Stockholm University

Musical Interaction with Students with Severe/Profound Intellectual and Multiple Disabilities

Linn Johnels

Doctoral Thesis in Special Education at Stockholm University, Sweden 2024

Musical Interaction with Students with Severe/ Profound Intellectual and Multiple Disabilities

Linn Johnels

Academic dissertation for the Degree of Doctor of Philosophy in Special Education at Stockholm University to be publicly defended on Friday 18 October 2024 at 09.00 in Auditorium 4, house 2, Albano, Albanovägen 18 and online via Zoom, public link is available through the department website.

Abstract

Research focusing on the educational strategies for students with severe/profound intellectual and multiple disabilities (S/PIMD) is strikingly scarce. Previous research has shown that the sensitive responsiveness of interaction partners, including school staff members, and motivating learning activities may promote interactive engagement (e.g., attention and initiation) in these learners. Interactive engagement is, in turn, considered important for learning and development more generally. Music interaction/therapy and multisensory storytelling are approaches that have shown positive results in supporting interactive engagement in individuals with S/PIMD. However, no known studies have investigated the effect of combining relevant elements from these approaches in educational settings with students with S/PIMD. The overall aim of the thesis was to develop new knowledge regarding how musical interaction with and without storytelling elements could support interactive engagement in students with S/PIMD using various methods in three sub-studies. Firstly, a scoping review of published peer-reviewed research was conducted to summarise the current evidence base on musical interaction with children and youths with S/PIMD (study 1). While the studies displayed great variation in quality and design, musical interaction was generally linked with positive outcomes for the participants with S/PIMD in several developmental domains, including communication, social interaction, engagement and cognition. Six categories of promising components of musical interaction were identified, namely the responsivity of the interaction partner, singing songs, structure and predictability in activities, a trusting alliance between interaction partners, long-term interventions, and using technology-mediated and multisensory music activities. Next, a novel pedagogical approach – MultiSensory Music Drama (MSMD) – was developed and empirically evaluated in terms of its effectiveness and social validity in school settings for students with S/PIMD (studies 2 and 3). To explore the effect of MSMD on interactive engagement, a study with a single-case-experimental design involving three participants was conducted (study 2). Data from the coded video observations showed higher levels of interactive engagement during MSMD relative to a control activity. Similarly, school staff members' ratings of interactive engagement were higher for two of the students during MSMD. In the last study, a special education teacher and her student with S/PIMD working with MSMD were followed in a qualitative, longitudinal case study. The results showed how the teacher, in her day-to-day teaching, implemented and developed MSMD lessons, with the process summarised in the following categories: *the reflective practitioner*; *enabling student agency*; and *the usefulness of MSMD*. Conclusions and implications from this thesis point to the importance of finding motivating pedagogical activities to support interactive engagement in students with S/PIMD. This thesis shows that MSMD could be an effective activity that enhances interactive engagement in participants with S/PIMD. The findings from the thesis collectively show how theoretically motivated and concrete learning activities, focusing on musical interaction with and without storytelling, support interactive engagement for these students. Also, it is perceived as feasible and helpful to integrate these into the everyday activities in schools.

Keywords: *Severe/profound intellectual and multiple disabilities, special education, musical interaction, multisensory music drama, responsive strategies, interactive engagement.*

Stockholm 2024
http://urn.kb.se/resolve?urn=urn:nbn:se:su:diva-232910

ISBN 978-91-8014-917-4
ISBN 978-91-8014-918-1

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ISBN print 978-91-8014-917-4
ISBN PDF 978-91-8014-918-1

Cover illustration: Lasse Johnels

Printed in Sweden by Universitetsservice US-AB, Stockholm 2024

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longitudinal case study. The results showed how the teacher, in her day-to-day teaching, implemented and developed MSMD lessons, with the process summarised in the following categories: *the reflective practitioner*; *enabling student agency*; and *the usefulness of MSMD*. Conclusions and implications from this thesis point to the importance of finding motivating pedagogical activities to support interactive engagement in students with S/PIMD. This thesis shows that MSMD could be an effective activity that enhances interactive engagement in participants with S/PIMD. The findings from the thesis collectively show how theoretically motivated and concrete learning activities, focusing on musical interaction with and without storytelling, support interactive engagement for these students. Also, it is perceived as feasible and helpful to integrate these into the everyday activities in schools.

Keywords: Severe/profound intellectual and multiple disabilities, special education, musical interaction, multisensory music drama, responsive strategies, interactive engagement.

Sammanfattning på svenska

Det behövs mer kunskap om undervisningsmetoder och aktiviteter för elever som har flerfunktionsnedsättning. Tidigare forskning har visat att lyhördhet och responsivitet hos skolpersonal samt motiverande aktiviteter i undervisningen kan gynna uppmärksamhet och engagemang hos eleverna, vilket i sin tur är viktiga förutsättningar för utveckling och lärande.

Musiksamspel/musikterapi och multisensoriskt berättande är arbetssätt som har visat sig stödja uppmärksamhet och engagemang hos elevgruppen. Forskningsbasen är emellertid begränsad, och det finns inga tidigare studier som på ett systematiskt sätt har kombinerat musikterapi och multisensoriskt berättande i en skolkontext för elever som har flerfunktionsnedsättning.

Det övergripande syftet i avhandlingen var att utvärdera och sammanfatta forskningsunderlaget om huruvida musiksamspel, med eller utan berättande och sinnestimulering, kan stödja uppmärksamhet och engagemang hos elever som har flerfunktionsnedsättning.

Olika metoder har använts i de olika delstudierna som ingick i avhandlingen. I ett första steg så gjordes en systematisk ”scoping” litteraturöversikt, där syftet var att summera och utvärdera tidigare forskning kring musiksamspel med barn och unga med flerfunktionsnedsättning (studie 1). Resultaten visade att studiernas forskningsdesigner liksom kvalitet varierade. Sex kategorier av vad som ansågs vara lovande komponenter i musiksamspel identifierades: att sampelspartnern var lyhörd och responsiv, att använda sånger, att ha en struktur och förutsägbarhet i musikaktiviteter, att ha en trygg relation med samspelspartnern, att insatser är långvariga och att använda teknikmedierade och multisensoriska musikaktiviteter. Sammanfattningsvis så visade studien att musiksamspel potentiellt kan stödja flera olika förmågor hos barn och unga med flerfunktionsnedsättning, såsom kommunikation, socialt samspel, engagemang och lärande.

I nästa steg så utvecklades och utvärderades ett nytt pedagogiskt arbetssätt – Multisensoriskt musikdrama (MSMD) – när det gäller dess potential att stödja engagemang och samspel för elever med flerfunktionsnedsättning i anpassad grundskola. Dessutom undersöktes hur skolpersonalen upplevde att arbetssättet passade in i skolkontexten (studie 2 och 3). För att utvärdera effektiviteten att stödja elevernas uppmärksamhet och engagemang genom MSMD, så genomfördes en experimentell studie där tre elever med flerfunktionsnedsättning jämfördes med sig själva under två olika

jämförelseaktiviteter, lektioner med MSMD och lektioner med bilderbokläsning (studie 2). Kodade videoobservationer av 16 lektioner per elev visade att lektionerna med MSMD var mer effektiva när det gäller att stödja engagemang och uppmärksamhet hos eleverna än jämförelseaktiviteten. På liknande sätt visade skolpersonalens skattningar på högre nivåer av engagemang för två av eleverna under MSMD lektioner jämfört med under lektioner med bilderbokläsning.

I den sista studien var en speciallärares arbete tillsammans med hennes elev med flerfunktionsnedsättning i fokus. Studien var en kvalitativ, longitudinell fallstudie, där samverkansforskning med läraren genomfördes för att implementera och utvärdera MSMD lektioner i hennes dagliga pedagogiska arbete under en period på fem veckor (studie 3). För att undersöka mer långsiktig användning av arbetssättet gjordes även en uppföljande intervju efter 20 månader. Resultatet summerades i tre kategorier: av (i) betydelsen av att vara en reflekterande praktiker; (ii) att skapa förutsättningar för elevens eget utforskande och delaktighet genom dynamisk balans i samspel aktiviteter; och (iii) att lärarens upplevde att arbetssättet med MSMD var motiverande för elever och användbart och relevant i relation till styrdokumenten.

Slutsatser och pedagogiska implikationer från avhandlingen rör vikten av att identifiera motiverande pedagogiska aktiviteter som kan stödja uppmärksamhet och engagemang hos elever som har flerfunktions–nedsättning. Avhandlingens sammantagna resultat visar hur teoretiskt motiverade och konkreta undervisningsaktiviteter med musiksamspel - med och utan sinnesstimulering och berättande - kan stödja uppmärksamhet och engagemang för elever med flerfunktionsnedsättning och att skolpersonal upplever det som ett användbart pedagogiskt verktyg i det dagliga skolarbetet. I synnerhet visar studien att MSMD kan vara ett arbetssätt som har positiva effekter för att stödja engagemang i lärandesituationer för elever som har flerfunktionsnedsättning.

Acknowledgements

A wise person once said: “There is no magical component in music, but there is”. From my experience as a musician educator and therapist, musical interaction provides opportunities to share expressions, intentions and emotions, including lots of joy. I would therefore like to thank all those that I have had the pleasure to meet in music therapy: students, clients, research participants, school staff members as well as relatives and colleagues for contributing to all moments of meeting in music. These have been, if not magical, then at least inspiring and meaningful!

On my journey in trying to become a PhD, I have had many persons without whom I would not have completed this book: Professor *Jenny Wilder*, my main supervisor, thank you for being such a great source of inspiration in research, one of the reasons why I wanted to become a PhD student in the first place! I am so grateful for your continual support, friendly encouragement and invaluable assistance along the way. My co-supervisors, Professor *Shakila Dada* and Professor *Simo Vehmas*, thank you both for your continual help with my PhD project and for generously sharing your vast knowledge in research. I am also grateful for the privilege to have participated in *the Research School in Special Education directed towards Early Intervention in Early Childhood Education*, which provided great resources and insight in developing research knowledge and skills that have been helpful for me in completing this work. Thank you Professors *Mara Westling Allodi*, *Sven Bölte*, *Henrik Danielsson*, *Mats Granlund* and *Eva Siljehag* for great support, knowledge and inspiration along the way. Also, I would like to express my deep gratitude to my current and prior fellow PhD students in the research school and at the *Department of Special Education* (no one mentioned no one forgotten!) for sharing experiences, giving support and continual encouragement during the way, which made the PhD student journey so much more meaningful and fun! I would also like to direct a special thank you to Professors *Henrik Danielsson* and *Samuel Odom* for their generous support and consultations concerning methodology in my first and second study. Thank you Professors *Henrik Danielsson*, *Mats Granlund* and *Helena Hemmingsson* for critically reviewing the thesis at my 50 and 90 % seminars and for providing valuable suggestions for revisions and future research.

I would also like to express my warm appreciation to all my former colleagues at the *Eldorado Resource Centre*, which both have contributed in

developing the approach of MultiSensory Music Drama and supported my work in so many ways. A special thank you also goes to my current colleagues at *the Swedish National Centre for Rett syndrome and related disorders*, thank you *Monika Dolik-Michno, Marianne Olsson, Åsa-Sara Sernheim, Magnus Starbrink, Lena Svedberg, Adam Thomas and Helena Wandin* for proofreading, continual support, encouragement and patience along the way.

I would also like to direct a special thank you to more informal groups of support: my friend and music therapy colleague *Anna-Karin Kuuse*, thank you for encouraging me to become a PhD student and for continual inspiration and reminding me not to forget to get out in the forest to get new ideas! *Ulf Jederlund*, thank you for sharing your knowledge and being an inspiration in early communication and music therapy. *Anna Rensfeldt Flink*, thank you for sharing the passion and challenges in research with people with S/PIMD. Also a warm thank you *Yu-Hsin Hsieh* for sharing and exchange experiences on this PhD student journey and for all your support and friendliness. *Sofia Wallin*, thank you for being my "partner in crime" during this PhD student adventure. Thank you for sharing it all – not the least beers and laughter! *Helena Wandin*, thank you for being my close and valued supportive companion, in clinical practice and in research.

I would also like to thank my very dear old friends, *Louise Andersson, Frida Blank, Maja Fredriksson, Anna Sandberg* and my sister *Sanna* for making life rich – through rain and through sunshine. To my loving *mum* and *dad*, thank you for your constant love and support and for giving me the courage to dare to dream. To my beloved children, *Lo* and *Majken*, thank you for making my life so much more fun, meaningful and rich. Finally, to my husband *Jakob*, my first reader and love, thank you for making life so beautiful and fun.

Context of the Thesis

The PhD project is a part of the Research School in Special Education directed towards Early Intervention in Early Childhood Education (Swedish Research Council 2017-03683). The research school is a collaboration between the Universities of Jönköping, Karolinska Institute, Linköping and Stockholm.

The thesis focuses on the context of education for students with S/PIMD who study an adapted curriculum focusing on daily living skills rather than academic achievement within the Swedish School Context.

The PhD project has been funded by the Department of Special Education at Stockholm University as well as the Swedish National Centre for Rett Syndrome and Related Disorders where the author is employed.

