

Music therapy following preterm birth

Results from a pragmatic trial evaluating the effects of parental singing on bonding and parent mental health

Tora Söderström Gaden

Thesis for the degree of Philosophiae Doctor (PhD)
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Scientific environment

This PhD-project is a shared enterprise between The Grieg Academy Music Therapy Research Centre (GAMUT) and RKBU West at the NORCE Norwegian Research Centre AS, and the Faculty of Humanities, University of Bergen. The work took place between June 2018 and July 2022 as a subproject of the larger trial *Longitudinal Study of music Therapy's Effectiveness for Premature infants and their caregivers* (LongSTEP). I, Tora S. Gaden, have been the project coordinator of the LongSTEP trial and main responsible for my thesis titled *Music therapy following preterm birth: Results from a pragmatic trial evaluating the effects of parental singing on bonding and mental health*.

LongSTEP is funded by the Research Council of Norway (RCN, project number 273534), under the program High-quality and Reliable Diagnostics, Treatment and Rehabilitation (BEHANDLING). Principal investigators of the LongSTEP trial have been Christian Gold (2018-2019) and Claire Ghetti (2019-2022).

The doctoral education was carried out within the general doctoral program at the Faculty of Humanities, University of Bergen, and the Grieg Research School (GRS) at the Grieg Academy from 2018-2022. The main supervisor of the thesis was Claire Ghetti, music therapist and professor of music therapy at the Grieg Academy Music Therapy Research Centre, GAMUT, University of Bergen. Co-supervisors were Christian Gold, music therapist and professor of music therapy at the NORCE Norwegian Research Centre AS and University of Bergen; and Ingrid Kvestad, clinical psychologist and research professor at RKBU West, NORCE Norwegian Research Centre AS.



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List of abbreviations

ANCOVA – Analysis of co-variance

BL – Baseline

CI – Confidence interval

DC – Discharge

EPDS – Edinburgh Postpartum Depression Scale

FCC – Family-centered care

GA – Gestational age

GAD-7 – General Anxiety Disorder

ICC – Intraclass correlation coefficient

ID – Infant-directed

IRR – Interrater reliability

KC – Kangaroo care

LBW – Low birth weight

M – Mean

MD – Median

MT – Music therapy

NBO – Newborn Behavioral Observation system

NICU – Neonatal intensive care unit

PBQ – Postpartum Bonding Questionnaire

PMA – Postmenstrual age

RCT – Randomized controlled trial

SC – Standard care

SD – Standard deviation

TD – Treatment delivery

TF – Treatment fidelity

TR – Treatment receipt

Preface

This PhD-project was realized as a part of LongSTEP – Longitudinal Study of music Therapy’s Effectiveness for Premature infants and their caregivers. LongSTEP is a multinational, pragmatic, randomized controlled trial that aims to provide high quality evidence of the long-term effects of music therapy (MT) on mother-infant bonding, parent mental health, and infant development.

My entrance into the LongSTEP project

I came to the LongSTEP project as a trained music therapist from the Norwegian Academy of Music in Oslo. Norwegian MT has strong humanistic roots and emphasizes context, the potential of the emergent musical relationship, user participation and individual resources (Ruud, 2008, 2010; Trondalen et al., 2010). After finishing my education, I worked full-time as a music therapist in the pediatric department at Akershus University hospital (AUH). There, a key responsibility of mine was the implementation of MT in the neonatal care unit.

The year before the LongSTEP trial commenced, AUH received external funding for a two-year project aiming to systematically develop a MT program for premature infants and their families that fit the Norwegian context (Janner et al., 2021). My interest in MT with families was partly rooted in a general interest in developmental psychology, which later led me to dedicate my master’s thesis to a MT group project for first-time mothers and their infants during their first year of life (Gaden, 2015; Gaden & Trondalen, 2018). As a singer myself, I am fascinated by the similarities that the early interaction between parents and infants has with that of musical interaction and improvisation. Across time and cultures, parents have used their voices to soothe, regulate, and interact with their babies. I entered professional training in MT with a firm belief that song and voice have a unique potential in working with families and young children. During the first year of my master’s program, on a study trip to New York, we visited the Louis Armstrong Center for Music and Medicine (LACMM), where I was introduced to MT with premature

infants and their parents for the first time. A music therapist presented video examples from practice, where we could observe infants' physiological signs such as their respiratory rate, oxygen saturation and heart rate on a monitor, and see how these improved when either the music therapist or a parent sang to them. This in-the-moment approach, using infant physiological measures to inform musical interaction, was new to me and sparked a curiosity about the potential of MT in neonatal care. While the physiological measures fascinated me, I also wondered about how music could help these families in less observable ways. The music therapist at LACMM also told us about how parents could process difficult experiences related to the preterm birth through songwriting and improvisation. Furthermore, she explained that because of the fragility of some of the babies, their parents could not always hold or touch them. In MT parents were supported in exploring careful singing as a way to "hold" their baby through the presence of their voice. I was moved by how such musical experiences could help parents and infants feel connected and closer to each other, even when they were separated by the walls of an incubator. I decided to pursue training to obtain the necessary clinical skills to one day take part in this practice myself. In 2015, while finishing my master's degree and before getting the music therapist position at AUH, I completed training in New York in the First Sounds: Rhythm, Breath, Lullaby model developed by Joanne Loewy with input from music therapists worldwide (Loewy, 2016).

During my first year in the pediatric department at AUH, nursing staff in the NICU underwent training in the clinical observational tool Newborn Behavioral Observation system (NBO) (Nugent et al., 2007). The training was part of the unit's ambition to expand their family-centered care practices, which already entailed a well-established Kangaroo Care program, where parents spend longer portions of time with their infant skin-to-skin on their chest. I joined the NBO-training and through this further expanded my knowledge on infant behavior, development, and early relationships. I soon discovered many similarities between NBO and the resource-oriented focus of my MT background – both approaches emphasizing collaboration, strengths, resources, and participation.

Based on this background, the PhD-position in the LongSTEP-project appeared as a golden opportunity to immerse myself more fully in my field of interest, and perhaps even partake in contributions for the further development and implementation of MT in neonatal care settings in Norway and potentially in other countries.

In contrast to the local, small-scale, explorative project I coordinated at AUH, LongSTEP was a big, international, longitudinal trial which, to a large degree, was designed and ready to implement at the time when I joined the team in June 2018. My role and tasks in the project were also quite well established when I started. Such a path into a research project is normal in the medical field, whereas in Norwegian MT research it is more common that candidates develop a project idea and its subsequent research design themselves, and then seek funding for the position. I would say that both approaches have their advantages and disadvantages. As I had no prior experience with quantitative research, it was reassuring to join an experienced research team that had already set up a methodologically rigorous research project. However, there was inevitably less room for me to freely choose the substudies I would complete within the frames of the main project. The LongSTEP team needed someone to take the lead in writing up and expanding upon the MT intervention protocol and its theoretical foundations, and they wanted to ensure that treatment fidelity was properly addressed in this large multinational trial. With my recent experience from the process of developing the MT program for the neonatal care unit at AUH, I found this focus on different aspects of the intervention interesting, and these tasks became two of the substudies and articles for the thesis. Existing research of MT with premature infants and their parents focuses predominantly on the time of hospitalization. Since the main trial would report longer-term effects, we decided it would be meaningful to make the results of the RCT at the timepoint of discharge the third substudy of my thesis. In addition to leading the subproject resulting in this thesis, I have been the project coordinator of the LongSTEP team with tasks including data entry; data quality assessment and monitoring; site visits; training and supervising the study's music therapists; developing treatment fidelity assessment tools; dissemination; and more.

Abstract

Background

Preterm birth is recognized as a major global health problem with potential adverse consequences for both infants and parents. The unexpected event of preterm birth is stressful, and often leaves parents with fears and worries for their baby's survival and health. Long-term sequelae of prematurity involve a range of health problems for infants, and some parents report mental health challenges, such as anxiety and depression, following preterm birth. A key assumption in the present research is that preterm birth may have a negative impact on parent-infant bonding. Previous research has confirmed positive effects of music therapy (MT) on premature infants' heart rate, respiratory rate, stress levels and feeding volume, as well as maternal anxiety. However, more large-scale, rigorous investigations of short- and long-term infant and parent outcomes, as well as mutual parent-infant outcomes like bonding, are necessary.

This PhD-project was realized as a part of the multinational, pragmatic, randomized controlled trial (RCT) "LongSTEP" (Longitudinal Study of music Therapy's Effectiveness for Premature infants and their caregivers). The overall aim of LongSTEP was to determine whether MT, delivered during a neonatal intensive care unit (NICU) stay, and/or during follow-up post-discharge, is superior to standard care in improving long-term mother-infant bonding, parent mental health and infant development.

Aims

The present thesis consists of three substudies for which all data were collected within LongSTEP. The aims for the substudies were: 1) to further develop and refine the theoretical framework and essential elements of the MT protocol; 2) to measure and report the short-term effects of the RCT on mother-infant bonding and parent mental health; and 3) to evaluate treatment fidelity of the MT intervention in both the NICU and post-discharge phases of the LongSTEP trial.

Methods and procedures

The LongSTEP trial was designed as a 2×2 factorial, multinational, pragmatic RCT. In this thesis, the short-term effects of the MT-intervention were analyzed at the timepoint of discharge from NICU. Participants were premature infants born before week 35 of gestation and their parents recruited from eight NICUs in five countries: Argentina, Colombia, Israel, Norway, and Poland. Participants were randomized to either the control group receiving standard care throughout NICU hospitalization or the intervention group receiving MT in addition to standard care. MT during NICU consisted of three weekly sessions throughout hospitalization. Parents were required to participate in at least two of three weekly sessions.

The MT approach was built on principles from resource-oriented MT, and developmental- and family-centered care. Musical interaction was tailored according to, and informed by, infant neurodevelopment and behavioral needs. Parents' voices were emphasized as a unique resource to build and strengthen the parent-infant bond. Hence, parental voice was promoted as the main musical source in our intervention. The role of the music therapist was a collaborative and supportive one, intentionally leaving space for parents' voices to be heard by minimal use of accompanying instruments (Article I). The primary outcome, mother-infant bonding, was measured with the Postpartum Bonding Questionnaire (PBQ). Secondary outcomes, maternal depression, and parent anxiety were measured with the Edinburgh Postpartum Depression Scale and Generalized Anxiety Disorder-7, respectively. Effects were examined by testing group differences at discharge by linear mixed effects models (ANCOVA), adjusted for site due to stratified randomization (Article II). Treatment fidelity was measured with Likert-scale-based questionnaires designed specifically for the study. The internal consistency of the questionnaires was evaluated with Cronbach's alpha. Interrater reliability was measured with an intraclass correlation coefficient and Gwet's AC. Fidelity scores were analyzed with descriptive methods (Article III).

Results

A total of 213 families were recruited between August 2018 and April 2020. 108 of them were randomized to standard care, 105 to MT. Treatment uptake and retainment in the study was very high, and the MT approach was feasible and safe. Treatment fidelity analyses showed that MT was delivered and received as intended across the included sites. This indicates high applicability of the approach across cultural contexts where parents' presence in NICU is high. However, we found no significant differences between groups, neither on the primary outcome, mother-infant bonding, nor the secondary outcomes, maternal depression, and parent anxiety, as measured at discharge from NICU. The estimated group effect (95% CI) for the PBQ was -0.61 (-1.82 to 0.59). At baseline, only 4% (9/209) of the mothers scored above the threshold for impaired bonding. For maternal depression, both groups scored below the clinical cutoff at baseline. Mothers in both groups, and fathers in the MT group, showed mild anxiety at baseline.

Conclusions

While the MT intervention with emphasis on parental singing was found feasible and well-accepted by the participants across the sites, results from the RCT showed no significant improvements of mother-infant bonding in the MT group. These results may be attributed to an already good mother-infant bonding in most enrolled participants at the outset. Our findings challenge the assumptions of problematic bonding and poor parent-mental health following preterm birth. We propose that future studies on the effect of resource-oriented MT approaches in NICU-settings with high parental presence and participation choose outcomes that are less pathology-oriented, such as parent self-efficacy, empowerment, and quality of life.

Sammendrag

Bakgrunn

Prematur fødsel er et stort globalt helseproblem som rammer både foreldre og barn. Som regel er prematur fødsel en uventet hendelse som medfører mye stress og bekymring for foreldre. Usikkerhet, og frykt for barnets liv og helse kan prege hverdagen på nyfødtintensivavdelingen. Prematurfødte barn er i risiko for en rekke langsiktige helseutfordringer, men også foreldre rapporterer belastninger og symptomer på psykisk uheld, som angst og depresjon, etter prematur fødsel. En sentral antakelse i dette forskningsprosjektet er at prematur fødsel kan ha en negativ innvirkning på utviklingen av kontakten og kvaliteten av båndet mellom forelder og barn, på engelsk kalt *bonding*. Tidligere forskning har funnet positive effekter av musikkterapi (MT) på premature nyfødte sin hjerterefrekvens, respirasjonsfrekvens, stressnivå og spising, samt på angst hos mødre. Det er imidlertid behov for mer forskning i form av større studier som undersøker både korttids- og langtidseffekter av musikkterapi etter prematur fødsel på barn, foreldre, og på felles utfallsmål som *bonding*.

