation of screeners for developmental language disorder and dyslexia. *Journal of research in reading*, 45(3), 277–298.
- Kratochwill, T. R., Hitchcock, J. H., Horner, R. H., Levin, J. R., Odom, S. L., Rindskopf, D. M., & Shadish, W. R. (2013). Singlecase intervention research design standards. *Remedial and Special Education*, 34(1), 26–38.
- Lane, K. L., & Beebe-Frankenberger, M. (2004). School-based interventions: The tools you need to succeed. Pearson/Allyn and Bacon.
- Lane, J. D., & Gast, D. L. (2014). Visual analysis in single case experimental design studies: Brief review and guidelines. *Neuropsychological Rehabilitation*, 24(3–4), 445–463.
- Mahoney, G. (1998). Child Behavior Rating Scale (Revised). Case Western Reserve University.
- Mahoney, G., Kim, J. M., & Lin, C. (2007). Pivotal behavior model of developmental learning. Infants & Young Children, 20, 311-325.
- McFerran, K. S., & Shoemark, H. (2013). How musical engagement promotes well-being in education contexts: The case of a young

man with profound and multiple disabilities. International Journal of Qualitative Studies on Health and Well-Being, 8, 20570.

- Munde, V., & Vlaskamp, C. (2015). Initiation of activities and alertness in individuals with profound intellectual and multiple disabilities. *Journal of Intellectual Disability Research*, 59(3), 284–292.
- Munde, V., & Zentel, P. (2020). Evidence-based practices for teaching learners with multiple disabilities, Oxford research encyclopedia of education (pp. 1–31). Oxford University Press.
- Nakken, H., & Vlaskamp, C. (2007). A need for a taxonomy for profound intellectual and multiple disabilities. *Journal of Policy and Practice in Intellectual Disabilities*, 4, 83–87.
- Nind, M. (2007). Supporting lifelong learning for people with profound and multiple learning difficulties. Support for learning, 22(3), 111–115.
- Odom, S. L., Brown, W. H., Frey, T., Karasu, N., Lee Smith-Canter, L., & Strain, P. S. (2003). Evidence-based practices for young children with autism: Contributions for single-subject design research. Focus on Autism and Other Developmental Disabilities, 18(3), 166–175.
- Östlund, D. (2015). Students with profound and multiple disabilities in education in Sweden: Teaching organisation and modes of student participation. *Research and Practice in Intellectual and Developmental Disabilities*, 2(2), 148–164.
- Penne, A., ten Brug, A., Munde, V., van der Putten, A., Vlaskamp, C., & Maes, B. (2012). Staff interactive style during multisensory storytelling with persons with profound intellectual and multiple disabilities. *Journal of Intellectual Disability Research*, 56(2), 167–178.
- Preece, D., & Zhao, Y. (2015). Multi-sensory storytelling: A tool for teaching or an intervention technique? *British Journal of Special Education*, 42(4), 429–443.
- Perry, M. M. R. (2003). Relating improvisational music therapy with severely and multiply disabled children to communication development. *Journal of Music Therapy*, 40(3), 227–246.
- Rensfeldt Flink, A., Broberg, M., Strid, K., Thunberg, G., & Johnels, J. Å. (2022). Following children with severe or profound intellectual and multiple disabilities and their mothers through a communication intervention: Single-case mixed-methods findings. International Journal of Developmental Disabilities, 1–19.
- Rushton, R., & Kossyvaki, L. (2022). The role of music within the home-lives of young people with profound and multiple learning disabilities: Parental perspectives. *British Journal of Learning Disabilities*, 50(1), 29-40.
- Scruggs, T. E., & Mastropieri, M. A. (2013). PND at 25: Past, present, and future trends in summarizing single-subject research. *Remedial and Special Education*, 34, 9–19.
- ten Brug, A. (2015). A good read: A study into the use and effects of multisensory storytelling—A storytelling method for persons with profound intellectual and multiple disabilities [PhD dissertation]. University of Groningen.
- ten Brug, A., Van der Putten, A. A. J., Penne, A., Maes, B., & Vlaskamp, C. (2016). Making a difference? A comparison between multi-sensory and regular storytelling for persons with profound intellectual and multiple disabilities. Journal of Intellectual Disability Research, 60(11), 1043–1053.
- Van Driel, J. H., & Berry, A. (2012). Teacher professional development focusing on pedagogical content knowledge. *Educational Researcher*, 41(1), 26–28.

- Van keer, I., Colla, S., Van Leeuwen, K., Vlaskamp, C., Ceulemans, E., Hoppenbrouwers, K., Desoete, A., & Maes, B. (2017). Exploring parental behavior and child interactive engagement: A study on children with a significant cognitive and motor developmental delay. *Research in Developmental Disabilities*, 64, 131–142.
- Van keer, I., Ceulemans, E., Bodner, N., Vandesande, S., Van Leeuwen, K., & Maes, B. (2019). Parent-child interaction: A micro-level sequential approach in children with a significant cognitive and motor developmental delay. *Research in Developmental Disabilities*, 85, 172–186.
- Van keer, I., Bodner, N., Ceulemans, E., Van Leeuwen, K., & Maes, B. (2020). Parental behavior and child interactive engagement: A longitudinal study on children with a significant cognitive and motor developmental delay. *Research in Developmental Disabilities*, 103, 103672.
- Vannest, K. J., & Ninci, J. (2015). Evaluating intervention effects in singlecase research designs. *Journal of Counseling & Development*, 93(4), 403–411.
- Vannest, K. J., Peltier, C., & Haas, A. (2018). Results reporting in single case experiments and single case meta-analysis. *Research in Developmental Disabilities*, 79, 10–18.
- Wandin, H., Lindberg, P., & Sonnander, K. (2020). Development of a tool to assess visual attention in Rett syndrome: A pilot study. Augmentative and Alternative Communication, 36(2), 118–127.
- Wandin, H., Lindberg, P., & Sonnander, K. (2021). Aided language modelling, responsive communication and eye-gaze technology as communication intervention for adults with Rett syndrome: Three experimental single case studies. *Disability and Rehabilitation: Assistive Technology*, 1–15.
- Wilder, J. (2008). Video observations of dyadic interaction: Behaviour style of presymbolic children. Scandinavian Journal of Disability Research, 10(2), 104–124.
- Wheeler, B. L. (2013). Individuals with severe and multiple disabilities, Guidelines for music therapy practice in developmental health (pp. 399-440). Barcelona.
- Young, H., Fenwick, M., Lambe, L., & Hogg, J. (2011). Multi-sensory storytelling as an aid to assisting people with profound intellectual disabilities to cope with sensitive issues: A multiple research methods analysis of engagement and outcomes. *European Journal* of Special Needs Education, 26(2), 127–142.

SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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