List of Publications

I. Johnels, L., Vehmas, S., & Wilder, J. (2023). 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. https://doi.org/10.1080/20473869.2021.1959875

II. Johnels, L., Wandin, H., Dada, S., & Wilder, J. (2024). The effect of MultiSensory Music Drama on the interactive engagement of students with severe/profound intellectual and multiple disabilities. *British Journal of Learning Disabilities*, *52*(1), 150-165. https://doi.org/10.1111/bld.12559

III. Johnels L., Wilder, J. & Vehmas, S. (2024). MultiSensory Music Drama with a student with multiple disabilities: A case study of teacher-researcher co-production. *Journal of Research in Special Educational Needs*, *24*(3), 530-541. http://dx.doi.org/10.1111/1471-3802.12649

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Abbreviations

AAC	Augmentative and Alternative Communication
CBRS	Child Behavior Rating Scale
EBP	Evidence-Based Practice
MSMD	MultiSensory Music Drama
PIMD	Profound Intellectual and Multiple Disabilities
PND	Percent of Non-overlapping Data
S/PIMD	Severe/Profound Intellectual and Multiple Disabilities

Introduction and Aims

Where there are humans, there is music. Across different times, geographical borders and cultures, music has been used to celebrate important occasions, express emotions and ideas and to strengthen cohesion between people. It has been argued that music is inherently human and provides an important function for communicative and social interactive purposes (Ilari, 2016).

Relatedly, storytelling has been used throughout the ages by groups of people with similar purposes to pass on traditions through generations and to create group cohesion through engaging and joyful activities (Penne et al., 2012).

The current thesis focuses on students with severe/profound intellectual and multiple disabilities (S/PIMD). The overarching aim of this thesis is to contribute with new knowledge regarding how musical interaction, with and without storytelling elements, may support students with S/PIMD with a focus on interactive engagement.

The thesis consists of three sub-studies. In the first study, a scoping review was performed of all peer-reviewed research published over the last 20 years regarding musical interaction with children and young people with S/PIMD. The study focused on mapping different features of the current evidence base, including those related to population characteristics, study designs, dependent variables (e.g. interactive engagement), reported effectiveness, scientific quality and promising components of musical interaction (study 1). Moving on from this, studies 2 and 3 focus on the development and scientific evaluation of a novel pedagogical approach called "MultiSensory Music Drama" (MSMD). MSMD combines relevant elements from musical interaction/therapy and multisensory storytelling. Study 2 aimed to evaluate the effects of the MSMD approach on the interactive engagement of three students with S/PIMD. In order to do this, a single case experimental design study was performed. Also, the social validity of the approach in a school setting was probed, as reported by school staff members. In study 3, a qualitative, longitudinal case-study approach was used to describe a researcher/teacher co-production process in developing, evaluating and refining MSMD lessons together with one student with S/PIMD. This was done through repeated stimulated recall-interviews, evaluation and joint planning. The goal was to provide an in-depth analysis of how the teacher related to, and challenged, the student during this particular pedagogical work.

Also, the teacher's perception of the social validity was investigated, as was the long-term feasibility of the MSMD approach in the school setting.
The overall research questions (RQ) of the thesis are:

1. What are the features of the current evidence base regarding musical interaction together with children and young people with S/PIMD? (study I).

2. To what extent is the MSMD approach effective in supporting interactive engagement in students with S/PIMD and perceived as feasible in a school context by school staff members? (studies II, III).

In order to provide a context and rationale for the thesis, definitions and characteristics of the target population are introduced in the following section. The purpose is to provide an overview on how the everyday functioning of this group of students may take form. Thereafter, the main outcome measurement, i.e. interactive engagement, as well as the theories and constructs that have guided the current work are presented. Then, the educational setting, including the organisation of educational activities, the learning goals as well as general teaching strategies used with the student population are described in order to provide a framework for the studies performed in the thesis. After that, prior empirical research on musical interaction and on storytelling with students with S/PIMD, along with a description of the background and constituent parts of the MSMD approach, are presented. Finally, epistemological assumptions relating to contrasting views on what constitutes "good" educational research within special education are discussed and related to the work carried out in this thesis.

Background

Severe/Profound Intellectual and Multiple Disabilities

In Swedish, the term "flerfunktionsnedsättning" is commonly used to describe the group of learners that is the focus of the current thesis (Socialstyrelsen, 2020; Wilder & Granlund, 2019). Many scholars have argued that research and evidence-based practices for people with the most profound disabilities are strikingly scarce, and that this population has been left behind in research about people with disabilities (Maes et al., 2021; Mietola et al., 2017; Nakken and Vlaskamp 2007; Rensfeldt Flink, 2022).

Internationally, different terms are utilised to refer to the current target group. In the context of (educational) research from Britain, the term "profound and multiple learning difficulties" is commonly used to describe learners with the most severe intellectual disabilities, complex comorbidities and extensive care needs (Ware, 2004). Also, in the context of research on small children (who may not yet have formal diagnoses), the term "significant cognitive and motor delay" is sometimes used (e.g., Dhondt et al., 2020; Van keer et al., 2019a). In 1996, the International Association for the Scientific Study of Intellectual Disability created a special interest research group about the target group in order to provide an international forum for the exchange of knowledge on best practices and scientific studies. This special interest research group utilises the term "profound intellectual and multiple disabilities" (PIMD). As defined by Nakken and Vlaskamp (2007), people with PIMD have (i) profound intellectual disabilities and (ii) profound motor disabilities. Also, additional severe or profound secondary disabilities or impairments are commonly present (Nakken & Vlaskamp, 2007). Bellamy et al., (2010) proposed a somewhat different definition: (i) profound intellectual disability and (ii) multiple disabilities including sensory and/or physical impairments and health problems (Bellamy et al., 2010). Because of difficulties in assessing cognitive abilities in persons with very low intellectual functioning (Maes et al., 2021; Nakken & Vlaskamp, 2007), many scholars have recently used the definition "severe/profound intellectual and multiple disabilities" (S/PIMD) to manage the difficulty of demarcation between profound and severe intellectual disability especially when an individual simultaneously has several other challenges (Griffiths & Smith, 2016; Rensfeldt Flink et al., 2023; Van Timmeren et al., 2017). This solution is adopted in the current thesis as well. To be consistent throughout the thesis,

the term S/PIMD will be used when referring to either one of the proposed terms described above.

The prevalence of people with S/PIMD in Sweden has clinically been estimated to be around 0.025% (Ölund et al., 2003). People with S/PIMD are dependent on others in all aspects of life. They need supportive relationships to facilitate daily living activities, but also to express their abilities, their will and their personalities. In addition, this group has several sensory-motor impairments and medical comorbidities (Axelsson et al., 2013; Nakken & Vlaskamp, 2002). Persons with S/PIMD constitute a heterogeneous group both concerning the aetiology of the impairment and their functioning, interests and behaviours. Their communication is often non-verbal, idiosyncratic and expressed by bodily movements, gestures and sounds (Griffiths & Smith, 2016; Rensfeldt, 2022; Wilder, 2014). Additionally, it is often hard to identify whether their communication is intentional (Wilder, 2014).

Theoretical Perspectives

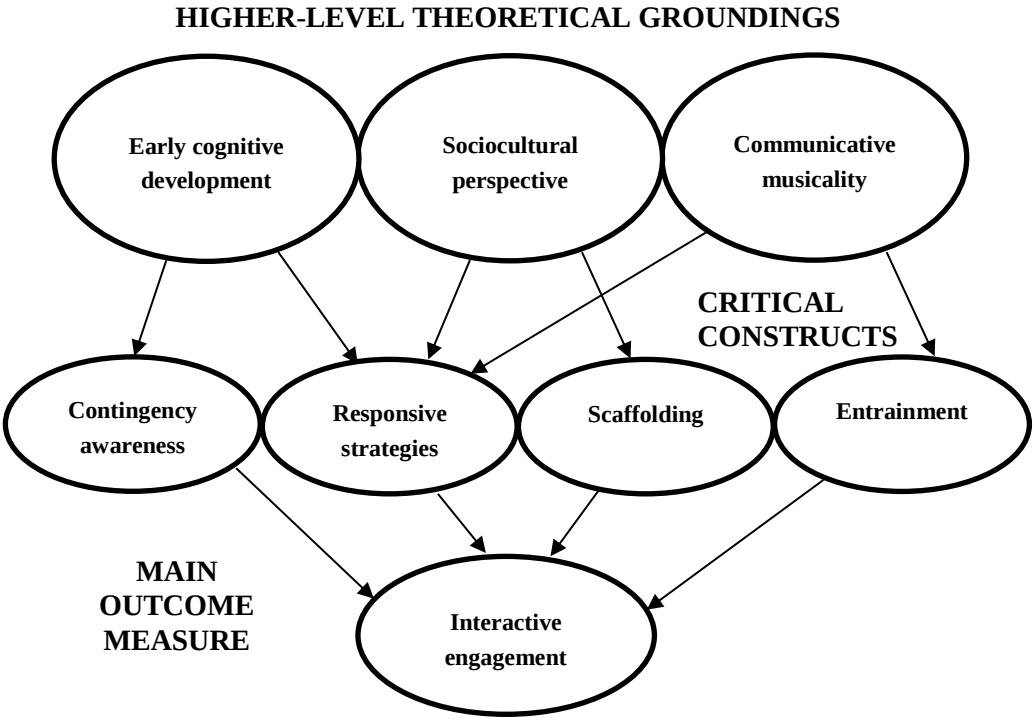
The value of an eclectic theoretical approach in special education research has been raised (e.g., Abbeduto & Boudreau, 2004; Odom, 2016; Pressley et al., 2023). As proposed by Odom (2016), "a variety of formal theories form the basis for practices in early childhood special education" (Odom, 2016, p.22), and the term "technical eclectic" is used by Odom to describe this approach to research. The current thesis has a similar eclectic approach. An overview of the theoretical framework and key theoretical constructs that have guided the work is provided in Figure 1. These can be understood at different levels of abstraction, from higher-level theoretical groundings to critical theoretical constructs that are more proximal in relation to the research questions and hypotheses that are being directly addressed. These theoretical groundings are seen to be influential on the main outcome measurement, which is interactive engagement. Therefore, interactive engagement will be discussed in relation to the highlighted theories in the text below.

Main Outcome Measure: Interactive Engagement

In this thesis, *interactive engagement* is considered a key outcome measure. The term "interactive engagement" has been used in research involving students with S/PIMD as an umbrella term for capacities such as attention, persistence, involvement, cooperation, initiation, joint activity, affect and alertness (Rensfeldt Flink et al., 2022: Van keer et al., 2017; 2020). In research, these complex, overlapping capacities and indicators are typically operationalised using observation coding schemes (ibid). It is further argued

that these capacities are considered so-called pivotal behaviours for development and learning (Kim & Mahoney, 2004; Mahoney et al., 2007; Van keer et al., 2017; 2020). Pivotal behaviours are behaviours that are central to the development of wide areas of functioning, such that a positive change in a pivotal behaviour potentially will produce improvements in a number of other outcomes. In the context of people with S/PIMD, Munde et al. (2012) specifically highlight alertness as the single most important prerequisite for learning and development and define alertness "as an individual's level of interaction and engagement with the environment, which becomes manifest and observable in the individual's behavior" (Munde et al., 2012, p. 6). Thus, for students with S/PIMD, focusing on activities and ways of interacting that promote interactive engagement is considered of key relevance to support learning and development more generally, i.e. interactive engagement is a pivotal behaviour.

Figure 1. Theoretical groundings and critical constructs for supporting the main outcome measure.



Early Cognitive Development

Firstly, theoretical underpinnings from cognitive and developmental psychology of how typically developing infants acquire cognitive and communicative skills (with the infant acting within early caregiver-child interactions) provide an important foundation for how the development of these skills can be fostered in students with S/PIMD (Bruner, 1985; Goldbart, 2018; Stern, 1977; Ware 2004; 2018). It has further been argued that several factors reciprocally support early cognitive development in both typical infants (e.g., Masek et al., 2021) and in children with S/PIMD (Van keer et al., 2017; 2019a; 2020), which will be detailed in the following section. To begin, in interactions between caregivers and infants, responsive strategies of the caregivers are considered imperative for communication development (Nind, 2007; Wandin et al., 2021). Examples of responsive strategies are waiting for the child's initiatives and providing time for responses, and interpreting sounds, movements and gestures as intentional and meaningful communication and responding accordingly (ibid). These strategies are

unconsciously and automatically performed in typical infant-caregiver interactions (Goldbart, 2018; Nind, 2007). However, it has been argued that patterns of social interaction are affected if a person has S/PIMD (Munde & Vlaskamp, 2014; Ware, 2004; Wilder, 2008). The complexity of their disabilities limits their ability to express themselves, and their communicative signals are commonly slower and very subtle (Dhondt et al., 2020; Olsson, 2004). In turn, this may result in the interaction partner being less responsive and also the one who takes the lead in interactional exchanges in a one-sided, asymmetrical manner (Dhondt et al., 2020; Ware, 2018). Although these patterns are often unconsciously performed by the interaction partner, they can result in a negative spiral when it comes to interaction and possibilities for stimulating early cognitive development for the person with S/PIMD (Ware 2004; 2018).

Also important for early cognitive development is alertness. As mentioned, alertness, attention and other indicators of the interactive engagement of the child are key enablers of learning and development (Munde & Zentel, 2020). In order to stay awake and alert, we need arousal, which we receive through our different senses and which energises our behaviours (Goldbart, 2018). Similarly to typically developing infants, individuals with S/PIMD often have low or fluctuating levels of alertness. Importantly, Van keer et al. (2017; 2020) showed that parents' responsive behaviours are significantly related to the child's interactive engagement. Although causal relations are hard to determine in cross-sectional analyses, the authors further argue that responsive strategies of the interaction partner may contribute to a chain of positive consequences, since they potentially both raise the interactive engagement in the person with S/PIMD, as well as motivate the interaction partner to be even more engaged and responsive in the interactive situation. Yet another crucial cornerstone in early cognitive development (emanating from Piaget's theories of stages in child development) is an increasingly sophisticated understanding of how and why things happen, and how cause and effect relations function. These lines of reasoning are related to the concept of "contingency awareness", i.e. the realisation that one's actions have consequences (Dunst et al., 2008) which is a prerequisite to intentionally participating in communication and interaction (Goldbart, 2018). In typically developing infants contingency awareness usually develops around six months of age through exploration of their own body (e.g., putting the thumb in the mouth) or later on with exploration of objects (pushing a toy which makes sounds) and in interaction with other people. Due to motor and sensory impairments in children with S/PIMD, they will commonly not be able to explore their environment in the same manner as typically developing infants and children do. This risks limiting their opportunities to receive feedback on their actions and to experience contingency awareness (Goldbart, 2018).