Dette doktorgradsprosjektet ble gjennomført som en del av “LongSTEP” (Longitudinal Study of music Therapy’s Effectiveness for Premature infants and their caregivers), en multinasjonal, pragmatisk, randomisert kontrollert studie (RCT). Hovedmålet med LongSTEP er å undersøke langtidseffekten av musikkterapi under innleggelse på nyfødtintensiv avdeling, og/eller som oppfølging etter utskrivelse, på båndet mellom mor og barn. I tillegg undersøkes både korttids- og langtidseffekter av musikkterapi på foreldrenes psykiske helse og barnets utvikling.

Mål

Doktorgradsprosjektet består av tre delstudier der alle data ble innsamlet gjennom LongSTEP. De tre delstudiene hadde følgende mål: 1) å utdype det teoretiske rammeverket for musikkterapi-intervensjonen og beskrive de essensielle elementene i intervensjonsprotokollen, 2) å rapportere korttidseffektene av musikkterapi under innleggelse på nyfødtintensiv på båndet mellom mor og barn, og på foreldrenes

psykiske helse, og 3) å evaluere *treatment fidelity* - om musikkterapien ble gjennomført i tråd med LongSTEPs intervensjonsprotokoll i begge intervensjonsfaser av studien (under innleggelse og ved oppfølging etter utskrivelse).

Metode og prosedyrer

LongSTEP ble designet som en 2×2 faktoriell, multinasjonal, pragmatisk RCT. Dette doktorgradsprosjektet rapporterer korttidseffektene fra studien målt ved utskrivelse fra nyfødtintensivavdelingen. Deltakere i studien var premature barn født før svangerskapsuke 35 og foreldrene deres. De ble rekruttert fra åtte nyfødtintensivavdelinger fordelt på fem land: Argentina, Colombia, Israel, Norge og Polen. Deltakerne ble randomisert til enten kontrollgruppen som mottok standardbehandling gjennom innleggelsen, eller til intervensjonsgruppen som fikk musikkterapi i tillegg. Musikkterapien besto av tre sesjoner per uke, der foreldrene måtte delta i minst to av dem. Musikkterapi-intervensjonen som ble evaluert i studien bygger på prinsipper fra ressursorientert musikkterapi og utviklingstilpasset- og familiesentrert omsorg. Musikalsk interaksjon ble individuelt tilpasset barnas utvikling og behov i øyeblikket. Foreldres stemme ble vektlagt som en unik ressurs for å styrke båndet mellom foreldre og barn, og var det viktigste musikalske elementet i sesjonene. Musikkterapeutens rolle var å støtte opp om den musikalske interaksjonen mellom foreldre og barn. For å gi rom til foreldrenes stemme og tilrettelegge for at de skulle bli hørt, brukte musikkterapeuten andre instrumenter sparsommelig (Artikkel I). Hovedutfall i studien var endringer i mor-barn-båndet målt med Postpartum Bonding Questionnaire (PBQ). Sekundærutfall var depresjon hos mor og angst hos begge foreldre målt med henholdsvis Edinburgh Postpartum Depression Scale og Generalized Anxiety Disorder-7. Effekten av musikkterapi ble målt ved å teste gruppeforskjeller med en lineær sammensatt modell, (ANCOVA), justert for behandlingssted på grunn av stratifisert randomisering (Artikkel II). Fidelity-målinger ble gjort med Likert-skalabaserte spørreskjemaer utviklet spesifikt for studien. Spørreskjemaenes interne konsistens ble evaluert med Cronbach's alpha, og interrater-reliabilitet med intraclass korrelasjonskoeffisient og Gwet's AC. Fidelity-skåringer ble analysert med deskriptive metoder (Artikkel III).

Resultater

I perioden mellom august 2018 og april 2020 ble 213 familier rekruttert til studien. Av disse ble 108 randomisert til standardbehandling og 105 til musikkterapi. En svært høy andel av deltakerne gjennomførte de planlagte sesjonene og ble værende i studien. Musikkterapien ble evaluert som sikker og gjennomførbar. Fidelity-analysen viste at musikkterapien ble gjennomført og mottatt som planlagt ved alle studiens behandlingssteder. Dette indikerer høy anvendelighet av tilnærmingen i ulike kulturelle kontekster og nyfødteintensivavdelinger med høy foreldre-tilstedeværelse. Vi fant imidlertid ingen signifikante forskjeller mellom gruppene på verken primær- eller sekundærutfall målt ved utskrivelse. Den estimerte gruppeeffekten (95% CI) for PBQ var -0.61 (-1.82 to 0.59). På baseline-målingen skåret kun 4% (9/209) av mødrene over terskelen for indikasjon på et problematisk mor-barn-bånd. I begge grupper skåret mødrene under terskelen for indikasjon på depresjon ved baseline. Mødre i begge grupper, og fedre i intervensjonsgruppen hadde indikasjoner på mild angst ved baseline.

Konklusjoner

Selv om musikkterapien i studien var godt mottatt av deltakerne og vurdert som sikker og gjennomførbar, kunne vi ikke påvise noen signifikant forbedring av mor-barn-båndet som et resultat av intervensjonen. Mangelen på effekt kan muligens forklares av den svært lave forekomsten av problemer med mor-barn-båndet i utgangspunktet. Funnene våre utfordrer antagelsen om at mange foreldre strever med båndet til barnet og har utfordringer med psykisk helse etter prematur fødsel. I fremtidige studier av ressursorienterte musikkterapitilnæringer ved prematur fødsel anbefaler vi å velge utfallsmål som er bedre i stand til å fange opp det som er hovedhensikten, nemlig å styrke ressurser. Det kan være for eksempel foreldres mestringstro, empowerment og livskvalitet.

List of Publications

Article I

Gaden, T. S., Ghetti, C., Kvestad, I., & Gold, C. (2021). The LongSTEP approach: Theoretical framework and intervention protocol for using parent-driven infant-directed singing as resource-oriented music therapy, *Nordic Journal of Music Therapy*. <https://doi.org/10.1080/08098131.2021.1921014>

Article II

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Article III

Gaden, T. S., Gold, C., Assmus, J., Kvestad, I., Stordal, A. S., Bieleninik, Ł., & Ghetti, C. (Submitted for review). Treatment fidelity in a pragmatic clinical trial of music therapy for premature infants and their parents: the LongSTEP trial.

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1. Background and theoretical framework

1.1 The global burden of preterm birth

Preterm birth is a global health problem that has major, medical, psychological, and socio-economic consequences worldwide (Chawanpaiboon et al., 2019). The World Health Organization estimates that 15 million babies are born prematurely every year, corresponding to greater than 1 in 10 babies (Howson et al., 2012). Out of these, approximately one million will not survive, which makes prematurity the second-leading cause of death in children under five years and the primary cause of death in the critical first month of life (Liu et al., 2016). Preterm birth is defined as any birth before 37 completed weeks of gestation, or fewer than 259 days since the first day of the woman's last menstrual period (Howson et al., 2012). The definition is further divided into degrees of prematurity based on infant gestational age (GA). Extremely preterm infants are born before 28 weeks GA, very preterm infants between 28- and 32-weeks GA, and moderate to late preterm infants between 32- and 37-weeks GA (Blencowe et al., 2013b). Birth weight is also used as an indicator of newborns' health, with lower birth weight indicating higher risk for complications (De Bernabé et al., 2004; Hack et al., 1995). By definition, low birth weight equals less than 2500 grams, very low birth weight less than 1500 grams, and extremely low birth weight less than 1000 grams (Hack et al., 1995).

While the rate of preterm birth is increasing in almost all countries, more than 60% of the cases occur in South-Asia and Africa (Howson et al., 2012). Due to unequal access to feasible and cost-effective treatment and care, there are also dramatic differences in survival rate and outcomes of preterm birth, depending on where in the world the baby is born. In low-income settings half of the babies born at or below 32 weeks GA die, while almost all babies in this group survive in high-income settings (WHO, 2018). More than 90% of extremely premature infants born in low-income countries die within the first days of life, while less than 10% of them die in high-income countries. Moreover, in middle-income settings, suboptimal use of

technology increases the burden of disability among preterm babies who survive (WHO, 2018).

1.1.1 Born too soon

All newborn babies are vulnerable, but premature infants are even more so. While premature infants' medical needs vary considerably depending on the degree of prematurity and related medical factors, most of them will spend weeks, and some even months, in the neonatal intensive care unit (NICU). Some premature infants primarily need time to physically grow and developmentally mature under controlled circumstances and end up with few or minor health challenges (Simeoni et al., 2004). Others need advanced treatment and care and face greater risks. The environment in the NICU also exposes neurodevelopmentally immature premature infants to potential harmful sensory input (Cheong et al., 2020). Medical advances and improved care in recent decades have increased infant survival rates following preterm birth. However, these infants of borderline viability are still at risk of developing a range of complications (Saigal & Doyle, 2008). Long-lasting sequelae of prematurity include a range of health problems such as learning disabilities, visual and hearing impairments, cerebral palsy, chronic lung diseases, and poor mental health (Blencowe et al., 2013a; Johnson & Marlow, 2011; Luu et al., 2017). Preterm birth places a burden on individuals and society, which calls for strategies to reduce long-term morbidity (Howson et al., 2012; Saigal & Doyle, 2008).

1.1.2 Premature parenthood

When a baby is born prematurely, parents also enter parenthood too soon. In most cases preterm birth occurs unexpectedly and parents' processes to mentally prepare for birth and the new baby are interrupted. Thus, their transition into parenthood is a non-normative one associated with psychological distress, as parents are forced to adjust their hopes and expectations (Gangi et al., 2013; Lasiuk et al., 2013). The normally joyful circumstances surrounding the life-changing event of the birth become more complex as parents might mourn the "hoped for baby", while still embracing the child that was born (Gangi et al., 2013). All this happens while also being thrown into the unfamiliar, highly technological, medicalized world of neonatal

intensive care. Mothers have described the emotional experience throughout their NICU stay as a “roller-coaster ride” (Redshaw, 1997), fluctuating in response to events during their baby’s recovery process. Central concerns are worries about infant survival and long-term care (Lasiuk et al., 2013; Raphael-Leff, 2018). Feelings of guilt, alienation, ambivalence towards the infant and medical staff, and doubts about one’s own caring abilities can be part of the experience (Goutaudier et al., 2011; Misund et al., 2014; Trumello et al., 2018). Evidence shows that mothers of premature infants are at greater risk of developing post-partum depression (Korja et al., 2008; Muller-Nix et al., 2004) and post-traumatic stress disorder (Brunson et al., 2021), and that fathers experience significant stress (Sloan et al., 2008). Parent mental health challenges may in turn threaten the early parent-infant relationship and bonding with their baby (Bidzan et al., 2009; Bozzette, 2007; Field, 2010; Hoffenkamp et al., 2012; Muller-Nix et al., 2004).

1.1.3 Two key models in neonatal care

Developmental care

Preterm birth means that the protected natural environment of the womb is exchanged with a highly technological and medicalized NICU environment with a range of sensory input that is unfamiliar and intrusive to the infant. The necessary and life-saving treatment and care provided to premature infants involves aspects that are potentially harmful and disturbing for the premature infant’s immature and developing nervous system. Although complications and mortality following preterm birth have been drastically reduced in high-income countries (Howson et al., 2012), premature infants’ neurodevelopmental outcomes remain a major issue of concern (Sizun & Westrup, 2004). The neuroplasticity of the developing premature infant’s brain allows for adaptation and growth (Brummelte, 2017). At the same time, medical procedures and handling also represent a risk for maladaptation in response to harmful or mismatched conditions in the environment (Als et al., 2004). Overstimulated premature infants will use considerable energy to adjust and recover – energy that could have been invested in neurodevelopment and growth (Schwartz, 2013). Examples of premature infant physiological responses to stress-inducing

noises are reduced oxygen saturation, increased rates of apnea and bradycardia, wide fluctuations in blood pressure, increased activation, in addition to agitation, crying, and sleep disturbances (Nöcker-Ribaupierre, 2013).

Developmental care is a broad category of medical and nursing interventions designed to decrease the stress of premature infants in the NICU and promote optimized organization (Sizun & Westrup, 2004). Assumptions of adequate care include control of external stimuli, clustering of nursing activities, and the physical positioning of the infant (Symington & Pinelli, 2006). Globally, developmental care is now widely accepted and practiced in most neonatal care units (Gibbins et al., 2008). While there is a consensus that premature infants need a supportive sensory environment where both sensory overstimulation and deprivation are counteracted, inconsistencies in understanding and defining what developmental care is, and the diverse ways it is applied into clinical practice, has also led to criticism and made it more challenging to draw conclusions on its effectiveness (Symington & Pinelli, 2006).

Developmental care strategies have been combined to form various programs and conceptual models, such as NIDCAP – the Newborn Individualized Developmental Care and Assessment Program (Als, 2009). Als first introduced the concept of developmental care in the 1980s with her synactive model of neonatal behavior organization (Als, 1982, 1986). Her model built on earlier work of Brazelton (Brazelton, 1974; Brazelton & Als, 1979) and was formulated as a framework for the assessment of premature infants' neurodevelopment and support for parents in the NICU environment. The synactive theory proposes that the implementation of care, in a way that takes infant's thresholds for disorganization into account, will support long-term developmental outcomes (Als & McAnulty, 2011).