In the current thesis, student interactive engagement and responsive and contingent interactions are seen to influence each other in a reciprocal manner and to conjointly influence student development and learning.

Sociocultural Perspective

Secondly, a sociocultural/Vygotskian perspective is important in this thesis, where learning is seen as an inter-personal event. A learner is able to master more with support from a more skilled person (e.g. a teacher) than he or she can master on his/her own, which corresponds to the description of being in the zone of proximal development for learning (Vygotsky, 1978). In the context of students with S/PIMD, this is indeed important, since it shifts the focus from learning and development as something that is fixed in the student, towards a view that learning is something that is co-created by the student together with the teacher or other supportive people in the social environment (Ware & Healey, 2018). A related concept is *scaffolding*, in which the teacher supports the student in the learning situation, which enables the student to accomplish much more than he or she would on their own (Wood et al., 1976), and the teacher then gradually reduces the support as the learner becomes more independent. In the context of students with S/PIMD, this can be related to the teacher's strategies to model actions and behaviours for the students before they can perform them themselves. Relatedly, the use of augmentative and alternative communication (AAC) may be construed as a form of scaffolding. Many scholars and practitioners have highlighted the benefits of using AAC as a means for communication and communication development with people with complex communication needs, including people with S/PIMD (Dada et al., 2021; Goldbart & Caton, 2010; Wallin et al., 2024; Wandin et al., 2023). AAC includes both unaided AAC (e.g., manual signs, gestures, facial expressions) and aided AAC (e.g., communication boards, speech output devices) that can assist the person with S/PIMD to make himself/herself understood and/or to understand others' communication (Dada et al., 2021).

Given the dependency students with S/PIMD have on people in their close environment, the balance between support and challenge is crucial in order to provide enjoyable learning situations (Nind, 2007). More generally, Bruner (1985) suggested that in good learning situations, neither the learner nor the teacher can be too controlling or passive. Related to the concept of scaffolding, the teacher needs to facilitate the learning activities in ways that make the learner actively engaged, which is a key aspect of the present work.

Communicative Musicality

Thirdly, the psychobiological framework of communicative musicality (Malloch & Trevarthen, 2009; Malloch, 1999) has been influential in the thesis. The theory of communicative musicality originates from research and practice in the field of music therapy and from research on infants, and proposes that all humans have an innate ability to use musical actions as meaningful expressions; thus, according to Trevarthen (2012) communicative musicality refers to the "human impulse to create and share music" (Trevarthen, 2012, p. 259). It is indeed proposed that the earliest interactions between infants and their caregivers have a musical structure and are characterised by musical elements, such as pulse, rhythm, melody structure and dynamics, (Bunt & Pavlicevic, 2001). Philips-Silver and Keller (2012) describe how when people play music and dance together, they commonly tune into a shared rhythmic timing and affective state. This synchronisation of movements and feelings is also called entrainment. Malloch and Trevarthen (2009) further suggest that communicative musicality has an important evolutionary function in the sharing of cultural narratives and emotions amongst humans. Thus music-making is inherently social according to the theory of communicative musicality. Relatedly, in the context of music therapy with people with autism and Rett syndrome (including people with S/PIMD), Wigram and Elefant (2009) emphasise that the innate communicative musicality is indeed evident in these populations. Importantly, music may provide an important means of expression: "when meeting a speechless client, the music therapist has the tools to promote communicative musicality, thus enabling a person to give their meaning a sound, and to sense that it has been received" (Wigram & Elefant, 2009, p. 442). Similarly, Bergström-Isacsson (2011) emphasises that music attracts a significant interest and elicits communicative responses to a large degree in persons with Rett syndrome, including people with S/PIMD.

In sum, these theories, constructs and strategies have all guided the research in the thesis.

Education for Students with S/PIMD

In this section, the organisational structure of education for students regarding setting and curriculum goals is presented. Thereafter, specific learning strategies used with the student population are presented.

Educational Setting and Learning Goals

In a larger historical perspective, students with S/PIMD in Sweden were considered as "uneducable" and they were placed in institutions or segregated

environments (Berthén, 2007; Östlund, 2012). In 1967, legislation (SFS 1967:940) made education for students with S/PIMD mandatory in Sweden. Nowadays, education is compulsory for children from the age of six.

For students with intellectual disabilities who are not expected to reach learning goals within the general school curriculum, there are two adapted forms of education: (i) education for students with intellectual disabilities, where students with mild to moderate intellectual disabilities receive their education and where the school subjects are similar to the general Swedish compulsory school but with more flexibility when it comes to progress and with more individualised learning goals; and (ii) education for students with more severe-profound intellectual and developmental disabilities where the curriculum is more related to daily living skills than to academic subjects, mainly in relation to arts activities, communication, motor abilities, activities of everyday living (with the aim of developing the student's skills related to managing everyday life routines) and practical applications of science and technology (Skolverket, 2024).

The current thesis focuses on students studying the latter educational curriculum. The school setting where students with S/PIMD generally receive their education, commonly consists of a heterogeneous group of students and the learning activities are typically organised in small classes with a high staff ratio (Östlund, 2015), often 1:1.

Research on Learning and General Teaching Strategies for Students with S/PIMD

It has been argued that educational research targeting students with S/PIMD is scarce (Arthur-Kelly et al., 2008; Hardesty-Jaynes, 2021; Maes et al., 2020; Munde & Zentel, 2020; Nind 2007; Ware, 2004; 2018). Relatedly, the National Agency for Special Needs Education and Schools in Sweden has emphasised that educational materials (i.e. activities, interventions, teaching strategies and materials) targeting this group of learners are very limited (Gröndahl, 2014).

It has further been discussed whether students with S/PIMD have educational needs that call for specific strategies and approaches in learning activities (Maes et al., 2020; Ware, 2004). Ware (2004) points out that these students spend much less time in attentive states, which affects their ability to learn. Also, the amount of voluntary actions and behaviours (attentive rates) is commonly low. In combination with differences in information processing and memory, special teaching strategies are warranted (Ware, 2004).

A few educational strategies have been customised and empirically evaluated for this group of learners specifically (Goldbart & Caton, 2010; Munde & Zentel, 2020; Nind & Strnadová, 2020; Ware, 2004). In large part,

these strategies closely mirror key concepts raised in the theoretical background of the thesis, and include:

- Approaches based on caregiver and infant interactions. Here responsive and pre-verbal interaction techniques are used, e.g., mirroring vocal sounds, gestures and movements of the student as well as tuning into the student's tempo and interests and finding a joint focus in activities (Nind & Hewitt, 2012). In an Australian context, the approach of intensive interaction was evaluated in an intervention study over 30 weeks for students with S/PIMD and autism. Analysis of video recordings on student involvement showed increases in the students' joint focus, positive affect and interactive involvement as compared to the baseline phase (Barber, 2008). A similar approach is the responsive environments approach (Ware, 2012). In a school-based research project, school staff members received coaching on how to become more responsive, which had a positive effect on the students' becoming more active in the interactions (Ware, 2012).
- "Cause and effect" activities, including the use of microswitches: The aim of these activities is for students to develop an understanding that their actions may affect objects and people in the environment (e.g., Lancioni et al., 2009). For instance, pressing a microswitch (i.e. an assistive technology device that may be activated with minimal effort with the use of, for instance, the head or a finger) which is connected to a music recorder or computer software, may enable a student to start the music and potentially understand the causality in the relation between these events. In their systematic review of microswitch technology to support self-determined responses in students with S/PIMD (e.g., choice-making or getting access to preferred stimuli), Roche et al. (2015) found 18 experimental studies, involving 45 participants, which all reported experimental data supporting the use of microswitches to enhance self-determination tasks for these students.
- Using objects as a reference and symbolic approaches: This means that a given object or prop is consistently linked to an activity, for instance, whenever it is time for a music lesson, a small drum is handed to the student. This is intended to signal what is about to happen for the student and to offer the student greater control. Two objects may also provide the student with the possibility of making choices (Goldbart & Caton, 2010). Also, symbolic approaches, such as pictures or photos, may be used as representations of the events in the world, in order to help the student to understand what will happen. Many different forms of symbols may be applied, such as books, individual cards or schedules (Goldbart & Caton, 2010). Object-based and symbolic approaches could be related to the earlier mentioned concept of AAC. It is often recommended that people in the social environment use aided AAC modelling (i.e. combining their

speech with pointing at picture symbols) when implementing AAC with students with S/PIMD (Wandin et al., 2023).

- Motivating learning activities including music, arts and storytelling: Approaches using music and other creative arts-based approaches and storytelling with props have been seen to engage students with S/PIMD (McFerran and Shoemark, 2013; Rainey Perry, 2003; ten Brug, 2015). Creative arts-based approaches (e.g., dance and drama) are mentioned in the practitioner literature (Butté & Unkovich, 2009; Vickers, 2005). Narrative and storytelling approaches, using combinations of language and multisensory props to construct a narrative, have also been seen to support interactive engagement in the population (Preece & Zhao, 2015; ten Brug, 2015). As mentioned, music interaction and therapy is commonly used in practice and explored in research (McFerran and Shoemark, 2013; Rainey Perry, 2003). Interestingly, similarities between music therapy and approaches using strategies from caregiver-infants interactions are highlighted, specifically to attentively tune into the student's initiatives, expressions, and tempo in musical activities (Goldbart & Caton, 2010). At the same time, the development of musical dialogues may not always occur automatically in children with S/PIMD due to sensory processing, delayed responses, uncontrolled or limited movement abilities. Hence, certain strategies are warranted to counterbalance this. Firstly, interjecting and making pauses are two opposite strategies mentioned. Interjecting means filling in the gap when the person makes a space in their communicative music, as opposed to making pauses and waiting for the person to take the initiative or respond. Secondly, the strategies of modelling and matching are suggested. Modelling refers to the therapist (or teacher) demonstrating something in a way that engages the person, and matching refers to picking up something (an action, sound or movement) that the person does and playfully imitating it with musical expressions. Thirdly, the function of music as a predictable framework for interaction is highlighted. Predictable and sustained sounds, variations of pitch and dynamics, as well as and routines and predictable turns, are all examples of musical elements that help the person to stay focused, engaged and able to predict what will happen, which in turn may enable him or her to join in (Wigram & Elefant 2009; Wigram, 2004). Prior research on music-based approaches and storytelling with the target population will be described in greater detail in the following sections.

Swedish (special) educational research focusing on students with S/PIMD is scarce. One important exception investigated contextual and interactional conditions of educational practices for students with S/PIMD (Östlund, 2015).

The findings highlighted the need for balanced teacher-student interactions and the need for a child-centred focus.

Given the amount of evidence that emphasises the importance of opportunities for students with S/PIMD to participate in balanced interactions where they have possibilities to take the initiative and be active (Munde & Vlaskamp, 2014; Shalev & Hetzroni, 2020; Ware, 2004; 2012; 2018), the concept of contingency awareness is of educational interest (Ware, 2004). This concept refers to an individual's realisation that one's actions have consequences (Dunst et al., 2008) which is a prerequisite to intentionally participating in communication and interaction (Goldbart, 2018). This is especially important since previous research has highlighted that children with severe disabilities, including students with S/PIMD, are at risk for developing learned helplessness, i.e., becoming passive rather than active learners (Dunst et al., 2008; Östlund, 2015). Responsive and contingent-rich everyday learning activities and environments are suggested to provide opportunities for development and learning for students with S/PIMD. Such activities and environments provide the student with opportunities to initiate actions and to gain opportunities to make choices. Also, students are given sufficient time to respond and to take a more active role in interactions (Ware, 2004; 2012; 2018). Hence, the knowledge, sensitivity, and attentiveness of people in the social environment (e.g., school staff members) become crucial to achieving these learning situations (Munde & Zentel, 2020; Nind, 2007).

Prior Research on Musical Interaction with People with S/PIMD

In many parts of the world, including Sweden, musical interaction and music-therapeutic approaches are integrated into education for these students (Bertén et al., 2022; McFerran & Shoemark, 2013). In the educational context for the target group, music is integrated into a broader curriculum on arts education (Sjöqvist et al., 2021). Musical interaction is defined as situations in which at least two persons are involved in joint music-making. By definition, music therapy involves a music therapist taking part in the musical interaction, thus musical interaction is compared with music therapy, a broader term referring to activities that can take place with different interaction partners. When discussing practice, research and theories related to active musical activities with primarily non-musical goals (e.g., communication, social interaction and engagement), the distinction between musical interaction and music therapy is not considered central.

Since the late 1960s music therapy or musical interaction with people with S/PIMD have had the aim of supporting non-musical goals such as non-verbal communication, social relatedness and engagement (Nordoff & Robbins,

1971; 2007; Wheeler, 2013; Wigram et al., 2002). In this tradition, it is assumed that we all have an innate musicality and that communication skills and motivation may lead not only to musical development but to more general socioemotional and cognitive development (Malloch & Trevarthen, 2009; Wigram et al., 2002). Musical interaction is commonly performed in structured, playful and pre-verbal music activities using mirroring, taking turns, anticipation and tuning in to a shared focus. A relatively large body of literature shows that music therapy is a potentially promising way of working to support interactive engagement behaviours (Kantor, 2020; McFerran & Shoemark, 2013; Rainey Perry 2003; Thompson & McFerran, 2015; Wheeler, 2013). For instance, Thompson and McFerran (2015) investigated the communicative behaviours of four adolescents with S/PIMD during music therapy sessions (as compared to play sessions) for six months. Using a coding scheme of potential communicative acts, their findings revealed more communicative behaviours during music therapy than during regular play sessions, although both conditions were seen to engage the students. Similarly, Rainey Perry (2003) investigated the communicative behaviours during music therapy with ten young students' with S/PIMD, where student actions during the music therapy were related to their general communicative level. It was found that the students' general communicative levels were reflected during music therapy, i.e. students at a more advanced communicative level (but still on a pre-symbolic level) were able to initiate more and stay engaged for longer periods. For the students at the earliest communicative level, repetition and a set structure in music activities were seen to support interactive engagement. Also, favourite musical instruments, and "suspense-heightening" pauses made by the music therapists were seen to engage the students more than was usually seen in school activities. Interestingly, the author suggested that the potential of music to sustain interaction is important since many students with S/PIMD need time to respond or initiate interactions. Hence, it is argued that music creates a supportive framework for interaction.