In the NIDCAP model of care, individualized support and care is based on reading each premature infant's behavioral cues and using this to inform a care-plan that enhances and builds upon infant strengths, while also supporting infant areas of sensitivity and vulnerability (Als & McAnulty, 2011). Behavioral observation

provides the basis from which to estimate the most appropriate adaptations to enhance developmental outcomes. All newborns, including those who are premature or medically fragile, display observable behaviors to communicate their state and needs. White (1959) was the first to use the word “competence” in descriptions of newborns’ capacities to interact with their environment. Brazelton (1974) viewed competence as a driving-force within newborns orienting them towards development. In NIDCAP, infant competence is seen as expressed in behaviors that can be observed among four main systems: 1) the autonomic system (e.g., respiration patterns, color fluctuations, visceral responses), 2) the motor system (e.g., muscle tone, postures and movement patterns), 3) the state system (level of awareness, sleep and awake states, transition patterns), and 4) the self-regulation system (infant strategies to rebalance the three other subsystems and the success of these strategies, as well as infant responses to external regulation) (Als & McAnulty, 2011 pp. 7-8).

Als (1986) also saw the potential for caregivers to contribute in making the NICU environment a better and safer place for the premature infant through an improved understanding of the premature infant’s transition from the womb to the NICU:

The preterm infant in the NICU, who is in essence a displaced fetus, emerges as keenly sensitive to all parameters of environmental impingement. This sensitivity of the very young nervous system provides a unique opportunity for the caregiver to make the alien environment organism-appropriate and developmentally supportive, rather than a hazardous place of increased stress. To improve the environmental structure and timing, input and experience of careproviders need to be based on a better understanding of the functioning of the transitional newborn infant. (Als 1986, p. 11)

Als also describes NIDCAP as a relationship-based intervention (2009) proclaiming the collaborative and mutually respectful relationship between each family and staff as the most important aspect of NIDCAP. This relationship is something that develops over time. In today’s description of the model the contributions of all involved parties in this collaborative relationship are highlighted:

Infants are considered individuals, persons, collaborators in care, supported and nurtured by their parents. Parents are considered infants' key nurturers, advocates and caregivers as well as collaborators in care decisions. Healthcare Professionals are partners in care with infants, parents and family members. (NIDCAP Federation International, n.d.).

The emphasis of the collaborative partnership between families and healthcare highlights how NIDCAP is also an example of family-centered care.

Family-centered care

Family-centered care (FCC) is a way of caring for children and their families within health services that ensures that care is planned around the whole family, not just the individual child. In FCC all family members are recognized as care recipients (Shields et al., 2006, p. 1318). Other terms have been used to connote similar approaches, including parental participation, care-by-parent, and partnership in care. These were all precursors that contributed to current pediatric care where FCC is recognized as a cornerstone (Jolley & Shields, 2009) and gold standard for practice (Cooper et al., 2007; Gooding et al., 2011). The consensus definition of FCC by the Academy of Pediatrics emphasizes a respectful family-professional partnership where strengths, cultures, traditions, and expertise brought by everyone involved is honored (Arango, 2011, p. 97). In the context of neonatal care FCC, an organized collaboration between families and physicians led to the establishment of The Principles for Family-Centered Neonatal Care (Harrison, 1993). These principles marked a paradigm shift within neonatal care and enabled greater integration of FCC within best practice (Johnson, 2000; Jolley & Shields, 2009). Later, the European Research Network on Early Developmental Care proposed eight procedures to be considered as principles of care in NICU-settings (Roué et al., 2017). The eight procedures are based on a high level of evidence and aim to meet infants' and families' universal needs during the critical neonatal period and include: 1) free 24 hour a day parental access with no limitations due to staff shifts or medical rounds, 2) psychological support for parents, 3) pain management, 4) a supportive environment, 5) postural support, 6) skin-to-skin contact, 7) breast feeding and lactation support,

and 8) sleep protection (Roué et al., 2017). Evidence supports that FCC can reduce the overall length of hospitalization and intensive care treatment, as well as positively impact feeding, growth, and neurodevelopment in hospitalized premature infants (Yu & Zhang, 2019). Parents who actively participate in the treatment and care of their baby alongside nursing staff demonstrate improved caregiving abilities and report less anxiety and depression (Ding et al., 2019).

Kangaroo Care

Kangaroo care (KC), also known as kangaroo mothering¹, and skin-to-skin care, is an example of a practice that integrates developmental care and family-centered care principles. During KC the infant is placed in an up-right position, skin-to-skin on a caregiver's chest (Ludington-Hoe et al., 2008). KC is recommended for infants from 28 weeks GA; however, evidence suggests that KC also benefits infants below this age (Clifford, 2001; Hunt, 2008). While it has many physiological benefits to premature infants, including cardio-respiratory stabilization, improved autonomic regulation, and increased weight gain, findings also suggest benefits to parents and the parent-infant bond (Hunt, 2008). Kangaroo care allows for higher incidents of breastfeeding, it reduces stress and anxiety in mothers; and also increases fathers' confidence in their caregiver role and promotes parent-infant bonding (Blomqvist et al., 2012; Conde-Agudelo et al., 2012; Fegran et al., 2008; Ludington-Hoe et al., 2008).

1.2 Bonding

Like in any other mammal, survival, healthy development, and the well-being of the human infant depend on the proximity and care of a parent or other close caregiver (Bowlby, 1969, 2008). There must be a certain bond – a sense of belonging, positive emotions, and responsibility perceived from the adult side, and an experience of care, protection, and predictability from the infant side. A parent-infant bond is generally

¹ Kangaroo mothering does not refer to just maternal-infant contact. Any close caregiver can provide KC to the infant (Ludington-Hoe, 2011).

considered to be unique and emotionally charged, based on presence, availability, sensitivity, and satisfaction of needs. The Merriam Webster dictionary ("Bonding") gives a common-sense definition of bonding as "The formation of a close relationship, as between a mother and child (...) especially through frequent or constant association". In psychology, sociology, nursing, and medical literature, bonding is usually used when referring to parents' emotional bond – predominantly the mother's perception of the bond towards her infant. Bonding is also widely referred to in MT literature from the settings of family work, pediatrics, and neonatal care (Edwards, 2011a, 2011b). Other terms such as maternal-infant bonding, mother-infant relationship, maternal bonding, and attachment are also used, although the latter refers to a distinct concept as distinguished below. I will use "bonding" when referring to the mother-infant bond in this thesis, and I lean on Kinsey and Hupcey's (2013) definition from their principle-based concept analysis:

Maternal-infant bonding is a maternal-driven process that occurs primarily throughout the first year of an infant's life, but may continue throughout a child's life. It is an affective state of the mother; maternal feelings and emotions towards the infant are the primary indicators of maternal-infant bonding. Behavioural and biological indicators may promote bonding, but are not sufficient to determine the quality of maternal-infant bonding nor are these indicators unique to the concept. (p. 1319)

This definition covers bonding in the post-partum period. However, it is important to add that maternal infant-bonding is a process that begins before the child is born, from early pregnancy lasting throughout it, and continuing after birth (Brandon et al., 2009; Ettenberger et al., 2021; Glover & Capron, 2017). Many mothers experience that the relationship with their unborn baby intensifies in the transition from the second to third trimester, once they feel the first fetal movements and the baby increasingly responds to external stimuli such as the mother's voice and touch (Cataudella et al., 2016; Ettenberger et al., 2021). In the case of preterm birth, this prenatal bonding and parental psychological preparation to parenthood is interrupted, causing stress which may negatively impact post-natal parent-infant interactions and

bonding (Cataudella et al., 2016; Cook et al., 2018; de Paula Eduardo et al., 2019; Muller-Nix et al., 2004).

1.2.1 Attachment and mutual regulation

Establishing healthy parent-infant bonding lays the foundation for the later development of infant attachment to caregivers (Ettenberger et al., 2021; Kinsey & Hupcey, 2013). Attachment refers to the child's emotional bond and relationship to their closest caregivers, normally their parents, which develops through close interplay and interaction throughout the first years of life (Bowlby, 1969).

Attachment theory was originally developed by Bowlby (1969) and further refined by Ainsworth who proposed the concept of different attachment styles that affect how infants relate and react to their environment (Ainsworth, 1964, 1969; Ainsworth & Bowlby, 1991). Children with secure, ambivalent, avoidant, or insecure attachment styles demonstrate different emotional and behavioral strategies in the face of stressful situations (Ainsworth et al., 2015). Where the securely attached infant will use their attachment figures as a base to explore the world from and return to them when facing a threat, insecurely attached infants may avoid their caregiver (Ainsworth et al., 2015; Bowlby, 2008). The child's pattern of attachment develops gradually from birth and is normally manifested as an identifiable attachment style around the age of 18 months (Ainsworth, 1979; Bowlby, 1969). Attachment style impacts later relationships and other social and developmental outcomes later in life (Winston & Chicot, 2016). An increased risk for disorganized attachment has been demonstrated in infants born very preterm or with very low birth weight, compared with full-term infants (Wolke et al., 2014).

In modern attachment theory of recent decades, Bowlby and Ainsworth's ideas have been expanded upon and evolved into a more complex and clinically relevant model that includes neurobiological processes such as affect and self-regulation (Schore & Schore, 2008). Self-regulation refers to the capacity to regulate emotions, cognition, and behavior, and is widely recognized for its important role in promoting healthy development and wellbeing across the lifespan (Murray et al., 2015). Infants develop their capacity for self-regulation through external regulation and co-regulation with

their parents. Mutual regulation (or co-regulation) happens when parents sensitively attune to infant cues and provide nurturing support through responsive actions (Samdan et al., 2020; Tronick, 1989). Mutual regulation is reciprocal and informed by the infant, parent, and mutual processes between them (Beeghly & Tronick, 2011). Successful mutual regulation can confirm parents' perception of themselves as good caregivers for their child (Landsem et al., 2015). However, mutual regulation can be threatened by maternal stress and mental health challenges (Choe et al., 2013; Tu et al., 2007).

1.2.2 Bonding following preterm birth

Traumatic events around the time of childbirth, physical or emotional separation from the infant following birth, and maternal mental health challenges may threaten healthy parent-infant bonding (Kinsey & Hupcey, 2013). In the case of preterm birth several of these conditions are present. Additionally, preterm infants are more difficult to interpret and understand as their behavior states change rapidly and communication cues are subtle (Bozzette, 2007). The combination of premature infants' inconsistent and often compromised communicative capacity with parents, who might be less sensitive because of psychological distress, can result in mismatched parent-infant interaction and overstimulation of the infant (Sansavini et al., 2015). This in turn, may affect healthy parent-infant bonding (Gutbrod & Wolke, 2004; Kim et al., 2020; Trumello et al., 2018). While a significant portion of literature demonstrates that preterm birth poses a risk for the parent-infant bond, there are also examples of the contrary where studies have found good mother infant-bonding following preterm birth (e.g., Bieleninik et al., 2021). A study comparing the bonding process of parents of preterm infants and parents of full-term infants found that parents of preterm infants had fewer bonding problems (Hoffenkamp et al., 2012). In a study investigating whether infants' gestational age at birth influenced the quality of the mother-infant and father-infant bond, mothers of preterm infants even reported higher feelings of bonding than mothers of full-term infants (Hall et al., 2015). For fathers, no differences were found between the gestational age groups (Hall et al., 2015). The evidence of the impact of preterm birth on parent-infant bonding is hence inconclusive, and more research is needed.

1.3 Voice, song and the parent-infant relationship

The capacity of hearing develops to become fully functional during the second trimester of pregnancy when the fetus is around 25 weeks of gestation (Graven & Browne, 2008). However, experiments measuring fetal movement in response to external sounds have observed responses as early as 19 weeks of gestation (Hepper & Shahidullah, 1994). The first sounds the fetus hears in-utero are the internal, bodily sounds of the mother (heartbeat, breath, circulation, and digestion), as well as external sounds related social activities and voices muffled by the amniotic fluid (Moon, 2011). After birth, full-term newborns demonstrate a preference for their mother's voice over a stranger's voice (DeCasper & Fifer, 1980). Newborns, including premature infants, also show a clear preference for infant-directed over adult-directed speech (Butler et al., 2014; Cooper & Aslin, 1990; Fernald, 1985; Pegg et al., 1992). The terms "protoconversation" (Bateson, 1975), "motherese" (Fernald, 1985; Newport, 1975), and "infant-directed (ID) speech" all refer to the ways in which adults adjust their way of speaking when interacting with infants. Infant-directed speech is characterized by shorter utterances, slower tempo, longer pauses, higher pitch, more exaggerated pitch contours and rhythmical contours, compared to when talking to adults (Fernald et al., 1989; Trainor et al., 2000). The concept of ID speech was later applied to singing (Trainor, 1996) and has been found to elicit many of the same types of infant responses as ID speech such as capturing attention and regulating arousal (Corbeil et al., 2013; Corbeil et al., 2015). Infant-directed singing, however, offers additional structure through musical form, including more predictable and distinct phrasing, melodic range, and rhythmical patterns (Trehub & Trainor, 1998; Trehub et al., 1997). Live maternal singing can increase physiological stability and positive communicative behavior of premature infants (Filippa et al., 2020; Filippa et al., 2017).