However, there are no known studies where the quality and nature of research on musical interaction with children and youths with S/PIMD have been evaluated and summarised systematically.

Prior Research on Storytelling and Drama with People with S/PIMD

Storytelling and other dramatised expressions involve direct interactions and engage attention, listening and interpretation (Grove, 2013) and can be used to entertain, instruct and describe and process events (ibid). For people with S/PIMD, who are less likely to appreciate regular stories because of their severe/profound intellectual disabilities, the approach of storytelling

supported by sensory stimuli (i.e. multisensory storytelling), was developed in the 1980-1990s (Fornefeld, 2012; Lambe & Hogg, 2013). The practice of multisensory storytelling is diverse, with several different ideas concerning the content and aim of the stories, as well as the way they are presented (Fornefeld, 2012; Grove, 2013) including in theatre formats (Brigg, 2012; Webb, 2012) in active groups (Fuller, 2013) or one-to-one storytelling sessions (Lambe & Hogg, 2013; ten Brug, 2015). Multisensory storytelling is to a large extent an individualised activity, where the form and content of the story are adjusted to the person with S/PIMD (Lambe & Hogg, 2013; ten Brug, 2015; Young et al., 2011). Many scholars emphasise the value of presenting the story in a consistent way and providing the individual with S/PIMD opportunities to explore the multisensory objects in order to support attention and alertness (Fuller, 2013; ten Brug, 2015). Interestingly, Preece and Zhao (2015) investigated the use of multisensory storytelling in five British schools for students with S/PIMD and found that the teachers continuously adapted and adjusted the stories in accordance with their pedagogic professional judgements of what would be the most appropriate for the student at the moment. The authors argue that for the approach of multisensory storytelling to be feasible in an educational context and in relation to curricular goals, a more fluid and adaptable application of the approach is needed (Preece & Zhao, 2015).

Although many authors state that a primary goal of multisensory storytelling is to provide an enjoyable activity for people with S/PIMD (Lambe & Hogg, 2013; ten Brug, 2015), research has shown that multisensory storytelling can enhance social interaction, engagement and attention in this population (Foloştină et al., 2015; Preece & Zhao, 2015; ten Brug, 2015; ten Brug et al., 2015; 2016; Young et al., 2011). In a study by Foloştină et al. (2015), an intervention that included the use of drama therapy and storytelling was used with the aim of developing social competences and engagement in institutionalised adults with S/PIMD. The results indicated an increase in social and communication skills; however, the qualitative non-controlled design highlights that need for further research. In a study by Young et al. (2011) behavioural observations during storytelling sessions showed positive changes in engagement with the story in seven out of eight participants with S/PIMD. In her thesis, ten Brug (2015) similarly showed that multisensory storytelling can increase listener's attention and engagement and support interaction between storyteller and listener.

Interestingly, adding music during interactive storytelling has been shown to be positively related to engagement and learning in classroom situations with children with autism (Carnahan et al., 2009). Specifically, these authors showed in a study with an ABCAC single-case-experimental design with six children that combining music and storytelling elements appeared to be linked with particular increases in child engagement for several children. Fornefeld (2012) the founder of the German application 'mehr-Sinn Geschichten' (i.e.

storytelling with all our senses) similarly emphasised the use of music and the prosody of language in multisensory storytelling, since this is described to support a process of sensory-aesthetical comprehension in individuals with S/PIMD. These findings might suggest that the combination of props-supported storytelling, dramatised interaction and musical interaction may confer added value compared with any of these activities and interventions considered alone. To date, however, research has been lacking when it comes to the systematic joint use of dramatised and multisensory storytelling and musical interaction in supporting interactive engagement in individuals with S/PIMD in educational contexts. The idea that combining these approaches might have added pedagogical value compared with each approach considered separately (Carnahan et al., 2009; Fornefeld, 2012), is in keeping with observations made in special education more generally when it comes to the importance of multi-component interventions (e.g., Wolff, 2011). Relatedly, in the context of children with S/PIMD, Van keer et al. (2019b) argue that in addition to encouraging interactions, the combination of multiple stimuli elicits higher levels of interactive engagement behaviours in children. Given the promising research findings, this calls for further research to explore and evaluate the potential benefits of using dramatised multisensory storytelling with musical support with students with S/PIMD.

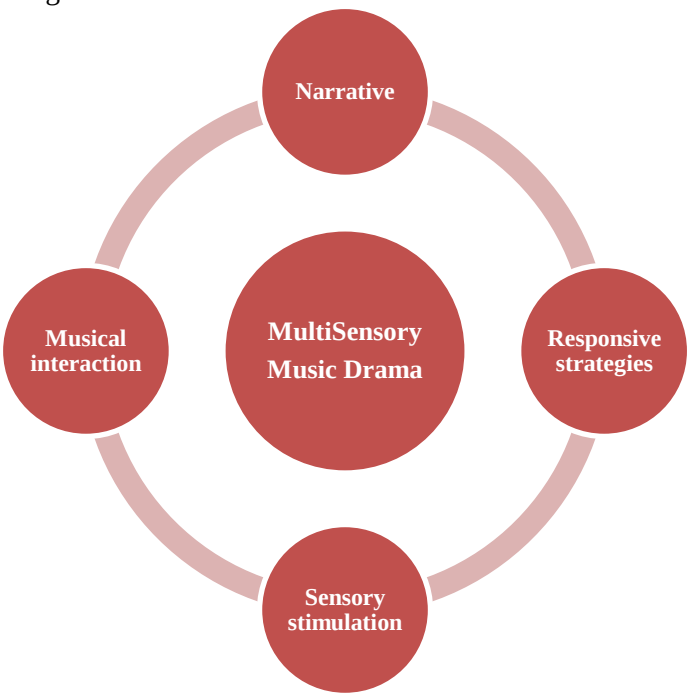
The Pedagogical Approach of MSMD

Two of the studies in the thesis focus on a custom-developed pedagogical approach called MultiSensory Music Drama (MSMD). In MSMD, musical interaction is systematically combined with dramatised sensory stimulation. MSMD is based on principles from musical interaction and multisensory storytelling and was developed by the author and her colleagues at an activity, education and culture centre for people with S/PIMD in Gothenburg. The goal of the MSMD approach is to support the interactive engagement of students with S/PIMD.

The central elements of MSMD are narrative, responsive strategies, sensory stimulation and musical interaction, as visualised in Figure 2. In MSMD, an individualised drama is created to fit the student's abilities (i.e. motor, communication, sensory abilities) and the student's preferences concerning favourite props, games, music as well as goals related to the student's syllabus. Also, the sensitive responsivity of the interaction partner is emphasised in the approach (i.e. the interaction partner is very attentive and responds to the reactions from the student, adjusts his or her energy and "tunes in" to the student). Additionally, the possibility for the student to make choices within structured, contingent activities is important to consider in order to provide prerequisites for student anticipation and agency. An MSMD lesson

is recommended to last approximately 15 minutes, although this also needs to be adjusted to the individual student.

Figure 2. Visualisation of the central elements of MSMD. In the MSMD approach, musical interaction and sensory stimulation as well as responsive partner strategies are used within a narrative. Based on Johnels et al., 2024b.



Evidence-Informed Practices and Co-Production in Educational Research

In this section, the epistemological assumptions that have guided the work of the thesis are described. The current thesis favours the idea of trying to apply the scientific method (i.e. raising hypotheses and testing them against empirical data) to educational matters in order to enhance understanding and support improvements in practice.

The framework of evidence-based practice (EBP) emerged in the 1970s with the purpose of improving clinical practice in medicine (Leach, 2006). In more recent years, an emphasis on identifying practices that have scientifically supported effectiveness has been expressed also in educational practices (Mitchell & Sutherland, 2020; Odom et al., 2003; Shavelson & Towne, 2002). However, within current educational research, there are discussions and disputes regarding the role of EBP (Biesta, 2007; Hargreaves, 1997; Imray et

al., 2023). On the one hand, advocates of EBP argue that interventions and approaches used in educational practice should be scientifically evaluated for their effectiveness, preferably through large-scale randomised control studies or other experimental or quasi-experimental studies, in order to find the most effective interventions for specific learning outcomes (Davies, 1999; Mitchell & Sutherland, 2020). On the other hand, opponents have questioned the appropriateness of EBP in educational contexts, arguing that EBP only favours "technocratic" and positivistic perspectives on research (effectiveness), while failing to include normative aspects and more cultural (interpretative) perspectives in educational research (Biesta, 2007; Evaldsson & Nilholm, 2009). Also, it has been argued that the framework of EBP reinforces a top-down relation between research and practice, which fails to acknowledge the expertise and professional judgements by teachers with regard to the best choice of teaching strategies appropriate for a specific group of learners under specific conditions (Biesta, 2007; Sanderson, 2003). Biesta (2007) further states that the frequent discussion regarding EBP has led to a more nuanced terminology, where terms such as "evidence-informed" or "evidence-aware" practices are preferred over EBP.

Munde and Zentel (2020) summarised in their review article the current state of EBP for teaching and learning for students with S/PIMD. They conclude that the major methodology used in empirical studies with the population is single-case-experimental design studies. Also, they argue for a broader understanding of the term "evidence", such that it should also include expert opinions, professional consensus and proxy perspectives (Munde & Zentel, 2020). Triangulating methods and paradigms might be especially relevant "if the research question is not merely 'Does it work?' but also involves issues of process, feasibility, appropriateness, and meaningfulness" (Onghena et al., 2019, p. 436).

As reported in many research and practice fields, including in special education, there is a gap between research and practice (Greenwood & Abbot, 2001). At least in part, this is described to be due to a clash of cultures and contrasting views on what constitutes good research and useful knowledge (Martin, 2010). Greenwood and Abbot (2001) give the following explanations for the identified lack of special educational research in practical settings. Firstly, it has been reported that research and practice communities are separated. Secondly, the relevance of educational research is commonly perceived as low by practitioners. Thirdly, there is a failure in research to produce feasible interventions that are useful in classroom settings. Finally, it has been reported that there is a lack of opportunities for practitioners and researchers to receive input from each other which otherwise could contribute to professional development (for both the researcher and the practitioner).

Co-production in research entails a collaboration between researchers and practitioners when conducting research studies (Martin, 2010; Norström et al., 2020). Co-production has the potential to decrease the research-practice gap

by providing a research focus on activities carried out in practical settings (Martin, 2010). Conversely, co-production may enhance the prospect of research being found useful in practical settings. However, it is also important to safeguard academic freedom and integrity in the research process. Martin (2010) described five different types of co-production in research that satisfy these complex goals to varying degrees. In type 1 research, practitioners are involved to a minimum extent – e.g., practitioners as informants. In type 5, practitioners are involved to a maximum extent, (e.g. practitioners function as co-researchers involved in design and dissemination. Similarly, educational design-based research has the aim of refining theory and making a difference in practice through collaboration performed in iterative cycles with relevant stakeholders (Armstrong et al., 2022; Getenet, 2019).

The thesis is informed by all these perspectives. In study 2, a single-case-experimental design study was performed to scientifically test the effectiveness of the MSMD approach on student interactive engagement as compared to a control activity. At the same time, the more nuanced understanding of "evidence" as proposed by some critics of a "traditional" understanding of EBP is arguably very important to include and consider (e.g., Biesta, 2007) – recognising that valuable knowledge contributions include aspects such as appropriateness, meaningfulness, and specificities in relation to certain persons and contexts (Munde & Zentel, 2020; Onghena et al., 2019). In study 1, the complementary benefits of both qualitative and quantitative research methods were therefore acknowledged, and thus the study included a qualitative content analysis of promising components of musical interaction. Additionally, a quality appraisal was applied to evaluate the effectiveness of the musical intervention in the context of the systematic work of a scoping review. Also, the current work is generally favourable to educational research that is done in close collaboration with practitioners, and collaborative research might be one avenue for bridging some of the gaps between research (whether more positivistic or interpretative) and educational practice. In order to develop, evaluate and refine MSMD lessons, study 3 consisted of a qualitative, longitudinal case study in which the researchers had a close collaboration and co-production together with an experienced special education teacher and her student. In addition, the long-term feasibility and social validity of this approach in the everyday school work of the teacher was investigated.

Methods

Methodological Overview

In the studies of the thesis, both quantitative and qualitative methods were applied to answer the research questions. The current work has been inspired by researchers who argue for the value of methodological pluralism, which "involves finding value in a variety of sources of information, including believing that no research method is inherently superior to any other" (May et al., 2017, p. 1) and that the merit of a methodological choice is dependent on the research question at hand. In table 1 an overview of designs, participants, instruments and data analysis used is presented.