Parents singing for their babies is an act of caregiving behavior found across cultures and has likely always been around (de l'Etoile, 2006; Dissanayake, 2009). Soothing lullabies and lively play songs sung by the baby's closest caregivers have unique regulating qualities and offer a way for parents and their child to share

intimate moments of connection. Malloch and Trevarthen (2009) argue that we are all born with the innate capacity of “communicative musicality”, which plays a vital role in early communication and the formation of early relationships. Communicative musicality is expressed in parent-infant interaction through the parameters pulse, quality, and narrative (Malloch, 1999; Malloch & Trevarthen, 2009). When mothers and infants communicate effectively, they are attuned to each other’s vocal and physical gestures, and these are appropriately timed and inflected (Malloch, 1999). Together, mother and infant are partners in a musical dialogue where movement – gestural, vocal, and emotional – allows communicative musicality to occur (Malloch, 1999 p. 49). In the attempt to define music’s therapeutic potential, Trevarthen and Malloch (2000) also point to music’s multi-faceted capacity to move humans:

We believe that music is therapeutic because it attunes to the essential efforts that the mind makes to regulate the body, both its inner neurochemical, hormonal, and metabolic processes, and in its purposeful engagements with the objects of the world, and with other people. Music, with dance and all the expressive arts, offers a direct way of engaging in the human need to be sympathized with – to have what is going on inside appreciated intuitively by another who may give aid and encouragement. (p. 11)

Their description illustrates the potential within musical interaction for meaningful experiences of relationship. It also addresses music’s regulating qualities, which in the case of singing may be considered particularly relevant. Compared to other musical instruments, singing is unique in the way that the source of music – the voice, is created from and resonates within the body itself. When a premature infant lies skin-to-skin on their parent’s chest and the parent sings, the vibrations of the voice provide a sensory experience that is not only heard but also felt through the infant’s body. Furthermore, singing not only affects the infant being sung for, but also the person singing. Singing affects breathing patterns, which provide a potential pathway to work with self-regulation and stress-reduction in parents (Gick, 2011). While singing, parents may experience a reduction in arousal, which can enable them to more sensitively attune and respond to their infants (Fancourt & Perkins, 2018a,

2018b). The voice is also deeply personal, allowing for expression of emotions and identity (McLean, 2016; McLean et al., 2019). In sum, parent-led ID singing has many unique qualities and can provide a frame where infant and parents can be together, co-create, co-regulate, and experience relationship.

1.4 Music therapy

Music therapy (MT) can be defined in a range of ways, and definitions are colored by distinct MT traditions and theories. The definition in the Oxford Handbook of Music Therapy entails relational aspects that resonate with the MT approach of LongSTEP:

Music therapy is a relational therapy involving the use of music in therapeutic processes with individuals or groups by a qualified practitioner who has undertaken appropriate training and undertakes ongoing professional development. It is a unique way of working in which the dynamic capacities of music and musical relating, are harnessed to serve the needs of the client, family or group who is seeking help. (Edwards, 2016, p. 2)

The emphasis on a qualified practitioner is also central. Music therapists develop a professional, clinical competence over years of education and practice, and use methods that are distinct to other music approaches and practices applied in health or community settings (e.g., by pedagogues or professional musicians).

1.4.1 Resource-oriented music therapy

The MT in LongSTEP is strongly influenced by Rolvsjord's resource-oriented approach to MT. Four sensitizing concepts characterize resource-oriented MT and serve as guiding values for our MT approach. These include 1) the nurturing of strengths, resources, and potentials; 2) collaboration rather than intervention; 3) viewing the individual within their context; and 4) seeing music as a resource (Rolvsjord, 2010, p. 74). Resources in this case involve the person's musical resources, including musical competence and potentials such as instrumental skills, knowledge of a repertoire of songs, and singing ability, as well as access to music (through, e.g., choirs, bands, music listening technology), and the social participation

and relationships that can be fostered through it (Rolvsjord, 2020 p. 75). The theoretical development of resource-oriented MT by Rolvsjord (2010) was originally situated in the context of mental health care with a particular focus on individual psychotherapeutic work. However, Rolvsjord explicitly states a hope that primary concepts and principles of resource-oriented MT can be transferable to other fields of MT practice (2010, p. 74). Resource-oriented MT is based on a contextual model of psychotherapy, “(...) a model that encompasses the whole therapeutic situation as promoter of change, with specific emphasis on the client’s own effort and competence related to the therapeutic process” (Rolvsjord, 2010 p. 12). The collaborative therapeutic relationship of a resource-oriented approach is characterized by equality, mutuality, and participation.

In line with a resource-oriented approach (Rolvsjord, 2010) we recognize and nurture parents’ and infants’ unique strengths, resources, and potentials. Premature infants are viewed as competent and actively contributing to shaping the musical interaction through expressing their needs and responding to music with behavioral cues. Parents are positioned as experts on their own infants, and their voices are considered a key resource when aiming to strengthen parent-infant bonding. The music therapist contributes to the therapeutic process with musical expertise, but we intentionally challenge positioning the therapist as an expert. We aim to promote equality and mutuality in the therapeutic relationship where all involved parts share responsibility and collaborate on what happens in MT (Rolvsjord, 2010).

1.4.2 Music therapy in neonatal care contexts

Music therapy has gradually been introduced to the context of neonatal care through the research and development of clinical approaches to meet the different needs premature infants and their families experience during NICU hospitalization. Early contributions to MT in the NICU field were largely infant-focused, used recorded music and primarily emphasized physiological and behavioral aspects such as heart rate (HR), respiratory rate (RR), oxygen saturation level, sucking response, feeding behavior and weight gain (Cassidy & Standley, 1995; Standley, 1991; Standley, 1998, 1999; Standley, 2015). Current MT practices in neonatal care involve a range

of approaches where music is used to promote infant development, support parents' psychosocial needs and facilitate the interaction and relational processes between them (Nöcker-Ribaupierre, 2013). However, the degree to which MT is established in routine clinical practice still varies greatly globally (Shoemark & Ettenberger, 2020), and more research into different aspects of what MT can offer families following preterm birth is needed (Bieleninik et al., 2016a; Yue et al., 2021).

Literature review: MT with premature infants and their parents

Since this thesis centers around a MT approach with a strong emphasis on parent participation, I delimited the scope of the review to literature concerning MT in NICU and follow-up where parents are involved as active participants. Furthermore, studies had to include parent outcomes and/or parent-infant mutual outcomes. Literature concerning other types of music interventions (e.g., recorded music, including recordings of parental voice, or hospital musicians) in neonatal care settings was excluded (Appendix 1 Search strategy and inclusion criteria). This overview is based on systematic database searches and manual searches and includes 34 peer-reviewed articles (Appendix 2 Reviewed literature).

The included articles were published between 2000 and 2022 in 23 journals and include contributions from fifteen different countries. A significant portion of the included literature comprises recent feasibility or pilot studies and reports from implementation processes (Bieleninik et al., 2020; Coombes & Muzaffar, 2021; Ghetti et al., 2021; Janner et al., 2021; Kehl et al., 2021; Loscalzo et al., 2021; Menke et al., 2021; Van Dokkum et al., 2020). These contributions show how the field is expanding regarding both clinical practice and research. Eighteen of the articles were published during the last two years, which is a noteworthy increase in studies of MT in NICU with parent participation.

Two systematic reviews and meta-analyses include RCTs with both infant and parent outcomes. A 2016 meta-analysis confirmed large favorable effects of MT on premature infants' respiratory rate and maternal anxiety (Bieleninik et al., 2016). A recent meta-analysis found that MT can effectively improve infant heart rate,

respiratory rate, oral feeding volume and stress level. Furthermore, the effect on maternal anxiety was confirmed (Yue et al., 2021). Both meta-analyses address that there is considerable heterogeneity among the included studies for many outcomes. Rigorously designed studies with larger sample sizes, that address both short-term and long-term outcomes for parents and infants, are thus called for (Bieleninik et al., 2016; Yue et al., 2021).

In contrast to research from the 1990s up until the early 2000s, which was predominately infant-focused and largely from the US, the literature in this review represents a larger selection of countries and outcomes, which might increase the applicability of the findings. Music therapy in NICU has grown substantially in Europe the last decade, and literature from 10 different European countries is included in this review. However, the search did not identify any literature from African countries, and very few studies from Asia. This may reflect the unequal implementation status of NICU MT globally, as well as differences in the degree to which parent participation is an area of interest. There are substantial differences between countries in the degree to which FCC and parent participation is an emphasized and established part of NICU standard care. This likely influences the aims and methods of MT in NICU where it is implemented. The reviewed literature reveals an increased interest in parent and parent-infant mutual outcomes of MT from around 2008 and onward. This can be interpreted to align with the ongoing shift towards FCC in neonatal care.

Several of the included studies emphasize FCC principles, such as planning MT around both parent and infant needs and including parents as active participants (Ettenberger et al., 2017; Kobus et al., 2021; Loscalzo et al., 2021; Menke et al., 2021; Shoemark & Dearn, 2008; Ullsten et al., 2021). Several forms of parent participation in MT are mentioned in the literature. Examples include singing (Haslbeck, 2014; McLean, 2016; Palazzi et al., 2021a; Palazzi et al., 2021b; Shoemark, 2018), often in combination with Kangaroo Care (Epstein et al., 2020; Ettenberger et al., 2017; Kehl et al., 2021; Kostilainen et al., 2020). Parent singing has also been suggested as part of neonatal procedural support and pain management

(Ullsten et al., 2021). Songwriting and modifying songs, as well as playing instruments, are other ways parents engage (Álvarez et al., 2021; Ettenberger & Ardila, 2018; Loewy, 2015; Loscalzo et al., 2021). Educational approaches, such as parent workshops, and single-session training/supervision in techniques, such as multi-modal stimulation and singing, have been suggested as complimentary approaches to MT (Coombes & Muzaffar, 2021; Shoemark, 2018; Whipple, 2000). Families who have infants with few complications and shorter NICU stays might benefit from such formats as they provide parents with musical and relational tools they can use on their own.

While most studies exemplify what parent participation in MT may entail, the studies differ substantially in the level of details they report regarding the degree to which parents attended sessions and the potential reasons why some did not participate where participation was low. In studies where KC and MT are combined, parents actively participate through their provision of skin-to-skin contact, but it is often left somewhat unclear as to how active parents have been musically (e.g., if they have participated in singing or if the music therapist has been the sole provider of music).

Only a few of the included studies explicitly address the participation of fathers (Bieleninik et al., 2020; Ghetti et al., 2021; Loscalzo et al., 2021; Mondanaro et al., 2016). Music therapy is well suited to meet the needs of fathers in the NICU through providing a culturally sensitive and normalizing environment where fathers can connect to their child and their own emotions and take-on an active role in the NICU environment (Mondanaro et al., 2016). Loscalzo et al. (2021) developed a MT approach based on Creative music therapy and the Rhythm, Breath, Lullaby-model called “Premature Triadic Music Therapy”, which specifically targets the inclusion of both mothers and fathers and was found feasible in an Italian NICU setting. Due to differences in access to paternity leave, particular efforts may be necessary to increase the participation of fathers in MT in some countries, although fathers may be more available to participate in MT after discharge (Bieleninik et al., 2020). Family-structures are changing in society, but this diversity is not yet reflected in the

literature. MT with premature infants and their parents is a largely heteronormative field, and no studies were found that describe MT with single or LGBTQIA² parents.

Qualitative studies provide insight to parent experiences with participation in MT. Singing can help validate parental identity and provide parents with emotional catharsis and self-coping practices, as well as a way of connecting with their child (McLean, 2016; McLean et al., 2019). While singing can provide parents opportunities to connect with their child, being asked to sing in front of others can be challenging (Bieleninik et al., 2020; Epstein et al., 2022; Ghetti et al., 2021). Singing is personal and parents may feel uncomfortable, embarrassed, and shy about it (Bieleninik et al., 2020). Singing without instrumental accompaniment may increase insecurity, but it also provides an opportunity for parents to discover their own singing ability and how their voice alone affects their baby (Epstein et al., 2022). Not surprisingly, parents recommend that MT should happen in individual rooms (Bieleninik et al., 2020; Ghetti et al., 2021).

Where previous literature has recommended cautiousness with introducing MT too early, some of the included studies evaluate the feasibility of introducing MT to more vulnerable patient groups (Epstein et al., 2020; Kehl et al., 2021; Van Dokkum et al., 2020). Extremely and very preterm infants have been found stable enough to participate in MT with live music, without signs of overstimulation (Van Dokkum et al., 2021). However, negative effects of MT were found in a study with preterm infants with severe brain injury and their mothers (Epstein et al., 2020). Infants demonstrated increased physiological and behavioral instability, and mothers had an increase in anxiety. More knowledge on when MT is contraindicated is needed. MT with particularly vulnerable infants and families may also require that MT is therapist-led to a larger degree, as specific knowledge on how to carefully tailor music to the infant is necessary to ensure that music does not cause harm (Van Dokkum et al., 2021). Although most premature infants stabilize and survive, MT in NICU may also involve end-of-life practices in palliative stages of treatment and

² LGBTQIA is an acronym for lesbian, gay, bisexual, transgender, queer/questioning, intersex, and asexual/aromantic/agender (Merriam Webster Dictionary, 2022).

when a child has passed (Ettenberger, 2017). Memory-making and rituals are central themes in such practices. Tailoring the MT to the family's needs, as well as being sensitive to culture-specific beliefs and practices concerning death, is crucial for end-of-life care.

While the evidence-base of MT during NICU demonstrates growth, the literature searches resulted in very few studies concerning MT after discharge from hospital. Two studies have investigated benefits of participation in parent-infant developmental music groups and found improvements in infants' social, cognitive, and communicative skills, as well as parent responsiveness (Hamm et al., 2017; Walworth, 2009). A clinical model in development for MT with NICU discharged dyads "CoPE with Music" suggest that MT post-discharge can restore mothers' parental efficacy and dyadic communication abilities, which are often negatively affected by preterm birth and NICU hospitalization (Calderon-Noy & Gilboa, 2021). In a retrospective qualitative inquiry, parents who had participated in MT during NICU seven years before reported that music continued to be a central part of their family life after discharge (Haslbeck et al., 2021). Some highlighted how they used the music intentionally to regulate and cope. Others called for MT to be continued after discharge (Haslbeck et al., 2021). More research on how MT can support families in the transition from NICU to home, and after discharge, is needed. This identified gap in the literature is addressed in the LongSTEP trial where the effect of MT during both NICU and post-discharge is investigated. Two LongSTEP feasibility studies from Norway and Poland concluded that MT post-discharge from hospital was relevant and feasible (Bieleninik et al., 2020; Ghetti et al., 2021). An Israeli LongSTEP cohort of parents who participated in MT post-discharge experienced that MT improved parent-infant communication skills and parental agency, and positively impacted the relationship with their baby (Epstein et al., 2022).