Table 1. Overview of the three studies in the thesis

	Title	Design	Participants	Instruments	Data analysis
I	Musical interaction with children and young people with severe or profound intellectual and multiple disabilities: a scoping review	Scoping review of peer-reviewed articles in English in the years 2000-2020 regarding musical interaction and children with S/PIMD.	n/a	Evidence-based librarianship critical appraisal checklist. PRISMA	Narrative synthesis. Inductive content analysis.
II	The effect of MultiSensory Music Drama on the interactive engagement of students with severe/profound intellectual and multiple disabilities	Single case experimental design study. Lessons with MSMD were compared with a control activity (picture book reading) in a Single-case-experimental design (ABAB design) study where the students' interactive engagement was assessed.	3 dyads of students with S/PIMD (8-12 years old) and their school staff members (one special education teacher and one teacher assistant per student).	Child behaviour rating scale (CBRS) and Engagement model: each occasion (students). Teacher post-intervention acceptability and importance of effects survey. (School staff).	Visual analysis. Statistical analysis (Percentage of non-overlapping data and Tau-U) Intra class correlation was used to examine inter-rater reliability.
III	MultiSensory Music Drama with a student with severe/profound intellectual and multiple disabilities: A case study of teacher-researcher co-production	Case study design. The co-production process and the implementation of MSMD with a teacher of a student with S/PIMD and Rett syndrome were described and evaluated.	One student with S/PIMD and Rett Syndrome. One special education teacher.	Interview checklist, follow-up interview. Engagement profile & scale. Teacher post-intervention acceptability and importance of effects survey. Field notes.	Inductive content analysis.

Study I

Design and Procedure

A scoping review with the aim of summarising and evaluating the current research regarding musical interaction with children and young people with S/PIMD was performed. Scoping reviews are commonly applied to provide a comprehensive overview of an emerging research field and to draw on evidence from different research methodologies (Peters et al., 2020). The review process in scoping reviews is commonly iterative, and changes and critical distinctions may appear during the review process (Oudshoorn et al., 2021). Commonly, much time is spent on screening due to broad search strings. In the current study, the inclusion criteria related to musical interaction focusing on children and young people with S/PIMD (between birth to 21 years of age) in peer-reviewed empirical articles written in English in the years 2000-2020. The exclusion criteria were: if the studies did not report any empirical data or if the musical interaction only involved listening to recorded music. Potential studies were identified through computer-assisted searches in the PsychINFO, Scopus and PubMed databases. Also, papers citing a prior systematic review on children and youths with disability and music (Brown & Jellison, 2012) were screened. Reference lists and relevant review articles were screened to find additional relevant articles. The study selection process followed the stages of the PRISMA guidelines (Liberati et al., 2009): (1) identification, (2) screening titles and abstracts, (3) eligibility and (4) inclusion. The three authors of the article screened all titles and abstracts, and two of the authors screened all full-text articles.

Data Analysis

A narrative synthesis was applied to summarise and evaluate the aggregation of research. The included studies were summarised using a standardised form related to the posed research questions which detailed the following variables: participant characteristics, study design and data collection methods, musical interaction activities, abilities and behaviours in focus, reported benefits/effectiveness, promising components and quality appraisal of the studies. In relation to the research question concerning reported promising components of musical interaction activities reported in the studies, we were analytically informed by the inductive content analysis approach used in systematic reviews (Elo & Kyngäs, 2008; Mikkonen & Kääriäinen, 2020). The quality appraisal was performed using the adapted Evidence-Based Librarianship Critical Appraisal Checklist. The included studies were rated in accordance with the guidelines described in Glynn (2006) and Van keer and Maes (2018). Following recommendations from Tarvainen et al. (2020), two

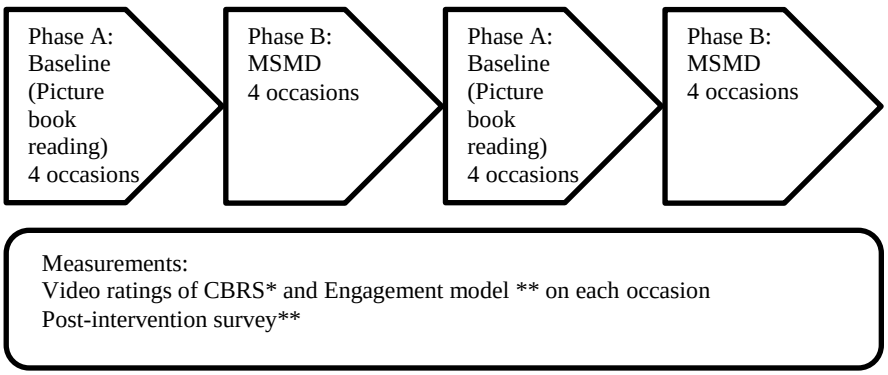
of the authors of the study performed an inter-rater reliability check on a randomly chosen 20% of the studies. The inter-rater reliability calculation resulted in point-by-point agreement of 86%.

Study II

Design

The study applied a single-case-experimental design in order to identify a possible effect of MSMD (the independent variable) compared to a control activity on student interactive engagement (dependent variable). The study adhered to principles for single-case-experimental design studies (Kratochwill et al., 2013) and the independent variable was systematically manipulated in an ABAB design. The 'A' phases (the control/baseline condition) consisted of joint picture book reading and the 'B' phases of MSMD lessons. The study included three attempts to demonstrate an intervention effect, i.e. between conditions A1 and B1; B1 and A2; A2 and B2. Each phase consisted of four lessons and a total of 16 lessons were carried out individually with each of the three participating students. The order of the phases for each student was randomised to lower the risk of order bias (http://slump.nu). Thus, two of the students started with four lessons of joint reading and one student started with four MSMD lessons. See Figure 3 for a visualisation of the design of the study.

Figure 3. Visualisation of the ABAB design used in study II (Or vice versa for one student, i.e. BABA).



*Rated by the second author of the study with a reliability check by an independent rater on 33% of the data points.
**Rated by the school staff members.

Participants and Setting

Three students with S/PIMD (8-12 years old) and six school staff members (one special education teacher and one teacher assistant per student) participated in the study. Inclusion criteria for the participating students were: 1) studied the curriculum focusing on daily living skills for students with severe/profound intellectual disabilities (In Swedish: kursplan ämnesområden); 2) were 7-12 years old; 3) had no or minimal verbal language and S/PIMD (as reported by parents). All teachers were fully qualified special education teachers and the teacher assistants had a secondary educational background.

As a first step, principals and teachers working in schools for students with S/PIMD in Gothenburg in Sweden were contacted. A special education teacher contacted us and showed interest in participation. To provide information about the study (i.e. information about the pedagogical approach of MSMD, participant selection criteria, data collection procedures etc.) a video conference with all teachers working at the school was held. Three teachers (from three of the school's four classes) volunteered to participate. They each suggested one student from their class who matched the inclusion criteria. Also, each teacher suggested a teacher assistant from their class for participation. The teachers contacted the parents of the students and provided them with contact information for the researchers as well as information about the study. Parents provided their informed written consent for their children to participate. Additionally, they provided information about their child's diagnoses and general and musical preferences (e.g. games, toys or songs they liked) and detailed their child's communication modes and need for different communication support. All the participating students met the S/PIMD inclusion criteria. Student 1 was a nine-year-old boy with cerebral palsy and microcephaly. He was non-ambulatory. He used eye contact, sounds and facial expressions and a few words in his communication. He was socially interested and he liked music, being outside, playing with water and soap bubbles. Student 2 was an eight-year-old girl with autism and epilepsy. She was ambulatory with support. She used eye gaze, sounds and gestures to communicate and had no verbal language. She was socially interested and liked music, movement, massage, playing with water and joint reading. Student 3 was a twelve-year-old boy with epilepsy. He was non-ambulatory. He used eye gaze, sounds and facial expressions to communicate and had no verbal language. He was socially interested and liked to be outside, children's songs, physical contact and joint reading.

MSMD and Control Activity

Prior to the data collection, an on-site meeting with each student's two participating school staff members (teacher and teacher assistant) was held.

The interests, strengths and communication modes for each student were summarised. Three examples of MSMDs were presented to the school staff members and each pair chose one MSMD for the student they worked with, which subsequently was individually adapted to the student (e.g. choosing songs, activities, props to explore and communication aids). Also, a pictorial booklet was created for each student. Lessons with joint picture book reading were used as the control activity. This control activity was chosen since it was a pedagogical activity that was commonly used with the students. Responsive strategies, pictorial support and a narrative element were applied in both conditions, whereas musical interaction activities and props-based sensory stimulation were used only in MSMD. In Figure 4, examples of materials used for one of the students in the different conditions are detailed.

Figure 4. Materials used for one of the students in study II. Based on Johnels et al., 2024a. To the left are the books that were used in the control activity and to the right are the materials that were used during the MSMD lessons.

Data Collection and Measures

The data collection was carried out in the school over two consecutive months during school hours. The lessons were performed individually with each student by the author who is a trained music educator and therapist. One of the students' school staff members was always present during the data collection to interpret communicative signals from the student and to detect if the student showed signs of discomfort. The lessons were carried out once a day, three to four times a week. All lessons were filmed with two Sony Handycam video cameras on a stand with the purpose of recording the student as well as the overall interaction.

CBRS

The Child Behavior Rating Scale, revised (CBRS; Mahoney, 1998), is an observation-based rating instrument that has been used to assess interactive

engagement in persons with S/PIMD (Hostyn et al., 2011; Van keer et al., 2017; 2020). In CBRS, 10 minutes of video recordings of interactions were coded on a five-point Likert scale. The coding procedure was conducted for each two-minute interval, i.e. in total five ratings per video. If the video exceeded 10 minutes, only the first 10 minutes were scored. The CBRS consists of two overarching constructs: attention and initiation and has seven subscales (the items and the subscales are the same). The attention construct consists of: attention to activity, persistence, involvement and compliance/cooperation. The initiation construct consists of: initiation to activities, initiation to adult, and affect. The CBRS has shown reliability and validity in previous research studies with the population (Hostyn et al., 2011; Van keer et al., 2017; 2020) although a recent study by Rensfeldt Flink et al. (2023) showed that exact point-by-point agreement may be hard to achieve. To evaluate inter-rater reliability in the current codings, two coders rated the video recordings from both conditions ('A' and 'B' phases) with CBRS. The second author of the study rated all 48 video recordings and an independent rater rated 33% of the video recordings (N=16) from both conditions. Intra class correlations with absolute agreement and single measures were applied to calculate inter-rater reliability. The obtained intra class correlations were judged as "good" (>.6) and "fair" (.4-.6) (Cicchetti, 1994). The two subscales in CBRS that constitute the main dependent variable, i.e. attention and initiation, had intra class correlations of .57 and .61, respectively. Given the well-known difficulty of performing these types of ratings with the current population (e.g., Rensfeldt Flink et al., 2023), the obtained intra class correlations were judged as satisfactory.

Engagement Model

As an additional measurement of the outcome, school staff members rated the students' engagement using the Engagement model (Carpenter et al., 2015). The school staff members independently rated five indicators of student engagement after each lesson: exploration, realisation, anticipation, initiation and persistence on a four-point Likert scale, which resulted in a total "engagement score" for each lesson.

Teacher Post-Intervention Acceptability and Importance of Effects Survey

In order to capture the school staff members' perception of the social validity and perceived pedagogical usefulness of the MSMD approach, a revised and translated version of the Teacher Post-Intervention Acceptability and Importance of Effects Survey (Lane & Beebe-Frankenberger, 2004) was completed. The survey consists of ten questions related to the social validity of the approach in the school context as well as the suitability of the approach in learning activities with the target population. The statements in the survey were rated on a Likert scale ranging from 1 to 7, where 1 corresponds to 'Do not agree at all' and 7 corresponds to 'Strongly agree'.

Data Analysis

CBRS and teacher ratings on the Engagement model were visually analysed within and across phases. In the visual analysis, the procedure as described by Lane and Gast was followed (2014). To calculate effect size the percentage of non-overlapping data (PND) was calculated, as well as Tau-U, which is a method that controls for possible positive baseline trends. Regarding PND, scores of > 90 are interpreted as highly effective, 70-90% as fairly effective, and <50% as unreliable or ineffective (Scruggs & Mastropieri, 2013). When it comes to Tau-U scores, 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).

Study III

Design

In this study, a qualitative, longitudinal case study design was used. Co-production between a special education teacher and researchers was employed to develop, implement and evaluate the feasibility of MSMD in a school context along with one student with S/PIMD and Rett syndrome. Co-production may be a fruitful way to bridge the research-practice gap and to produce research that is perceived as useful in classroom settings (Greenwood & Abbot, 2001; Martin, 2010). Also, we were analytically inspired by educational designed-based research which is a researcher – practitioner collaboration performed in iterative cycles to refine theory and practice (Armstrong et al., 2022; Getenet, 2019). In the current study, the participating teacher was involved in the planning, performance, evaluation and development of the MSMD lessons through stimulated recall interviews and reflecting upon her own strategies related to the student's interactive engagement during the lessons. The social validity and long-term feasibility were investigated through a post-intervention survey and a semi-structured follow-up interview.

Participants and Setting

One special education teacher and one student with S/PIMD and Rett syndrome participated in the study.

As a first step, contact was made with the special education teacher who was working in a compulsory school for students with S/PIMD in the Gothenburg area. The teacher was a fully qualified special education teacher and had worked in the school setting for three and a half years. A description

of the study was given by the author. The teacher confirmed her interest in participating in the study and the principal of her school was contacted.