The LongSTEP trial

Through conducting a rigorously designed, large-scale, multinational randomized controlled trial, we aim to contribute to the knowledgebase with high quality evidence of the long-term effects of MT on parent-infant bonding, parent mental

health, and infant development. A key feature of LongSTEP is the provision of MT both in the NICU phase and/or for a six-month follow-up period after discharge. The MT emphasizes parent participation through singing and use of voice, optimally with both parents present and actively engaged. Assessments are completed up until the child reaches 24 months corrected age (Ghetti et al., 2019). To our knowledge, LongSTEP is the first RCT on MT with premature infants and their parents to follow families through the infants' first two years of life. Through our study we aim to contribute to a deeper insight into the potential benefit of MT in different stages following preterm birth.

1.5 Aims of the thesis

This thesis consists of three substudies, for which all data were collected in the LongSTEP trial. The substudies were chosen to fulfil parts of the LongSTEP trial's aims and were designed accordingly. All three substudies concern different aspects of the MT intervention and had the following aims:

I) *Theoretical framework and intervention protocol:*

To thoroughly describe and expand upon the theoretical framework of LongSTEP's MT approach and present the guiding principles and operationalized elements of the MT intervention protocol.

II) *Short-term results of the RCT:*

To measure the short-term effects of the LongSTEP MT approach, reporting the results of the RCT from the timepoint of discharge from NICU.

III) *Treatment fidelity in the LongSTEP trial:*

To develop and evaluate treatment fidelity procedures and assessment tools. To assess treatment fidelity in the areas of treatment delivery – whether the MT was implemented in a uniform way across the different therapists and contexts of the study and treatment receipt – and to what extent parents perceived the MT to be in line with essential elements of the protocol.

2. Design, materials, and methods

To situate the substudies of this thesis in relation to the main trial, methodological aspects of the LongSTEP trial that apply to all three substudies will be provided before describing specifics unique to each substudy.

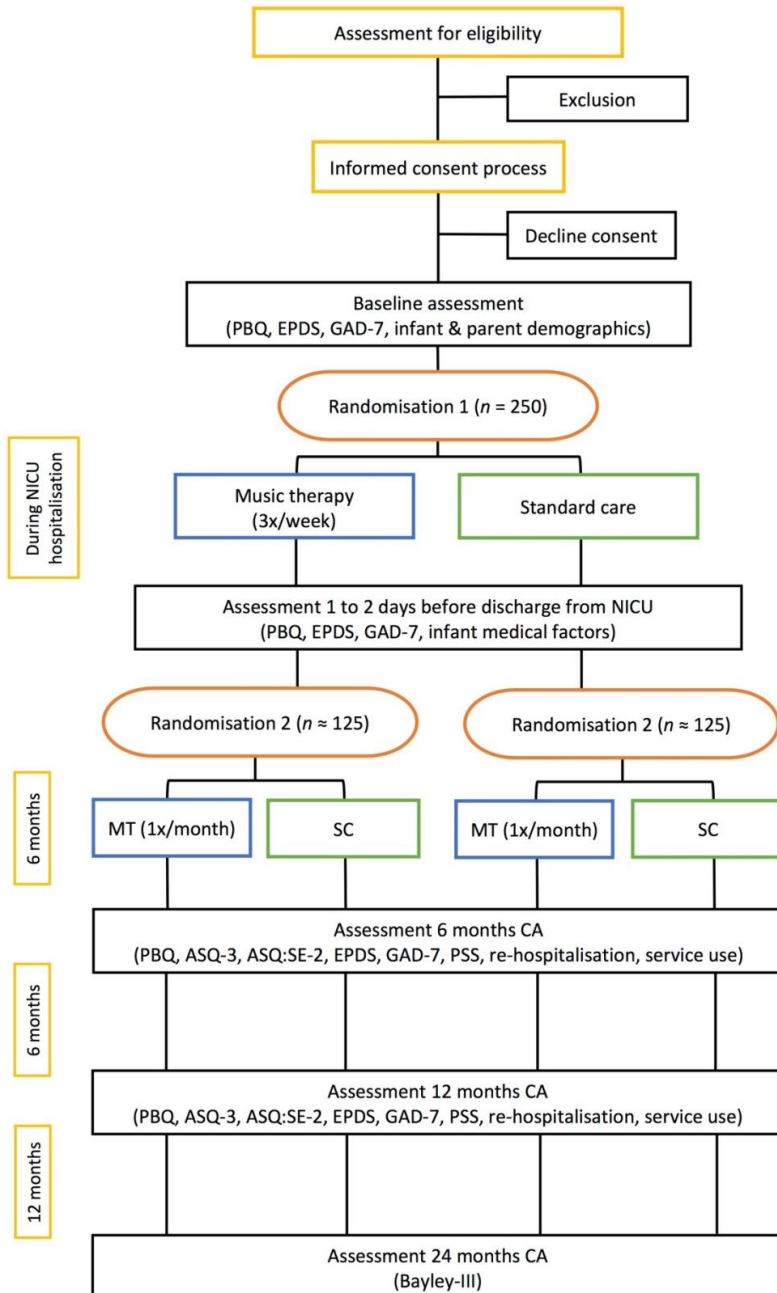
2.1 Design

LongSTEP is designed as a multinational, multicenter, assessor-blind, 2×2 factorial, pragmatic randomized controlled trial (Ghetti et al., 2019) informed by two feasibility studies (Bieleninik et al., 2020; Ghetti et al., 2021), enrolling families with preterm infants from eight NICUs in Argentina, Colombia, Israel, Norway, and Poland. The main study's primary objective was to investigate changes in mother-infant-bonding at the infant's six-months corrected age. Secondary objectives were investigating mother-infant bonding at the timepoints of discharge, 12 and 24 months, as well as child development at 6, 12 and 24 months; and parent mental health outcomes, including maternal depression, parent anxiety and stress, over 12 months (Ghetti et al., 2019) (See Figure 1 for LongSTEP study design and participant flow).

LongSTEP is registered at clinicaltrials.gov with the identifier NCT03564184.

A pragmatic approach was deemed appropriate for the LongSTEP trial to accommodate for differences between sites and maximize the applicability and relevance of the findings. Some of our sites had not yet systematically implemented MT in their NICUs, and applicability to local routine care and practice was considered of particular interest and importance for the relevance and successful implementation of the study.

Figure 1 LongSTEP study design and participant flow, reprinted from Ghetti et al. 2019



2.2 A pragmatic trial

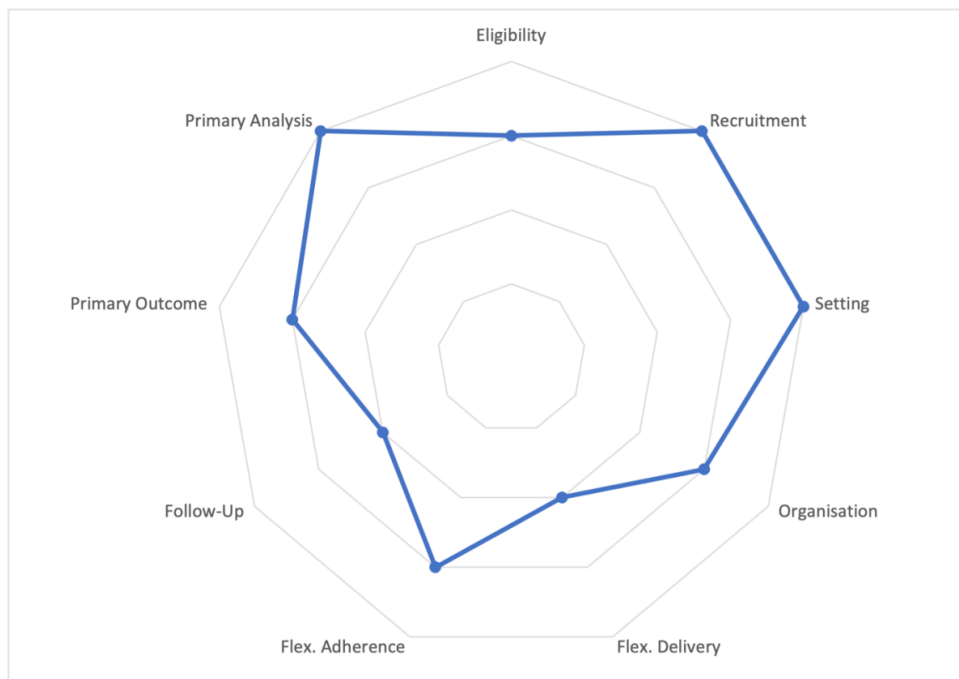
LongSTEP is a pragmatic trial, measuring *effectiveness* – the benefit the treatment produces in routine clinical practice. This as opposed to what is referred to as explanatory trials that measure *efficacy* – the benefit a treatment produces under ideal conditions often using carefully defined subjects in a research clinic (MacPherson, 2004). Research collaborations within the field of MT are increasingly becoming more international, particularly in the context of large randomized controlled trials where multisite collaborations are often necessary to assure a sufficient sample size. However, internationally conducted research also leads to multiple complexities in terms of cultural, linguistic, social, and practice-related differences. Pragmatic trials have gained popularity in recent decades due to their potential for wide generalizability and clinical applicability since therapies and interventions are evaluated as they would be used in usual care and routine clinical settings, accommodating for heterogeneity among participants, treatments, and contexts (Patsopoulos, 2022). Loudon et al. (2015) have proposed a four-step tool for aiding in the design of pragmatic trials – the PRECIS-2. The tool involves evaluation of where nine different domains of the trial are placed on an explanatory-pragmatic continuum, which consists of a 5-point scale ranging from very explanatory (1) to very pragmatic (5). The tool is to some extent subjective, and the scores of the domains in Table 1 and Figure 2 represent my understanding of the levels of pragmatism in LongSTEP. Overall, LongSTEP is a rather pragmatic trial. Study settings are identical to usual care, and inclusion criteria are intentionally broad to reflect real-life heterogeneity. On the domains such as flexibility of intervention delivery and follow-up, however, LongSTEP is leaning slightly towards a more explanatory approach. The intervention is provided in accordance with a protocol, and we have procedures to assess fidelity, while follow-up includes visits that would not have been part of routine clinical practice.

Table 1 Scoring of the PRECIS-2 domains

Domain	Score	Comment
Eligibility	4	Participants were similar to those who would receive MT in usual care but some limitations regarding GA and medical stability
Recruitment	5	Recruitment happened through usual engagement with participants in clinic
Setting	5	Settings were identical to usual care
Organization	4	Some additional resources were required to train music therapists and assessors
Delivery	3	MT was delivered in line with flexible intervention protocol, supervision was provided to improve compliance, and assessment of fidelity of delivery after intervention period
Adherence	4	Participants were encouraged to participate in a more active manner than might be the case in usual care MT.
Follow-up	3	Follow-up included more appointments and contact than usual with MT sessions and assessments post-discharge
Primary outcome	4	The primary outcome was relevant and of importance to participants, but based on inconclusive evidence
Primary analysis	5	An intention-to-treat analysis was done, including all available data

Note: See Loudon et al (2015) and <https://www.precis-2.org/Help/Documentation/HowTo> for scoring system and description of domains.

Figure 2 PRECIS-2 Wheel



Note: The PRECIS-2 wheel is a graphic representation of the explanatory-pragmatic continuum, where the outer ring indicates a very pragmatic approach. Each step inwards decreases the degree of pragmatism.

2.3 Participants and study settings

Preterm infants and their parents in the LongSTEP trial were recruited from seven level III and one level IV NICU³ in Argentina, Colombia, Israel, Norway, and Poland. Eligible premature infants were (1) born before 35 weeks gestational age (GA), (2) likely to be hospitalized at least two weeks from inclusion, and (3) declared by NICU staff as medically stable to start MT (typically after 26 weeks post-menstrual age (PMA)). In addition, parent inclusion criteria included (1) written, site-specific informed consent; (2) a willingness to engage in at least two of three MT

³ Level III NICUs are hospital NICUs organized with personnel and equipment to provide continuous life support and comprehensive care for extremely high-risk newborn infants and those with critical illness. Level IV NICUs provide the highest level of neonatal care at a regional level with the full range of specialized services (Barfield et al., 2012).

weekly sessions; (3) sufficient understanding of the respective national language(s) to answer the questionnaires and participate in MT; and (4) the capacity to complete the intervention and questionnaires (Ghetti et al., 2019).

The participating NICUs had staff available with the necessary scientific and clinical expertise to conduct the trial, including neonatologists, nurses and at least one master's degree prepared music therapist.⁴ A key characteristic common for the included sites was high parental presence in the NICU, with one or both parents being present daily.