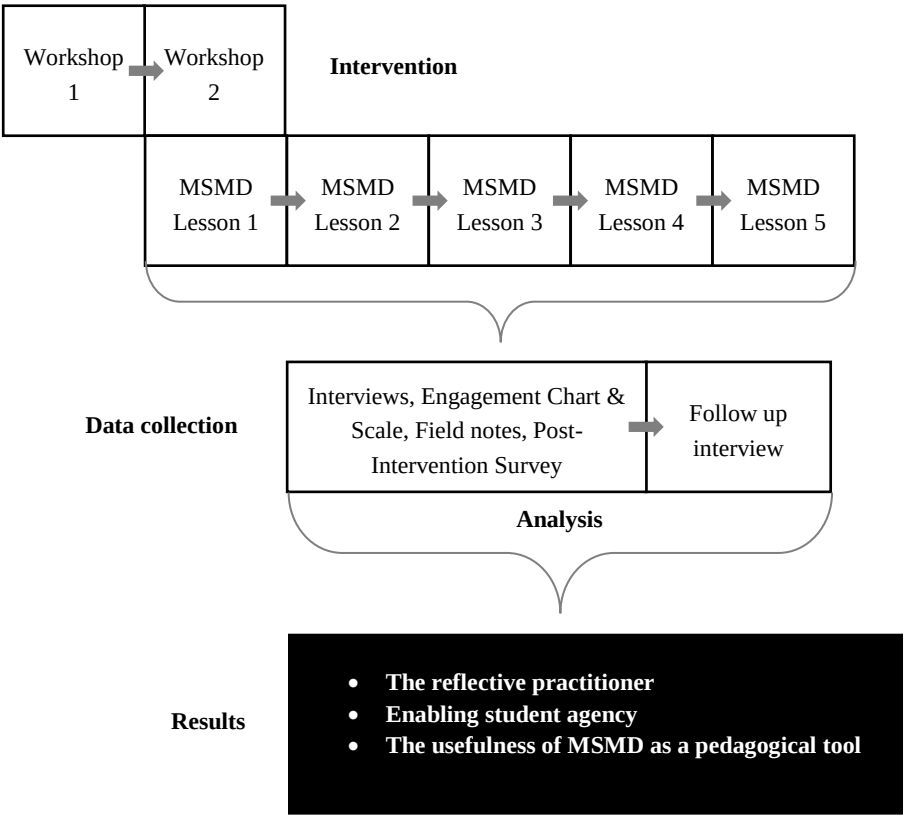
The teacher and the author discussed possible choices for a participating student with regard to the inclusion criteria and the teacher suggested an eight-year-old student with S/PIMD and Rett syndrome. The teacher then contacted the student's parents and provided them with contact information for the researchers and information about the study. The parents gave their informed written and oral consent for their child to participate in the study. Also, they provided information about their child's diagnosis and general and musical preferences as well as modes of communication. The student communicated non-verbally, including through movements, gestures, sounds and eye gaze. If she liked something, or wanted to confirm an option, she looked straight at the person or at the object. If she was uninterested, she looked away or looked at something else. Her individual educational goals concerned participating in turn-taking and confirming if she wanted to continue or end an activity (through eye gaze). She was socially interested and liked listening to music, including children's songs, watching movies and looking at an IPAD application with animal sounds.

Procedure and Data Collection

The author held two initial workshops with the teacher (five hours in total). During the workshops, underlying theories, strategies as well as practical instruction on how to create MSMDs were provided. Also, a protocol checklist regarding key underlying ideas and elements of MSMD was presented. The data collection procedure consisted of three phases: In the first phase, the author and the teacher mapped student abilities and interests, music tastes, modes of communication and how the student expressed engagement. This was an important prerequisite to make the individualised MSMD, which was called: "An adventure in the forest". Also, music interaction activities were discussed and adapted to the musical abilities and skills of both the student as well as the teacher. The second phase consisted of performing, reflecting on and refining the MSMD lessons which were carried out by the teacher once a week over five consecutive weeks in the school during school hours. Each lesson was filmed with a Sony Handycam video camera on a stand with the purpose of recording the student as well as the overall interaction. After each lesson, the author and teacher watched the video recording from the lesson. Then, they scored student engagement and reflected upon strategies used by the teacher as well as discussed improvements to be made to the next lesson in stimulated recall interviews. These discussions and reflections included that the teacher planned to adjust her strategies or support for the student with regard to the student's performance during the following lesson. The overall storyline of MSMD was kept during all lessons. The third phase consisted of a post-intervention survey and a follow-up interview, which was carried out

20 months after the last MSMD lesson was performed. In Figure 5 an overview of the data collection, measurements used for data analysis and final results are visualised.

Figure 5. Overview of the data collection, analysis and final results of study III. Based on Johnels et al., 2024b.



Measures

Stimulated Recall Interviews

Stimulated recall interviews have been used to access teacher perspectives on teaching activities (Ankney, 2016; Odena, 2001; Powell, 2014). It has been argued that stimulated recall interviews are useful for capturing complex and interactive behaviour in naturalistic and complex settings (Lyle, 2003). The stimulated recall interviews with the teacher were performed during the same or the following day as the MSMD lessons, except on one occasion, when the interview was performed after six days due to holidays. The interviews lasted between 12 and 30 minutes.

Engagement Chart & Scale

The engagement chart and scale (Carpenter et al., 2015) were used to assess the student's interactive engagement during the lessons and to reflect on improvements for the next lesson in relation to the student's engagement.

Field Notes

The author made field notes during the implementation and her visits to the school, which were used to triangulate the other data sources in the data analysis.

Teacher Post-Intervention Acceptability and Importance of Effects Survey

After the final MSMD lesson had been carried out, the teacher filled in a revised and translated version of the "Teacher post-intervention acceptability and importance of effects survey" (Lane & Beebe-Frankenberger, 2004). In line with Carnahan et al. (2009), the survey was used to capture the teacher's perspectives regarding the feasibility of MSMD in the school context and the applicability of the approach in relation to the student's syllabus.

Follow-Up Interview

A semi-structured follow-up interview was performed 20 months after the MSMD lessons had been carried out. The aim was to investigate how the teacher perceived the long-term-feasibility of the MSMD approach in the school setting, and whether she had continued to use the approach in her regular pedagogical work.

Data Analysis

Inductive content analysis was used to analyse the data from the stimulated recall interviews and the follow-up interview (Elo & Kyngäs, 2008; Kyngäs, 2020). The additional data sources (the Engagement chart and scale, field notes and post-intervention survey) were used to triangulate and validate the interview data. The results from the inductive content analysis were written narratively.

Ethical Approval and Ethical Considerations

With regard to studies 2 and 3, which included school staff members (teachers and teacher assistants) and their students with /SPIMD, an ethical vetting was conducted. Ethical approval was obtained from the ethical review board in Uppsala, Sweden (Dnr: 2019-05328).

In studies 2 and 3 the parents of the participating students were given oral and written information before they consented to their children participating.

Also, informed consent was obtained from the school staff members and the principals of the schools where the studies were conducted. All participants (the students' parents and school staff members) received information that withdrawal from the study (without any further explanation) was possible at any time.

Given the severe/profound intellectual disabilities of the participating students, it was not possible to inform them about the purpose or the procedure used in the studies. Consequently, they were not able to give their assent or informed consent to participate in the studies. Several scholars have raised this as being an ethical dilemma when conducting research on this target population, i.e. that they run the risk of being left out of research due to their inability to consent to participation (Maes et al., 2021; Mietola et al., 2017; Rensfeldt Flink, 2022). Hence, it is important to find acceptable ways to handle this. To carefully monitor the participants during the data collection and to be attentive to any signs of discomfort is suggested as an important strategy (Maes et al., 2021; Rensfeldt Flink, 2022). During the data collection of the current studies we were very vigilant for signs of discomfort from the students. In study 2, two lessons were ended earlier than planned, due to signs of discomfort in one student.

Also, it is important to consider ethical consequences in relation to the school staff members participating in the studies. Specifically, care was taken to avoid placing an additional workload on them. In study 2, the author prepared three examples of MSMDs from which the participating school staff members could choose to use with their specific students. Thereafter the author adapted the MSMD to each student. She also collected and brought the props, musical instruments and books to the lessons, so that the school staff did not have to do more additional work than necessary. In a similar manner, the author helped the teacher in study 3 to create an MSMD for the specific student. She also provided props and musical instruments for the drama and coached the teacher in how the teacher could accompany her singing in the different musical interaction activities.

Results

Study I

This scoping review study aimed at mapping and describing the existing research evidence regarding musical interaction activities with children and young people with S/PIMD from the last 20 years (2000-2020).

Twenty-five studies were identified. Regarding participant characteristics, a heterogeneity of terms to describe the target group was found. With regard to the aetiology of the intellectual disability, a large group with a known aetiology were individuals with Rett syndrome. Generally, there were few participants in each study and their ages ranged from four to 19 years old. In total, 187 participants with S/PIMD were included in the review.

A majority of the studies had a case study design. Regarding the content of the musical interaction activities, a majority of the studies entailed a shared music activity including vocalisations and playing instruments, sensory stimulation and moving to music. Yet another category consisted of technology-mediated and multisensory activities.

Regarding which dependent variables were focused upon, several studies had two or more behaviours or abilities as dependent variables that they sought to improve or stimulate by using musical interaction. In a large majority of the studies, 'social interaction and communication' were targeted. Another large group comprised 'engagement, emotions and attention'.

Concerning the data collection methods and research instruments used, video observations were used as the main data collection method in a large proportion of the studies. Predominantly, author-constructed research instruments were used to code behaviours and outcomes.

Regarding the reported effectiveness and benefits of the musical interaction activities, all but one study reported effectiveness, partial effectiveness or positive benefits on the dependent variables. The manner in which this was expressed varied due to the different types of research design.

In the inductive content analysis, six themes were identified as promising components in musical interaction: the responsivity of the interaction partner, singing songs, structure and predictability in the activities, long-term interventions, technology-mediated and multisensory music activities and a trusting alliance between interaction partners.

The quality assessment showed a range in the level of detail reported and the overall quality of the studies. The quality of the studies varied from poor

validity (12%) to high validity (88%). None of the studies made any claims of being representative of the entire population or being free from bias in participant selection/recruitment.

In conclusion, the review highlights the potential of musical interaction activities in a target population where both research and practice are aligned with many challenges. We hope that the study will provide insights into further research areas and give a reference for practice in this understudied and important research area.

The results from study 1 showed, amongst other things, also the potential of augmenting musical interaction with multisensory elements when addressing the needs of children and young people with S/PIMD.

Study II

The aim of study 2 was to investigate the effect of MSMD lessons compared to a control condition with picture book reading lessons on the interactive engagement of three students with S/PIMD. Video observations using the CBRS and school staff members' ratings of the Engagement model were applied to assess the outcome measure. Also, the study investigated the school staff members' perceptions of the social validity and pedagogical usefulness of the MSMD approach in the school context. These perceptions were captured in a post-intervention survey.

The results from coded video observations using the CBRS showed significantly higher levels of interactive engagement for all students during MSMD compared to the control condition according to visual and statistical analysis using PND and Tau-U. The PND was 100% for students 1 and 2 on both subscales (attention and initiation). For student 3, the PND was 75% on the attention subscale and 63% on the initiation subscale, with no immediate change in scores between the different phases. The baseline was stable for all three students.

The results of the school staff members' ratings of student engagement showed significantly higher levels of interactive engagement during MSMD compared to the control activity according to visual and statistical analysis for two of the students, namely students 1 and 2. PND was 88% for student 1, which corresponds to a "fairly good effect" and 100% for student 2 which corresponds to "highly effective" according to the benchmarks of Scruggs and Mastropieri (2013). For student 3, there was no statistically significant difference in visual analysis between MSMD and the control condition, which both showed a highly variable pattern from lesson to lesson. The Tau-U calculations followed the same pattern as the PND scores.

Regarding the second aim of the study – to investigate the school staff members' perception of the pedagogical usefulness of the approach – a

translated and adapted post-intervention survey was used. The overall sum score concerning the pedagogical usefulness of the approach was very high, ranging from 86 to 93% of the maximum score. Items relating to the fit of the MSMD approach in the school context as well as to the student population, received the highest score possible from all school staff members. The items that received the lowest scores related to statements concerning improvement of students' overall performance and lasting intervention effects from participating in MSMD. In the section for comments at the end of the survey, the school staff members elaborated on their ratings and several explained that a continuation of the MSMD lessons may have resulted in lasting positive effects on the students. For instance, one school staff member highlighted: "Through repetition, the development of his abilities takes place. I think [student 1] would need a continuation of the MSMD lessons to achieve his potential and improve the overall performance" (School staff member 1). Another one described it similarly: "Regarding statement 8 [MSMD will have lasting positive effects], I think you will have positive effects if you continue with this [MSMD] as a part of his daily schedule in the school, which will support memory and recognition" (School staff member 2).

Study III

The aim of study 3 was to describe a teacher-researcher co-production and to provide an in-depth analysis of how the teacher related to and challenged a student with S/PIMD and Rett syndrome while iteratively performing MSMD lessons. Also, the study investigated the teacher's perception of the long-term feasibility of the MSMD approach in her everyday work with her students.

The inductive content analysis resulted in three categories related to the aims of the study.

The first category was called "the reflexive practitioner". This category captured the way the teacher highlighted that the iterative and reflexive discussions during the stimulated recall interviews had made her more aware of her own actions and strategies during the MSMD lessons. Also, she stated that it had made her more attentive and sensitive in relation to the student's mood and subtle communicative signals.

She raised several specific strategies she had become aware of during the stimulated recall interviews (e.g., reducing her own tempo during the lessons, increasing her use of AAC strategies, for instance, aided AAC modelling and manual signing). Encouragingly, in the last stimulated recall interview, the teacher highlighted that the repeatedly reflexive discussions had made her feel more aware and confident about her own performance and had supported her in interacting more confidently with the student during the MSMD lessons.

The second category was called “enabling student agency”. The teacher mentioned that continuous attentiveness to student reactions and responses was imperative in order to be able to adjust each lesson to the student’s daily performance. She further highlighted the importance of finding a balance of intensity in both activities and interaction style to engage the student enough, but not too much. She said that sometimes the student got overly excited, which limited the student’s possibilities to interact and participate. Also, she raised the importance of timekeeping to maintain student engagement and energy throughout the whole lesson. In the follow-up interview, the teacher reported that the student managed to stay engaged and active longer during the MSMD lessons when she could anticipate what was about to happen.