LongSTEP is a multinational and culturally diverse study. While the sites had in common high levels of parent presence in the NICU, the collaborating countries differ from each other regarding many contextual and cultural factors, such as organization of health care, and attitudes and practices of music and singing in everyday life. The place of MT in neonatal care is likely governed by such contextual and cultural factors (Shoemark & Ettenberger, 2020). To recognize this diversity and provide a more nuanced backdrop for the study, our methodological choices, and ultimately our findings, I will provide a more detailed description of the study settings than we have been able to publish elsewhere. The descriptions are based on site visits I did for the LongSTEP trial in 2019 and personal communication with our research partners, where they were asked to identify site-specific characteristics that were important to them. The purpose of the site visits was to oversee study progress and conduct data quality auditing and furthermore gain first-hand insight into site-specific challenges and strengths. The site visits were also an opportunity for learning about contextual and cultural factors that shaped our research partners' and participants' everyday lives, and ultimately our research.

2.3.1 Argentina

The Argentinian site Hospital Fernandez (HF) in Buenos Aires is a public hospital serving some of the most vulnerable families in the district. It is known for its high-

⁴ Three of the eleven music therapists were in the finishing stage of their education and had completed all clinical components of the training but had not yet completed their final thesis.

quality services, and many travel from quite far to access the care there. Argentina is a conservative, Catholic country, and when I visited in 2019 the call for the right for safe, legal abortions was very visible in the public debate. Teen pregnancy numbers were high, particularly in rural areas, which was also notable in the NICU at HF where several young mothers were admitted after preterm birth. Healthcare in Argentina is primarily public, and mothers receive economic compensation to be with their child during NICU hospitalization. The NICU at HF is a small and quite crowded open-bay style unit, consisting of two rooms: one with incubators and one with open cribs for more stable infants. The room with incubators is quite noisy, possibly due to less modern medical equipment. A couple of rocking chairs are available for parents to sit by the incubators, but there are fewer chairs than the number of incubators/beds. A private foundation funds facilities to stay overnight for mothers with a long traveling distance. The NICU have a well-established KC program, but MT was introduced to the NICU there for the first time through the study. Argentina has a strong musical tradition and is particularly known for Tango music. Folkloric music also has very strong roots in some regions. People are used to singing as part of social settings and celebrations, but the level of comfort with singing varies individually.

2.3.2 Colombia

In Colombia participants were recruited from two sites: Hospital Fundación de Santa Fe Bogotá (FSFB) and Clínica de la Mujer (CM), both located in the capital Bogotá. The two hospitals are private, modern hospitals serving a population from low-to high income economic backgrounds, with the majority being middle class citizens with additional private health care insurance. Colombia's health care system is partly public, and partly private, but private health care is dominant. Citizens are obliged to have health insurance and choose insurance plans with different ranges of coverage from private or public companies. Colombia is a society with high levels of inequality and large socioeconomic gaps, which affects the accessibility, quality, and continuity of health care (Ardila et al., 2020). Mothers are offered 4.5 months of maternity leave, and fathers two weeks, but beyond this there is no further economic

compensation available for parents when their child is hospitalized. The FSFB NICU has approximately 600 yearly admissions and is a very modern, large 32 bed unit, with separate rooms for each family separated by transparent glass walls. According to our Colombian team, the families' general feedback is that they like to feel part of what is going on in the unit, and that a need for more privacy is rarely brought up. There are no facilities available for parents to stay overnight, but there are no restrictions to visitation hours either. This unit was the only one of our sites that did not have KC as an explicitly stated part of standard care. The NICU at CM is smaller, with a 19-bed capacity. They have approximately 400 admissions per year. Facilities are modern with individual rooms and a clear focus on family-centered care, including a well-established KC program. There are no restrictions to visitation hours, but the family-rooms are not equipped for parents to stay overnight. Music is a big part of Colombian culture, and many popular musical styles are directly connected to specific dance styles or specific regions of the country. Colombia is culturally diverse with many minority groups that have their own musical traditions and practices, including indigenous populations who use music in ceremonies. Colombians are used to singing, and singing is largely a collective act for social settings such as parties, celebrations, protests, gatherings, and religious settings. Both the Colombian sites had already established MT in the NICU. At FSFB music has been declared an integral part of their humanized health care services through the donations-based program "Hospital Lleno de Música" – "A hospital filled with music".

2.3.3 Israel

Meir Medical Centre (MMC) is a public hospital situated in Kfar Saba, Israel, providing health care to a population from low-to high income backgrounds, with the majority being middle income, working middle class. Approximately 60% of patients are Jewish, and 40% Arab, representing a variety of cultural and religious backgrounds. The unit admits between 350-400 families yearly and is a certified NIDCAP unit with a family-centered-care philosophy and KC program; it had also implemented MT prior to the study. There are no limitations regarding visiting hours,

but facilities for parents to stay overnight are not available. Welfare policies are in place that provide parents with economic compensation while caring for their hospitalized baby. The NICU consists of three open-bay rooms serving different levels of care and is decorated with pastel colored wallpaper, and pictures on the walls that give a more home-like atmosphere.

Everyday practices involving music vary widely within the multi-cultural population of Israel. The secular Israeli population is more accustomed to listening to recorded or live music and less used to singing in their daily lives. The Jewish religious population is more familiar with singing in the synagogues, at the Shabath table, and at other family events. In the Jewish Orthodox population listening to a sole woman's voice is prohibited and therefore these women are not used to singing by themselves. The Muslim religious population is accustomed to collective singing in the Mosque or through individual praying. Very conservative Muslims consider singing as sacred and will therefore only sing as a part of religious acts. However, music is central part of secular Palestinians' everyday life. They have a strong musical culture with a widespread practice of singing as a shared activity related to social gatherings, celebrations, and rituals.

2.3.4 Norway

In Norway participants were recruited from three sites: Akershus University Hospital (AUH), Oslo University Hospital Rikshospitalet (OUH) and Haukeland University Hospital (HUH). All three are public hospitals serving the general Norwegian population from all socio-economic backgrounds. Healthcare in Norway is public and financed by taxation, giving Norwegian citizens' rights by law to equal healthcare. Welfare policies ensure financial support to parents who are caring for their sick child enabling them to be away from work without financial loss. Hospitalized children have a lawful right to always have a parent with them. Family-centered care (FCC) is well-established in Norwegian NICUs, although specific practices and approaches to FCC vary between units. All the Norwegian NICUs in the study had established KC programs, and the NICUs of AUH and OUS were in an early phase of implementing MT prior to joining the LongSTEP trial. HUH have a well-established MT program

in the pediatric ward but had not yet started to provide MT in the NICU before entering LongSTEP.

AUH is a modern hospital in Lørenskog, a suburban area outside of Oslo. The hospital has a multi-cultural uptake area with many families that are first- or second-generation immigrants to Norway. The NICU is designed with a combination of individual family rooms where parents stay with the infant 24/7 and three open-bay rooms where infants are placed in accordance with medical needs; parents can stay overnight. The NICU has approximately 640 admissions yearly, with the majority being premature infants. The family-centered practices of the unit, in addition to KC, include breastfeeding support, NBO and early release with support from the newborn ambulant team.

OUH is situated in Norway's capital Oslo. The NICU provides intensive care to the most critically ill newborns in the South-East of Norway. It is an older unit that is not optimally constructed for family-centered care. It does not have facilities for families to stay overnight inside the unit, but rooms are offered nearby. However, it is a certified "mother-infant-friendly" NICU, offering breastfeeding support and practicing zero separation once the infant is stable enough. It is a 20-bed unit where 60% of the beds are dedicated to intensive care. They have approximately 700 admissions yearly, treating a wide range of conditions including premature infants born as early as 23 weeks GA and critically ill full-term newborns.

HUH is located in Bergen, Norway's second largest city. The NICU is a 16-bed unit that admits approximately 170 premature infants yearly from Bergen and other parts of Western Norway. During the study, a new pediatric department was under construction, and the NICU was situated in a temporary space consisting of both individual family rooms, where parents could stay overnight, and open-bay rooms with incubators. Treatment and care are delivered in accordance with family-centered care and NIDCAP principles.

Norway has a vivid music scene and the highest output of recorded music per capita worldwide (International Federation of the Phonographic Industry, 2018 in Ullsten et al., 2020). However, the general population is more accustomed to music listening than actively engaging with music. Singing in everyday life is mostly associated family life with children, and Norway has a rich tradition and repertoire of folk music, children's songs, and nursery rhymes (Ruud, 2015).

2.3.5 Poland

The Polish site was Szpital Miejski w Rudzie Śląskiej Sp. z o.o. (SRS), situated in Ruda Śląska, an industrial small town near Katowice with a predominantly working-class and middle-income population. Health care in Poland is primarily public, and mothers are provided with paid maternity leave the first twenty weeks following birth, while fathers only have two weeks paid leave that can be used before the child turns two years old. It is common practice for fathers to not take out their portion of leave right after birth, and as a result fathers' presence in the NICU is lower than mothers'. The SRS NICU is a 40 bed-unit, where nine of these are dedicated to intensive care. Family-centered care is not yet well established in Polish NICUs; however, SRS is an exception in that regard. A former head neonatologist was influenced by a Swedish neonatologist⁵ and began establishing family-centered care practices in the SRS NICU. The NICU has no restrictions to visitation hours and offers rooming-in where parents can stay with their infant 24 hours a day. They also have an established KC program. However, the NICU had no prior experience with MT, thus it was introduced to the NICU for the first time through the study. Everyday practices involving music in Poland are mainly centered around music listening. Music education in Poland is predominantly theoretical and does not involve much active playing or singing, which might influence peoples' attitudes to and comfort with singing later in life. Poland is a Catholic country, and some people sing in religious settings, while singing in other settings of everyday life is less common.

⁵ The neonatologist established a collaboration with a NICU in Uppsala, Sweden. Sweden is among the forerunners in neonatal care practice and research and has one of the highest survival rates in the world for premature infants (Ullsten et al., 2020).

2.4 Sample size and power calculations

The LongSTEP trial aimed to recruit a total of 250 participants, with each NICU encouraged to recruit approximately 50 families. With 250 participants the study was intended to have 80% power to detect a difference of 4 points on the PBQ (SD=8) as the minimal clinically important difference. This was based on a two-sided 2.5% significance level (i.e., Bonferroni-corrected 5% for two main comparisons – MT vs. SC during NICU and MT vs. SC after NICU), clustered by country (ICC 0.01), and 20% attrition (Ghetti et al., 2019).

2.5 Trial procedures and intervention

2.5.1 Randomization and blinding

All enrolled participants were randomized to either standard care (control group) or standard care plus MT (intervention group) using a 1:1 randomization stratified by site with random block sizes of two or four. Participants were randomized twice, with the first randomization occurring after informed consent and baseline assessments, and the second randomization when discharged from the NICU. To carry out the randomizations, we used the online system www.sealedenvelope.com, which was administered by a core team member with no involvement in the study's clinical work. In case of multiple pregnancies, only the first-born infant was included and randomized, while remaining siblings received the same interventions for ethical and practical reasons. Due to the nature of the intervention, it was not possible to blind participants, providers, and data collectors. Data analysts were kept blinded to participant allocation until analyses were completed (Ghetti et al., 2019).

2.5.2 Intervention

Music therapy with premature infants and their families is a complex phenomenon that in everyday clinical practice involves a high degree of individualization, flexibility, and improvisation. When designing the intervention for LongSTEP, we aimed to allow for these aspects to still be present through articulating essential elements/principles of the MT approach rather than creating a detailed, rigorous

intervention protocol. Our challenge was to provide the music therapists with specific enough guidelines and training to implement the MT consistently, without compromising necessary adjustments for the therapists' particular cultural context, NICU, and individual needs of the families they would meet. We also wanted to assess treatment fidelity in our trial – specifically on the areas of treatment delivery and receipt across sites to better understand the applicability of our MT approach in different countries, including countries where MT had not yet been systematically implemented in NICU settings.

Music therapy in both intervention phases followed an intervention protocol with guiding principles and essential elements (Ghetti et al., 2019). While staying in the NICU, families were offered three individual MT sessions per week. Before commencing the MT process with a family, the music therapists were recommended to meet with the family twice. First to introduce the concepts and aims of MT, then to begin developing rapport and map out the family's musical preferences and experience, as well as their cultural backgrounds and beliefs. This could take place outside the unit and with parents alone, if suitable. The second meeting was to observe parent-infant interaction in a situation involving handling of the infant, such as diaper changing, bathing, or feeding. This is to form an initial idea about infant maturity, expression, as well as parent strategies to understand and respond to their baby's needs in the moment.

Per protocol treatment was defined as a minimum of six sessions, and families could have a maximum of 27 sessions in total throughout their admission. Depending on the infant's medical condition and state, as well as the NICU facilities in question, sessions were realized either at bedside in an open bay unit or in family-rooms where this was available. Parents and the music therapist would sit next to the incubator or cot, or parents could have skin-to-skin-time or feed the baby during MT. NICU sessions would last approximately 30 minutes, with time spent actively making music dependent upon infant tolerance.

Musical interaction consisted primarily of parent-led, infant-directed singing, supported by the music therapist where the singing was matched to infant state and cues such that the infant “directed” the way the parents and music therapist used their voices. In line with recommendations for MT with premature infants (Nöcker-Ribaupierre, 2013), musical complexity and interaction was progressively expanded upon in alignment with infant GA and readiness in the moment. For infants aged from approximately 26–32 weeks PMA, MT entailed cautious use of parental singing and toned voice using single notes, simple melodies or short musical phrases adapted from children’s songs or parent-preferred music. From 32 weeks PMA and onwards, these elements were expanded upon adding increased musical complexity and interplay. Accompanying instruments were used sparingly to underline the importance of parental voice as an innate resource that is accessible and of unique importance to their baby. Sometimes a guitar or ocean disk was used to support parents’ initial engagement in sessions or facilitate singing when parents felt too insecure to do so without accompaniment. Non-music session time was devoted to therapist-parent dialogue on the family’s needs both before and during the session as needed.

Post-discharge MT sessions were provided monthly over a six-month period in the families’ homes, in connection to follow-up visits at the hospital, or in other health-care settings convenient for the family. During the first month, two sessions were provided to give information, complete assessment, and build rapport as some of the families had their first experiences with MT in this phase. The first session was provided approximately two weeks post-discharge. These sessions had a slightly more set structure, longer periods of active music-making, and increased musical complexity, since infants discharged from hospital are typically developmentally more mature and stable. Sessions lasted approximately 45 minutes to an hour and started with approximately 10 minutes of dialogue on the families’ situation, including changes, development, or challenges since last session, as well as possible approaches to address their current needs and interests in the session. Like in the NICU phase sessions, musical interaction was tailored to infant development over

time. Over the six-month period when sessions were provided, infant and family needs likely changed, and monthly sessions allowed parents to develop skills over time and practice them in their daily lives between sessions. Siblings and other family members who were present could be included in the sessions to make the interactions more relevant to everyday-life family dynamics.

In line with resource-oriented music therapy, the therapeutic encounter across both phases was regarded as a collaboration based on equality, mutuality, and participation between all parts (Rolvsjord, 2016). Aims included the nurturing of parents' and infants' strengths and potentials, and the promotion of dialogical processes between them to support mutual regulation, parent-infant bonding, parent wellbeing and infant development. We also aimed for parents to become comfortable with using musical interaction with their baby by themselves in their everyday lives and contexts, both between sessions during NICU hospitalization and at home.

2.5.3 Standard care

Standard care (SC) varied across sites and included a range of medical, nursing, developmental and supportive care, with the exclusion of other MT approaches. All NICUs defined themselves as family-centered. All except one of the Colombian NICUs (FSFB) included Kangaroo Care as part of SC.

2.6 Ethical considerations, data safety, and user involvement

The study was carried out in line with the ethical principles of the Declaration of Helsinki for medical research involving human subjects (World Medical Association, 2018). Ethics approval for the LongSTEP trial was granted by The Regional Committees for Medical and Health Research Ethics (2018/994/REK Nord, 03 July 2018, see Appendix 3). Each site also obtained ethics approvals in accordance with local and national procedures for clinical research. Informed consent from participants was obtained postnatally following routine practices in each NICU. Local staff involved in the LongSTEP trial provided a written and oral explanation of the

project's aims, the duration of involvement, expected benefits to participants and others, the nature of the interventions, procedures involved in participation, and any potential risks. It was emphasized that enrolment in the study was voluntary: they could withdraw at any time from all or part of the study and any decision they took in this respect would have no bearing on the care the infant and family received. Participants were informed that the outcomes of the study were to be published but no details would be divulged from which the participant could be identified (See Appendix 4 for participant information and consent form).

Data were primarily collected through paper questionnaires that were later entered by local site investigators, or a LongSTEP core team member, into "OpenClinica" – a platform for electronic data capture and clinical data management. Norwegian research partners also used the survey platform Qualtrics to collect data from parents. To maintain participant confidentiality, all information was to be stored with ID code numbers generated upon data entry in OpenClinica. All data was double entered with a required waiting period of 24 hours before second data entry. When each timepoint was completed, data exports were done by the core team statistician and uploaded for storage in the University of Bergen's system for information security SAFE (secure access to research data and e-infrastructure). All analyses were carried out within the SAFE remote desktop solution, which disables downloads of datasets to your own computer. An independent data safety monitoring committee monitored data safety and trial procedures throughout the trial, holding meetings every six months. A user advisory group with parents of premature infants provided consultation in the planning of the study, before and during the intervention period, and in the development of the treatment fidelity questionnaire for parents.

2.7 Design of the substudies of the thesis

The three substudies constituting the thesis are closely related through their focus on different aspects of the MT intervention and resulted in three articles. The specifics unique to each substudy are described in the following.

2.7.1 Substudy I

This substudy was designed as a theoretical study with the dual purpose of elaborating on the theoretical background of the LongSTEP trial's intervention, which was briefly described in the study protocol paper (Ghetti et al., 2019), and presenting the guiding principles, essential elements and proposed mechanisms of action concerning the MT approach in the form of an intervention protocol. The resulting article is a theoretical article, synthesizing and contextualizing the concepts and theories that inform the MT intervention developed for the LongSTEP trial.

2.7.2 Substudy II

Design

The part of the LongSTEP trial investigated within the scope of this thesis was designed as a two-arm parallel, multinational, pragmatic, randomized controlled trial. The substudy addresses one of LongSTEP's secondary aims: the evaluation of short-term effects of MT on the mother-infant bonding and parent mental health at the timepoint of discharge from NICU. Participants in this substudy were families randomized to standard care or standard care plus MT during NICU hospitalization.

Outcome measures

The primary outcome was mother-infant bonding measured by the Postpartum Bonding Questionnaire (PBQ) (Brockington et al., 2006; Brockington et al., 2001) at the timepoints of baseline and discharge. The PBQ is a 25-item self-report screening instrument consisting of statements concerning mothers' feelings, experiences, and attitudes towards their infant. It has four factors with a different number of items rated on a 6-point Likert-scale with responses ranging from 'always' to 'never'. Factor 1 – general impaired bonding – has 12 items; factor 2 – rejection and pathological anger – has 7 items; factor 3 – anxiety about the infant – has 4 items; and factor 4 – incipient abuse – has 2 items. The total score ranges from 0–125, with higher scores implying impaired bonding; additionally, each factor has its own suggested cut-off point. In this study the PBQ total score was the main outcome, with factor 1 – general impaired bonding included as an outcome in the exploratory

analysis. A PBQ total score of ≥ 26 points indicates impaired bonding, and ≥ 40 points indicates a severe bonding disorder. For factor 1, a score of ≥ 12 points indicates impaired bonding (Brockington et al., 2006).

Secondary outcomes included parent anxiety and maternal depression. Anxiety was measured for both parents using the Generalized Anxiety Disorder self-report tool (GAD-7) (Spitzer et al., 2006). The GAD-7 total score ranges from 0–21, with the score of 5 indicating mild anxiety, 10 moderate anxiety, and 15 severe anxiety (Johnson et al., 2019). Maternal depression was measured by the Edinburgh Postnatal Depression Scale (EPDS), a self-report tool with total score range from 0–30 where >10 points indicate depression (McBride et al., 2014).

Baseline and discharge assessments also included parent demographics and infant medical factors, including measures of common health issues for preterm infants such as systemic infections, bronchopulmonary dysplasia, neonatal necrotizing enterocolitis, retinopathy of prematurity, intraventricular hemorrhage, hydrocephalus, and periventricular leukomalacia. Baseline assessments were done before randomization, and discharge assessments were completed 1–2 days prior to discharge, with assessments completed up to 2 weeks after discharge considered as valid.

Statistical analysis

A detailed plan of analysis was made prior to data export. In line with a pragmatic approach, an intention-to-treat (ITT) approach was applied, using all available data from all 213 participants as randomized, regardless of the intervention received. Special approaches for handling missing data were deemed unnecessary because the rate of missingness was low ($<10\%$). Descriptive methods were used to characterize the sample, including means (SD), medians (SE) and percentages. The effects of the intervention were examined by testing for group differences in PBQ total score at discharge with linear mixed-effects models (ANCOVA) adjusted for site due to stratified randomization. The same analyses were carried out with secondary outcomes. The quality of the models was evaluated by residual plots. Exploratory

analyses of PBQ total score were conducted for pre-specified subgroups including sex; GA at birth (<28 weeks, 28 to <32 weeks, 32 to <35 weeks]); hearing status measured at discharge (normal vs. abnormal); PBQ factor 1 impaired bonding score (≥ 12 vs. < 12); parental socioeconomic status (education level); average parental skin-to-skin care during NICU (4 or more days per week vs. fewer than 4 days per week); and treatment per protocol (≥ 6 MT sessions) (Ghetti et al., 2019). Statistical analyses were carried out using statistics software R version 4.1.0 (R Project, 2022) with a two-sided 5% significance level.

2.7.3 Substudy III

Participants and raters

Participants in this substudy were drawn from the pool of participants who were allocated to MT in NICU or/and post-discharge phases. Evaluation of the treatment-fidelity involved two rater groups: music therapist self-raters, which consisted of the music therapists who carried out the MT in the LongSTEP trial; and external raters who were persons affiliated with the respective sites that had an education in MT or psychology and understood the language used in the sessions. The treatment fidelity (TF) of two domains was evaluated:

- Music therapist *treatment delivery* (TD), which concerns therapist adherence to essential elements of the intervention protocol.
- Parent *treatment receipt* (TR), which concerns whether parents were able to perceive essential elements of the intervention (Bellg et al., 2004).

Outcome measures and data

Development of treatment fidelity questionnaires

Assessment of TF within MT trials has not been common practice, but there are examples of trials reporting separate TF analyses, or addressing aspects of fidelity in research protocols, intervention guidelines, or when reporting RCT results (Baker et al., 2019; Erkkilä et al., 2008; Erkkilä et al., 2011; Geretsegger et al., 2015; Robb et al., 2011). However, to our knowledge no other trials in the field of MT with

premature infants and their parents have assessed TF, and we did not find any existing TF assessment tools that fit our intervention. Therefore, we had to develop assessment tools ourselves, which is not itself unusual for complex interventions such as MT that often entail highly individualized interactions between a therapist and clients (Skivington et al., 2021). In some cases, it might be sufficient to make adaptations to existing tools while in others they must be developed from the ground up to fit the intervention in question. When developing TF assessment tools, we followed a process similar to what Feely et al. (2018) describe in their five-step field guide to fidelity measurement. They suggest four key measurement-related criteria to consider when developing a new measure or modifying existing measures: (1) the organization of the fidelity measurement tool; (2) items to be included on the list; (3) phrasing of the items; and (4) response choices (Feely et al., 2018 p. 143). When organizing the tool, the structure of the intervention and delivery should inform the design of it. In LongSTEP, we articulated a set of seven⁶ essential and distinguishable elements when developing the intervention protocol, which should be present in all sessions regardless of which phase MT was delivered in or the infant's PMA and state. These elements formed the basis of the TF tools. We had decided to measure both treatment delivery in the form of therapist adherence and treatment receipt (Bellg et al., 2004). This meant that TF assessment would include both music therapist self-ratings, external ratings, and parent ratings, each of which would have different perspectives on, and levels of insight into, the MT process. This, in turn, meant that we had to develop different variants of the TF tools. Items were re-phrased versions of the intervention's essential elements into statements. For response options, we chose Likert-scaled items (each from 0-6), with anchors "I completely disagree" and "I completely agree". A threshold of ≥ 4 was decided *a priori* as a satisfactory level of fidelity per item, with higher numbers indicating better therapist adherence and parent perceived receipt of the intervention's essential elements.

⁶ Essential element 1 concerning observation and dialogue on infant needs was split into two items in the treatment fidelity assessment. This first element would normally involve checking in, and if necessary, adjusting infant *physical positioning* to support infant regulation and readiness for interaction, which would not be possible to assess from audio recordings. The video recordings from the post-discharge phase, however, allowed for such observation, and consequently a separate item on infant physical positioning was added.

Four treatment delivery questionnaires were created in accordance with a) the phase within which MT was delivered, b) which elements could feasibly be distinguished in audio versus video recordings of sessions, and c) who was completing the rating. These were as follows: Treatment Delivery Questionnaire for Music Therapist Self-ratings, NICU (seven items) and post-discharge version (eight items); Treatment Delivery Questionnaire for External Raters, NICU (six items); and post-discharge version (seven items). One element was not observable for all raters because of a reliance on familiarity with the family's music preferences and was therefore only included in the music therapist and parent questionnaires, resulting in different numbers of items for each variant of the tool. Another element regarding infant physical positioning was only observable with video recordings and was only included in the post-discharge tools. The fifth questionnaire we developed was the Treatment Receipt Questionnaire (nine items) assessing parents' perception of the intervention. One additional item that was only included in the TR questionnaire concerned parents' experiences of their voices being unique and important in relation to their baby, as it was something neither the music therapists nor external raters could provide feedback on.

Treatment delivery questionnaires were pilot tested by two members of the study core team and deemed functional. The treatment receipt questionnaire was presented to and discussed with the user advisory group who suggested simplifying the wording. Changes were made accordingly before implementation.

Audio and video recordings

The music therapists were responsible for audio recording all MT sessions during NICU and video recording all MT sessions post-discharge for treatment delivery (TD) assessment purposes. Audio recording was chosen for the NICU phase due to practical and ethical considerations. Many NICUs are designed as open-bay units with families residing very close to each other. Video recording in such cases would involve a significant risk of recording persons who were not part of the study and had not given consent. Also, premature infants in the NICU are very small, and it would have required an advanced camera setup to be able to record both the subtle cues

from the infants and the parents interacting with them. Although audio recordings would provide us with less information than videos, it was considered the best solution for recording the NICU phase sessions. Follow-up sessions post-discharge were encouraged to take place in the families' homes, which eliminated the risk of recording persons other than the participating family. Video instructions were to aim for a frame that showed both parent(s) and infant. One of the sites did not allow any form of video recording for the study's purposes and recorded audio instead of video for the post-discharge sessions as well.