The third category concerned the long-term feasibility and was called “the usefulness of MSMD as a pedagogical tool”. As in study 2, the teacher filled in a translated and adapted post-intervention survey regarding the usefulness of the approach after the performed MSMD lessons. The total sum score was 93% of the highest possible score. The teacher expressed high satisfaction with the MSMD approach and found it to be useful in relation to the subjects in the syllabus as well as highly motivating for the student. She emphasised that music and sensory exploration were highly motivating and useful in learning activities with her students more generally, and both are constituent parts of the MSMD approach. In the follow-up interview, the teacher reported that she had continued with the approach with all her students in a small group setting, as had several of her colleagues. She further stated that she did not have the same possibilities to detect all communicative signals from the student in the group setting; on the other hand, during these lessons the student was able to interact with her peers too. The teacher reported that the main obstacle regarding the long-term usefulness of the MSMD approach in her everyday work with the students was finding the time to come up with new MSMDs. A potential solution mentioned by the teacher was to cooperate with other teachers, such as the music teacher.

Discussion

General Discussion

The findings from the first study of the thesis suggest that there is a steadily increasing evidence base for using musical interaction to support developmental goals in education for students with S/PIMD. Also, the combination of musical interaction with narrative support and dramatised sensory stimulation (MSMD) was found in the second study to be effective in supporting social interactive engagement in some students with S/PIMD, and, in both the second and the third studies, school staff members perceived the approach to be useful in relation to their educational work and to curricular goals.

The empirical findings can be understood in the light of the earlier mentioned theories that have guided the research in the thesis and that have been hypothesised to support interactive engagement, namely early cognitive development, sociocultural perspective and communicative musicality. From these perspectives, development is suggested to take place within supportive and encouraging interactions (Nind, 2007; Ware, 2012) and interactive engagement is fostered when students are met by a balance of support and challenges and with motivating activities (Maes et al., 2020; Munde & Zentel, 2020; Nind 2007; Nind & Strnadová, 2020; Van keer et al., 2019b; Ware 2004). This developmental dynamic is also closely aligned with the concept of scaffolding, where the teacher models different learning strategies, for instance, how to use AAC devices or jointly playing on a drum, before the student can move towards increasing independence. Moreover, from the perspective of early cognitive development, continuous opportunities for active sensory exploration are important, in order for students to be able to experience that they can have an impact on their environment and potentially develop an understanding of contingency awareness and intentionality. The findings in the current thesis have thus suggested that responsive and contingent-rich everyday learning activities and environments, where students are given opportunities to take an active role in interactions, are given enough time to respond, and are provided with re-occurring opportunities to make choices and explore with all their senses, might support student interactive engagement, which is imperative for learning and development more generally (Munde & Zentel, 2020; Van keer et al., 2017; 2020).

Another important aspect of student development is the need for learning activities to be individualised and "designed according to the strengths and needs of each individual learner" (Munde & Zentel, 2020, p.1). To do this, an important first step is mapping of student abilities, difficulties and interests, preferably through collaboration between different stakeholders, (e.g., parents, school staff members, health care staff) to find appropriate and motivating learning activities and individualised learning goals for the students (Maes et al., 2020; Munde & Zentel, 2020). Relatedly, there is a need to adjust the tempo and affective state to the student in order to form supportive circumstances for the student to engage in learning activities. In the theoretical framework of communicative musicality, this kind of co-regulation in tempo, quality and emotion is described as a mutual "dance-like" turn-taking that is inherently human and that takes place already in the earliest interactions between infants and their caregivers (i.e., entrainment). In the context of students with S/PIMD, however, specific interactive strategies might be warranted due to delayed (and very few) initiatives and responses from the students, as well as uncontrolled and limited movement abilities. To provide a predictable framework for learning activities, pauses, repetition, and specific interactive strategies (e.g., matching, interjecting) are suggested to support student interactive engagement. Moreover, it has been suggested that the music itself provides a supportive or "holding" framework that contributes to sustaining engagement in interactions for students with S/PIMD and other severe disabilities (Wigram & Elefant, 2009).

Thus, taken together, it appears as if musical interaction with narrative support and dramatised sensory stimulation is conducive to creating conditions for interactive engagement in students with S/PIMD. More detailed discussions are provided below regarding the characteristics and findings of the three sub-studies included in this thesis.

In study 1, the aim was to map and evaluate current research relating to musical interaction and children and youths with S/PIMD. In a large majority of the published studies, musical interaction was seen to be effective or beneficial to support the outcome measures or behaviours that were focused upon in the studies, which is a promising finding with relevance for planning and conducting educational practice with the population. Also worth discussing here, is the well-acknowledged difference in terminology used to describe persons with S/PIMD (e.g. Maes et al., 2021, Nakken & Vlaskamp, 2007; Rensfeldt Flink, 2022; Ware 2004) which became apparent in the current study as well. In fact, the results showed that neither of the terms PIMD or S/PIMD was used, suggesting that these terms are not established in research concerning musical interaction/music therapy. Several reasons have been suggested to explain the causes for differences in nomenclature in the field of S/PIMD research, including differences between countries as well as factors related to the diagnostic features of small children, where a diagnosis may not yet have been determined when children are young (Maes et al., 2021;

Rensfeldt Flink, 2022). Differences in settings – such as health care, habilitation services, community disability services or schools – may also play a role. Regardless of the exact reason for the lack of consensus in nomenclature, Maes et al. (2021) emphasise the importance of detailing participant characteristics in research studies in order to compare and aggregate research findings from different studies and provide a growing evidence base.

Regarding the data collection methods and measurements, detailed video observations were the most commonly used method in the studies according to study 1, which in turn could be analysed either quantitatively using coding schemes or qualitatively. As stated by Maes et al. (2021), besides proxy reports, behavioural observations using video recordings are predominantly used in data collection with this target group. In comparison to "in vivo" observations, observations of video recordings enable more detailed coding procedures and inter-rater reliability checks. It is argued that hindering factors in observational studies with the population are the idiosyncratic behaviours and expressions in people with S/PIMD, which may make coding difficult. Commonly, observation studies with the target group are aligned with relatively low levels of inter-rater reliability, compared to other target groups (Maes et al., 2021; Rensfeldt Flink et al., 2023).

In study 1, a qualitative content analysis was performed regarding the reported promising components within musical interaction. The findings were summarised in sex categories which are well aligned with the theories that have guided the research in the thesis, e.g., using responsive and supportive (scaffolding) strategies, structure, pauses and repetition in multisensory and long-term music activities to promote contingency awareness and interactive engagement in the child with S/PIMD. These may all have practical implications for the organisation of educational practices for this group of students and when planning and conducting intervention research. Also, several of the suggested categories that were found in study 1 are integrated into the pedagogical approach of MSMD.

In study 2, which is one of very few studies that have performed an experimental evaluation of an educational approach for this population, the novel pedagogical approach of MSMD was the focus. Although new with regard to the systematic formalisation of the approach, this way of working is far from totally novel; instead, the approach is built on important principles and established facts concerning the support and learning for the target population more generally. As described above, categories of promising components of musical interaction from the scoping review are included in the MSMD approach (i.e. using responsive strategies, multisensory stimulation and structure and predictability in music activities). A few other experimental studies on the population have explored one of the core ingredients of MSMD in experimental design studies; for instance, Wandin et al. (2021) conducted a single-case-experimental design study to explore the

effect of a communication intervention package, including responsive strategies, on expressive communication and visual attention in three participants with Rett syndrome. Also, Ghetti (2002) investigated the alertness levels of six children and youths with S/PIMD during three different increasingly active music interventions (i.e. rhythmic stimulation, singing songs, multisensory rhythm instrument playing) compared to baseline. Relatedly, ten Brug et al. (2016) performed a non-randomised control study where an intervention group took part in reading multisensory storytelling books and a comparison group read regular books. The percentages of attention from the participants with S/PIMD towards the book/stimuli and storyteller were scored and compared statistically. To be able to draw conclusions regarding which methods and strategies that promote development and learning in students with S/PIMD, experimental studies, such as single-case-experimental design studies, may thus be important.

In study 2, the effect of different pedagogical conditions on interactive engagement was examined. Interactive engagement has been found to be an important prerequisite for learning more generally (Van keer et al., 2017; 2020), which motivates research investigating pedagogical interventions that support it. Since students with S/PIMD are unable to self-report (Maes et al., 2021), other ways to assess this construct must be applied. As suggested by Maes et al. (2021), a combination of different information sources may be the most valid way to do this. In line with this way of reasoning, the current study assessed interactive engagement through ratings of video observations, performed by the second author of the study, as well as inter-rater reliability checks, performed by an independent rater (inter-rater reliability was deemed adequate although far from perfect). Also, school staff members rated indicators of interactive engagement using a British pedagogical rating instrument (Carpenter et al., 2015). The results provided evidence that for two of the three students, MSMD was more conducive than the control activity in supporting interactive engagement, whereas for the third student the results were more ambiguous and fluctuating, regardless of activity. It can be concluded that MSMD seems to be effective in supporting interactive engagement in some students with S/PIMD.

In study 3, we were specifically interested in soliciting the perspective of a teacher working with students with S/PIMD on the usefulness of the MSMD approach. It has been argued that the relevance of educational research may be perceived as low by practitioners (Greenwood & Abbot, 2001), and in particular, that a "top-down" approach in research may fail to produce interventions and approaches that are perceived as feasible by practitioners. Hence, the current study was a qualitative, longitudinal case study, inspired by co-production and design-based research, in which there was close collaboration with an experienced special education teacher.

Several insights were gained through this case study concerning facilitators and barriers when implementing educational work for students with S/PIMD.

The insights concerned the MSMD approach more specifically, but there were also more general insights regarding what constitutes important professional knowledge when working with the target population. For instance, the importance of responsive and warm interactions, where the student is both encouraged and challenged (i.e. using scaffolding) was highlighted in the study. This is in line with Nind (2007), who emphasises the importance of a warm and supportive social environment that encourages and expects learning although progress is subtle and slow. Also, balanced dynamics and tempo in the MSMD lessons were found to be important for student agency and endurance. The teacher stated that in order for the student to be able to be active the whole lesson, the teacher needed to balance moments of vitality and excitement while also making time for recovery (i.e., entrainment). Otherwise, the student might become overly-exited, presumably due to her inability to regulate arousal, which is a common feature in Rett Syndrome (Bergström-Isacsson, 2011). Additionally, the teacher highlighted that the structure and repetition of the MSMD lessons made the student seem more aware of what was about to happen, and that this made her more actively engaged. This result can be directly related to the concept of contingency awareness, which is argued to be fundamental to cognitive development and learning in students with S/PIMD (Dunst et al., 2008; Ware, 2004).

Regarding facilitators, the teacher was positive about taking part in the study and she emphasised that the participation in the co-production process – which entailed continuous time for reflections on her own teaching strategies and performance during the MSMD lessons – had made her more aware of her own strategies and had made her more attentive and sensitive in relation to the student's subtle expressions. The fact that the concept and formalisation of the MSMD approach is mainly research-driven ("top-down"), did not seem to influence the teacher in a negative way. In fact, the teacher's specific pedagogical knowledge concerning continuous adjustments that needed to be done in relation to her specific knowledge of the student helped to improve the lessons substantially. Hence, it can be argued that the MSMD approach had become a pedagogical tool "owned" by the teacher. This could be related to arguments raised by Preece and Zhao (2015), who emphasise the importance of pedagogic professional knowledge judgements in order to adapt interventions and methods to their specific pedagogical context and to specific students. Interestingly, in the follow-up interview, the teacher described how she had developed the MSMD lessons from an individual activity into a group activity, which had benefits such as peer interactions and increased feasibility in the school context.

Another interesting finding was related to the performance of musical interaction activities within the MSMD lessons. The teacher had no prior experience of playing musical instruments (although she used to sing with her students). Before the data collection, the author coached the teacher on how she could accompany her singing. Consequently, the teacher used a big

African djembe drum and a specially constructed guitar (with a built-in bracket that is moved to change the chords on the guitar) to accompany her singing during the lessons. The teacher's positive experience indicates that the MSMD approach could be used regardless of previous knowledge of playing musical instruments, although this proposal needs to be specifically explored in future research.

Regarding barriers to using the approach, the main obstacle mentioned by the teacher was finding the time to create new MSMDs. She stated that although she could relate the approach to all school subjects in the students' syllabus, it was hard to find time to invent new MSMDs. A potential solution mentioned by the teacher was to cooperate with other teachers, such as the music teacher.

Relating to long-term feasibility, in the follow-up interview, the teacher stated that she had continued to use the MSMD approach in the school context and that several of her colleagues in the school had started to use the approach as well. The promising findings from the current study suggest that MSMD may be a feasible pedagogical approach when working with students with S/PIMD. However, in order to probe the generalizability of the approach with the student population, more research, preferably with other research groups and with a much larger sample is called for. Relatedly, in future research it would be interesting to investigate, in a co-productive manner, how MSMDs developed by school staff would be constituted and organised.

Methodological Discussion

The work in this thesis has been inspired by researchers who argue for the value of theoretical eclecticism (Abbeduto & Boudreau, 2004; Odom, 2016; Pressley et al., 2023) and methodological pluralism (Ferri et al., 2011; Maes et al., 2021; May et al., 2017) which highlight the benefits of diverse theories, methods and analysis techniques in complex research fields, such as the research field of special education (Ferri et al., 2011; Odom, 2016) and in research concerning people with S/PIMD more specifically (Maes et al., 2021).

In the thesis this was reflected in the following manner. The first study scoped the current state of available research regarding musical interaction performed with the target population. For this purpose, a scoping review was chosen. Scoping reviews are commonly suggested in emergent research fields where the evidence is drawn from different types of research methodologies and paradigms (Peters et al., 2020). Also, the screening processes in scoping reviews are commonly time-consuming and critical distinctions may need to be changed iteratively due to broad search strings and research questions (Oudshoorn et al., 2021). In the current review, a quality appraisal of the

included studies was conducted, which can be argued to reflect the more technocratic role of research – i.e. the issue of "what works" (Biesta, 2007). Also, a qualitative content analysis of what was perceived as promising components of music interaction in the included studies was performed and we included both qualitative and quantitative research studies, which could be argued to reflect the more cultural and interpretative role of research related to "what matters" (Biesta, 2007). As proposed by Onghena et al. (2019), triangulation of research methods and paradigms may indeed provide a more nuanced view of complex research fields, as is the case with the one at hand.

In studies 2 and 3, the novel pedagogical approach of MSMD was scientifically examined and evaluated not "only" for its effectiveness but also for its perceived appropriateness and social validity with relevant stakeholders. The aim was to test whether the combination of musical interaction, multisensory exploration within a narrative frame and being performed in a responsive manner enhanced students' interactive engagement. All this was done in a single-case-experimental design study, where MSMD was compared with a control condition (picture book reading) that was a commonly occurring educational activity in the school setting. As proposed by many scholars, single-case-experimental design studies may be the most feasible choice to test the effectiveness of interventions and to gather an evidence base of support for people with S/PIMD (Munde & Zentel, 2020; Van keer & Maes, 2018) where the findings from many single-case-experimental design studies (on the same outcome measures) can be aggregated to form an evidence base (Munde & Zentel, 2020; Van keer & Maes, 2018). In order to gather a valid and reliable aggregation, several requirements need to be fulfilled in the research studies, e.g., one needs to sufficiently describe participant characteristics (Maes et al., 2021), and show transparency in data collection and analysis regarding the measures and procedures used (Van keer & Maes, 2018), which we tried to achieve.

Also, the social validity and feasibility of the approach as perceived by stakeholders (i.e. teachers and teacher assistants) were investigated in studies 2 and 3. It has been argued that for interventional research to attain social validity in school contexts, it is imperative to gain educators' input on the interventions (Komesidou et al., 2022). This was done to some extent in study 2, where the participating school staff members' perceptions of the MSMD approach were investigated in the "teacher post-intervention acceptability and importance of effect survey" (Lane & Beebe-Frankenberger, 2004). In study 3, this approach was carried out to a considerably greater extent, by including features of co-production (Martin, 2010) and design-based research (Armstrong et al., 2022; Getenet, 2019). The co-production process between the researchers and the special educator included the development, implementation and evaluation of the MSMD lessons in the educational context. The extent of co-production between researchers and practitioners can vary and can be related to the design, evidence gathering, analysis and

dissemination. In study 3, the extent of co-production is suggested to correspond to levels 2 or 3, according to typology by Martin (2010). That is, the participating teacher was involved in the development and the evaluation of the pedagogical approach, although the design and general conception were research-driven. In future research, other or additional forms of collaborative co-production may be tested. A specific research objective relating directly to design-based research and co-production is to explore the manner in which the MSMD lessons were transformed from an individual activity into a group activity.

Limitations and Directions for Future Research

Apart from the limitations described in the previous sections, some additional limitations of the studies performed in the thesis need to be highlighted, along with a discussion of potentially fruitful directions for future research.

A commonly acknowledged limitation in research with people with S/PIMD, that applies to the current thesis as well, is the limited possibilities to generalise the results (Maes et al., 2021). As has been reported in the background, the prevalence of S/PIMD in the total population is low (approx. 0.025%, Ölund et al., 2003). Hence, it is not surprising with small sample sizes in studies of people with S/PIMD (Maes et al., 2021). With that being said, a small sample size does limit the possibility to utilise generally accepted statistical measures, which in turn potentially will affect the possibility of capturing intervention effects and providing study validity (e.g., Field, 2013). Indeed, the number of randomised controlled trials in the field of S/PIMD is very small, if existing at all (Munde & Zentel, 2020). It has been proposed that results from several single-case-experimental design studies may be one viable way to try to circumvent this, and that findings gathered from different studies on the same outcome measures may be aggregated through meta-analyses and hence help build an evidence base (Rensfeldt Flink, 2022; Van keer & Maes, 2018). Although the studies of the thesis also include very few participants, it is hoped that they will contribute to the building of such an evidence pool of studies. In order to enable such a cumulative aggregation of findings, several methodological requirements need to be fulfilled, e.g., detailed participant descriptions and transparency in implementation, data collection and data analysis, which we tried to obtain. Another complementary option is to develop and collaborate in larger research consortia across scientific institutions and countries (Maes et al., 2021).

Relatedly, an issue that was brought up in study 1, and that was also a challenge that needed to be handled in study 2, is the well-recognised lack of validated research instruments for the target population (Maes et al., 2021). As reported in study 1, researcher-developed instruments were most

commonly used, and only in a few studies, did the researchers use validated instruments. In study 2, the choice of research instrument to assess the outcome measure (i.e. interactive engagement) was thoroughly considered. The CBRS was chosen since it has been used in other studies studying interactive engagement in the same population (e.g., Van keer et al., 2017; 2020; Rensfeldt Flink et al., 2023). As has been raised by several scholars, the idiosyncratic behaviours in people with S/PIMD may hinder the interpretation of rating scales such as the CBRS. To circumvent this, preparatory work, such as coding workshops and communication profiles of participants may be used, which was done in the current study. The author held a workshop (2 h) with the two raters on how to rate interactive engagement using the CBRS in video recordings of MSMD lessons. As mentioned, the obtained reliability was considered adequate.

Yet another threat to validity may be the risk of bias due to social desirability in proxies or stakeholders (Maes et al., 2021). Additionally, in relation to the MSMD approach – which has been customised by the author – there is also a risk that our interests and professional investments may affect the conclusions. As regards the risk of bias in study 2, several measures were used to counteract this. Specifically, a triangulation of researcher ratings of CBRS was validated by independent school staff member ratings using the Engagement model. Also, inter-rater reliability was assessed for 33% of the data points with the assistance of an independent rater. Thus, several measures were taken to ensure the conclusions were solid and trustworthy. With that being said, it has been proposed that other, more objective measures might help develop research. Interestingly, physiological measures may be used to complement and validate observational data (Maes et al., 2020; 2021). For instance, Sterkenburg et al. (2017) used measures of skin conductance to validate attachment behaviours, and Vos et al. (2013) and Hammann et al. (2022) used heart rate variables to validate codings of emotions in people with S/PIMD. It is further suggested that the combination of assessment methods from both qualitative and quantitative data as well as combining information from several sources such as professionals, parents and researchers may provide more complete answers to research questions related to the complex research field regarding people with S/PIMD (Maes et al., 2021; Munde & Zentel, 2020).

In the post-intervention survey, in which the school staff members rated the social validity and feasibility of the MSMD approach, several school staff members gave nuanced answers and pointed out, amongst other things, that transferable effects beyond the MSMD lessons remain unclear. This might indicate that the participating school staff members felt safe to express their views on the topic. With regard to study 3, the aim was not to recruit a teacher who was completely neutral in relation to the MSMD approach. In fact, contact was made with a special education teacher who had specifically shown interest in learning more about the MSMD approach. Hence, the purpose of

the study was to provide a transparent and illustrative description of how the MSMD approach was appropriated and developed by an experienced and interested teacher in a school setting, which may inspire other researchers and practitioners to use it in other settings for this group of learners. Beyond her positive statements, the fact that the teacher and several of her colleagues had continued to use the MSMD approach after the research study had been finalised, may indicate that the approach was in fact perceived as feasible and socially valid.

Regarding the assessment of student interactive engagement in study 2, which has been found to be an important prerequisite for development and learning for the population (Munde & Zentel, 2020; Van keer et al., 2017; 2020), interactive engagement was only assessed "in the moment", during the lessons, and thus we are unable to make any claims regarding potential transferable or lasting effects on student interactive engagement across situations and/or interaction partners. Even so, interactive engagement in the moment may be seen as both a means and an end, where raised attention or increases in initiatives taken provide the student with a meaningful experience and increased potential for learning and development. With that being said, in future research it would be interesting to scientifically evaluate, in a controlled follow-up study, whether a long-term intervention with MSMD would reveal generalised effects, including transferable abilities, on the students' interactive engagement. Given the almost complete lack of controlled group studies focusing on educational outcomes for students with S/PIMD, this seems to be an important task for future research. Numerous considerations would need to be made to achieve this, including the choice (or development) of appropriate and sensitive outcome measures.

Another aspect of studies 2 and 3 that could be questioned is related to the fact that the MSMD approach was delivered in a one-to-one format. This is not always (economically and organizationally) feasible in a school setting. From this perspective, it would be particularly interesting to learn more about the way a teacher, such as the one in study 3, transformed and developed MSMD into a group learning activity and what affordances this might have in relation to student learning, including peer interactions.

In study 3, the perspective of one practitioner on the feasibility of the MSMD approach was explored in her everyday school work. Interestingly, the findings mirrored several aspects raised in prior research on positive teaching strategies when working with students with S/PIMD. For instance, Maes et al. (2020) emphasise the importance of patience (i.e. waiting for student initiatives), responsiveness (including, playfulness and creativity) and the practitioner having possibilities to reflect upon their own teaching. This is all in line with the findings from the study, which revealed several important aspects which have direct implications for similar educational practices. With that being said, it was only the perspective of one teacher, and in future research it would be interesting to continue to unravel what constitutes

teachers' pedagogical content knowledge (Gudmunsdottir & Shulman, 1987) for this group of learners in a more representative sample.

Implications for Practice

Based on the findings of the thesis, the following implications for educational practice are recommended.

Musical interaction activities have been shown to be beneficial in supporting interactive engagement in children and young people with S/PIMD. In study 1, six promising components were revealed which are suggested to be useful in educational settings with these learners, and these can all be considered by professionals. Also, the pedagogical approach of MSMD, which combines several strategies that have been found to be useful with this group of learners (i.e. responsive strategies, multisensory stimulation and structured musical activities within a narrative framework), may provide practitioners with an additional learning activity for these learners. A potential obstacle regarding the feasibility of the MSMD approach, as was mentioned by the teacher in study 3, was finding the time to create new MSMDs. An MSMD manual for practitioners is currently being developed, which also includes several examples of MSMDs to use with the students. Potentially, this may provide a useful pedagogical tool for practitioners working with students with S/PIMD.

Secondly, supportive and responsive interaction partners are important for student development and learning. It has been suggested that strategies used within early caregiver-infant interactions may provide insights into how these supportive, responsive strategies may be performed (Goldbart, 2018; Nind, 2007; Ware 2004; 2018). Interestingly, high qualitative interactions have been suggested to have musical elements: "[W]hen working optionally it can be characterised as a dance, a symbiotic sharing of time and space to exchange meanings, take turns and validate the efforts of both partners to communicate" (Maes et al., 2020, p. 49). Moving on from this, it has been suggested that the music itself may promote the interactive engagement of both interaction partners since music has the potential to sustain and energise the interaction by providing a "holding" musical framework (Wigram & Elefant, 2009).

Thirdly, time for self-reflection as a way to build pedagogical content knowledge for school staff members working with this group of learners was raised in study 3, as in prior research (e.g., Maes et al., 2020; Nind & Strnadová, 2020). In study 3, the participating teacher specifically emphasised that self-reflection had made her more aware of what she was doing and thus more confident in providing the appropriate support for the student. Indeed, balancing adjustments in tempo and intensity in the activities, as well as continuously challenging the student during the learning activities, were

raised by the teacher as key enablers for student development. Given the commonly expressed risk of these students becoming passive (Dunst et al., 2008; Östlund, 2015), this is indeed important to consider for practitioners working with these learners.

While many of these strategies may be seen as beneficial for all students, it has been argued that they may be of even greater importance for students with S/PIMD (Maes et al., 2020; Nind, 2007), because their learning trajectories are far from straightforward and very slow. Thus, Nind and Strnadová (2020) highlight that while these teaching strategies "benefit from specialised knowledge (such as early developmental stages) they also share common characteristics of good teaching". Ware (2004) further states that teaching approaches and interventions that are structured, long-term and well-grounded in theory are successful. Thus, it is argued that school staff members working with this population need continuous training to increase their educational skills: "Hence, we need to combine some sort of critical realism with an effort not to entrench low expectations; we need to include reference to what people can do in the best environments with people who can interpret their communication" (Nind & Strnadová, 2020, p.2).

Conclusions

Research focusing on effective and helpful educational strategies for supporting students with severe learning difficulties in our schools is strikingly sparse, and this is especially the case in relation to students with S/PIMD who have the most extensive combinations of intellectual and motor disabilities. It has been shown that learners with S/PIMD typically spend limited time in attentive states (Munde et al., 2012; Ware, 2004) and take few initiatives (attentive rates) which can negatively affect their possibilities to learn (Ware, 2004). In addition, differences in their memory and information processing call for specific strategies and approaches in learning activities (Ware, 2004). Encouragingly, prior research has suggested that when learning activities are led by responsive and supportive teachers who have knowledge about early developmental stages, this has been shown to have beneficial effects on a number of outcomes, including interactive engagement. More generally, individualised, structured and motivating educational activities – which include possibilities for multisensory exploration, use of AAC, as well as aesthetic experiences – have been suggested to support interactive engagement in students with S/PIMD. The findings and conclusions of this thesis suggest that there is an evidence base for using musical interaction to support developmental goals related to education for students with S/PIMD. Also, the combination of musical interaction with dramatised sensory stimulation (MSMD) was found to be effective in supporting social interactive

engagement in some students with S/PIMD, and school staff members perceived the approach as feasible and helpful in relation to their educational work and well aligned with curricular goals. Given the scarcity of learning strategies and activities for the student population, the findings of the thesis are of great relevance for developing practice and for inspiring future research – which is still much needed - in the field of special education for students with S/PIMD.

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