Approximately 10% of each therapist's participants were included in the treatment delivery analysis. These participants were randomly selected using www.randomresult.com with the "Pick items" function. If the recorded material from the selected participant was not possible to use due to low quality, missingness or because the participant had dropped out of the study, a new participant was drawn randomly. For the sample our strategy was to evaluate sessions that were distributed over time, avoiding the first and last session because we expected that these sessions might include minimal levels of interaction between parents and infants or singing. The first sessions were used to explain and demonstrate aspects of the intervention, and in the last sessions a significant portion of time would be used to sum up content from the MT process, as well as dialogue about parents' continuation of using music independent of the music therapist. From the NICU phase recordings of session three, five and seven were analyzed for participants who received a minimum of seven sessions. If they had fewer sessions, we analyzed number two, four and six. In cases where participants had more than 10 sessions, the 11th session was added with the intention of evaluating drifting from the intervention protocol; however, we did not have a sufficiently large sample to carry out this analysis ($N = 39$). From the post-discharge phase, recordings of two sessions per participant were evaluated. The sessions evaluated were either number three and six, three and five, or four and six, based on useable video. Some recordings had very poor quality and were impossible to use, and in the final sample the session number varied due to missing or damaged recordings.

Treatment fidelity assessment procedures

For the assessment of treatment delivery, each therapists' sessions were reviewed by the corresponding therapist and two external raters who understood the language spoken and were educated in MT or psychology. The questionnaires included descriptions of behaviors related to each item to look for. Further instructions were to listen to or watch the recorded session once in its entirety, while filling out the questionnaire. Questionnaires were uploaded to SAFE, and a member of the core team entered all data. Parent ratings of treatment receipt were collected at the six-month assessment timepoint. Parents were instructed to think back on their experiences with MT as a whole and evaluate the degree to which they perceived the guiding principles of the MT intervention.

Statistical analysis

A detailed plan of analysis was made prior to data export. Descriptive methods were used for characteristics of the two participant samples for TD and TR. Categorical data were analyzed with frequency and percentage, and numerical data with mean, standard deviation, and range due to normally distributed data. The TF questionnaires' internal consistency was evaluated with Cronbach's alpha (Cronbach, 1951), with Alphas of ≥ 0.70 indicating good internal consistency (Tavakol & Dennick, 2011). Interrater reliability (IRR) between music therapists and external raters was evaluated per item, and the total score was determined with intraclass correlation coefficient (ICC) with a two-way model, single measurement, and absolute agreement. We also calculated the agreement of categorical items dichotomized to above/below the threshold of satisfactory adherence (≥ 4). Because of the high prevalence of single outcomes for some of the items, we used Gwet's AC (Gwet, 2008) instead of kappa, due to the known weaknesses of kappa in this case (Viera & Garrett, 2005). Mean TD scores per item, therapist, and composite total score (mean scores across external ratings and music therapist self-ratings) were calculated from ratings from the two intervention phases (NICU and post-discharge). Mean TR scores per item, total score per participant, and composite total score across intervention conditions (MT in NICU, post-discharge or both) were calculated from

parent ratings. Statistical analyses were done with software R version 4.1.0 (R project, 2022). Graphics were made with Matlab (MathWorks, 2021).

3. Summary of results

3.1 Demographics and clinical characteristics of LongSTEP participants

We recruited 213 families to the LongSTEP trial. Half of the enrolled infants were girls (51%), and the average GA at birth was 30 weeks. Post-menstrual age at enrollment was at average 33 weeks. Average birth weight was 1414 grams, and average weight at enrollment was 1618 grams (See Table 1 in Gaden et al. 2022). Mothers' and fathers' average age was 33 and 36 years, respectively. Mothers had an average of 16 years education, ranging from four to 22 years. Fathers' average education level was 15 years, ranging from three to 25. In terms of usual work situation, 72% of mothers and 93% of fathers were full-time or self-employed. Of the mothers, 92% were married or living together with their partner. Average baseline PBQ total score was 7.4 ranging from 0 to 62 points. Nine of the 209 mothers who completed assessments (4%) scored above the threshold on factor 1 (≥ 12) for indications of impaired bonding⁷ (See Table 2 in Gaden et al. 2022, and Appendix 5).

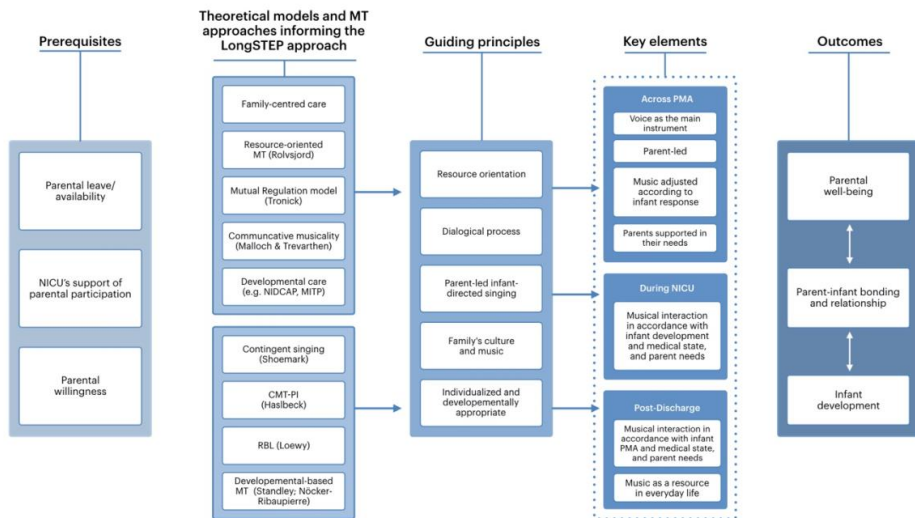
3.2 Article I: The LongSTEP approach – Theoretical framework and intervention protocol for using parent-driven infant-directed singing as resource-oriented music therapy

The main aim of article I was to thoroughly describe the theoretical models, concepts, and approaches informing the MT approach developed for LongSTEP and to demonstrate how these are manifested in the MT sessions through the guiding principles and essential elements of the intervention. In the attempt to fulfil these two aims, article I served multiple purposes, amongst them providing an intervention protocol to make the study replicable, and operationalizing essential elements into

⁷ In the published article (Gaden et al., 2022) there is an error in Table 2 concerning the PBQ factor 1 scores. The correct values for "Above factor 1 threshold (≥ 12) for impaired bonding" are 9/209 (4%) of the total participants, with 5/104 (5%) in the MT group and 4/105 (4%) in the SC group. See Appendix 5 for corrected table.

observable behaviors for later treatment fidelity analysis purposes. Two models resulted from the research process. The first (Figure 3) is a conceptual model that provides an overview over the relation between the MT approach's prerequisites; informing theoretical models, concepts, and approaches; proposed guiding principles and key elements; and intended outcomes. Resource-orientation, with MT as a dialogical, collaborative process that is tailored to infant developmental needs and families' cultural and musical background, with an emphasis on parent voice as the primary instrument, are key features of the LongSTEP approach. The approach is feasible to implement in culturally-diverse settings where parent presence in NICU is high but tailoring to local contexts and circumstances is required. It also requires sufficient psychosocial and musical support for parents since the approach places a high demand on parents with its emphasis on parent voice and active participation.

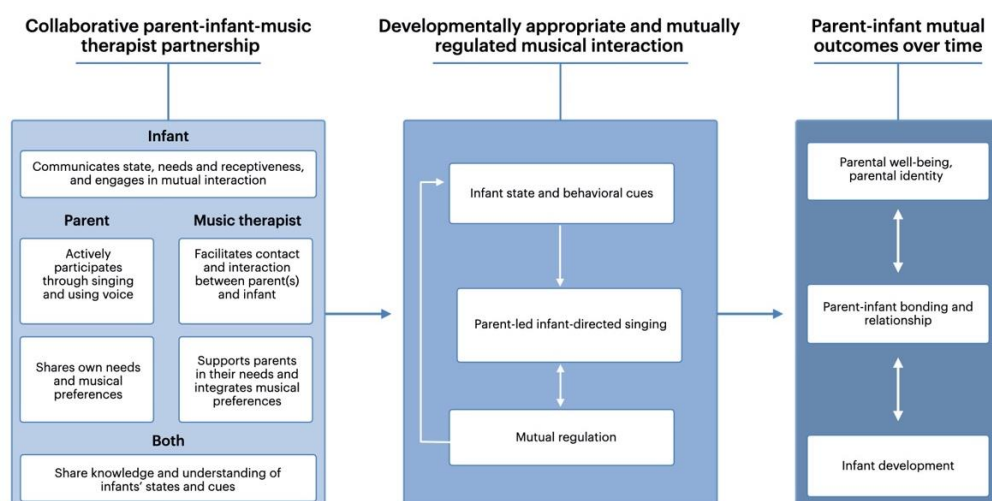
Figure 3 Conceptual model of the theoretical framework and key elements of the LongSTEP approach. Reprinted from Gaden et al., 2021



Note: See Appendix 6 for a larger scaled version of the figure.

The second model illustrates the proposed mechanisms of the MT approach. In psychotherapy research the term “mechanism” explains how the intervention translates into events that lead to the outcome and reflect the steps or processes through which therapy actually unfolds and produces change (Kazdin, 2007). In the second model (Figure 4) we suggest two main mechanisms that we hypothesize might produce change in parent-infant mutual outcomes over time: 1) the collaborative parent-infant-music therapist partnership and 2) developmentally appropriate and mutually regulated musical interaction.

*Figure 4 Proposed mechanisms of action of the LongSTEP approach.
Reprinted from Gaden et al., 2021*

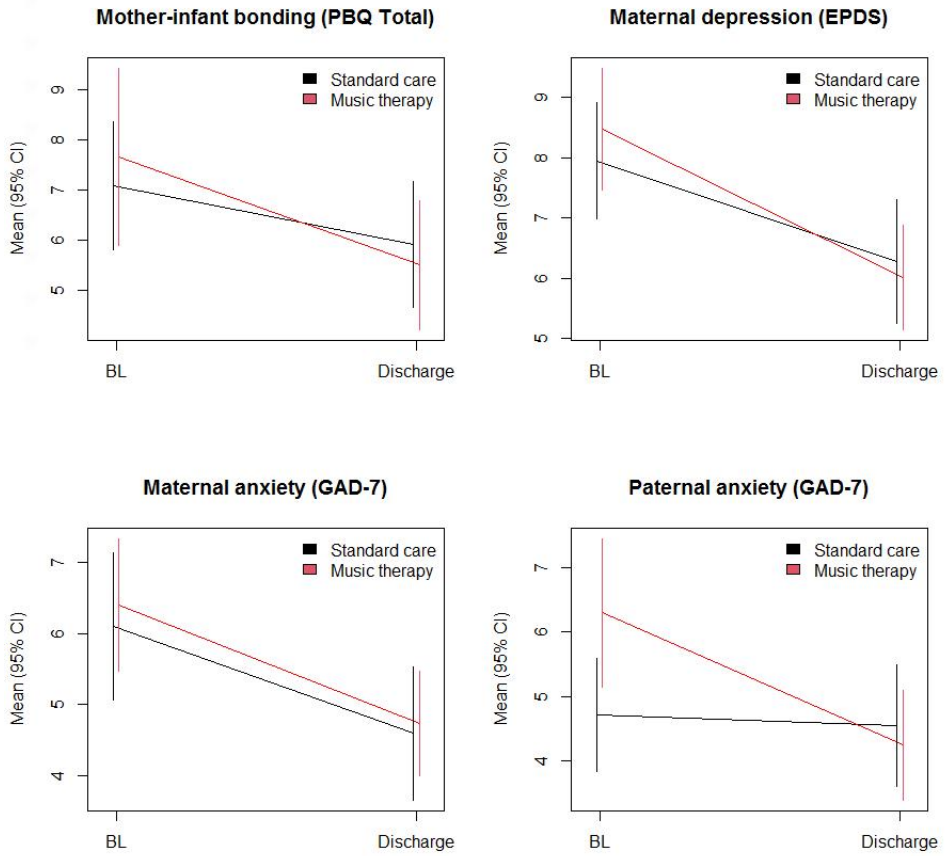


Note: See Appendix 7 for a larger scaled version of the figure.

3.3 Article II: Short-term music therapy for families with preterm Infants – A randomized trial

In article II we measured the short-term effects of the MT intervention during NICU hospitalization on mother-infant bonding measured by the PBQ in a pragmatic randomized controlled trial. Secondary outcomes included parent anxiety measured by the GAD-7 and maternal depression measured by the EPDS. We found that the PBQ mean total score (SD) decreased from 7.66 (9.05) at baseline to 5.51 (6.65) at discharge in the MT group, and from 7.08 (6.55) at baseline to 5.92 (6.38) at discharge in the SC group (Figure 5). However, no significant difference between groups was detected. The estimated group effect (95% CI) for the PBQ was -0.61 (-1.82 to 0.59). The narrow CI suggests that we can exclude both beneficial and harmful effects. The same applied to secondary outcomes and predefined subgroups. Estimated group effects (95% CI) for anxiety measured by GAD-7 were 0.14 (-0.85 to 1.12) for mothers, -0.90 (-2.03 to 0.22) for fathers, and -0.60 (-1.72 to 0.52) for maternal depression measured by EPDS (Figure 5). Uptake of the intervention was high with 87 of 105 (83%) participants receiving per protocol MT (≥ 6). The average (M) number of MT sessions was 10. Of the 213 families enrolled in the study 208 (98%) completed assessment at discharge. There were no adverse events related to the intervention. The very high levels of treatment uptake and study retention, together with the lack of adverse events, suggest that the MT approach of LongSTEP is feasible and safe, although with no short-term effect on parent-infant bonding and parental mental health as measured in the study.

Figure 5 Changes from baseline to discharge. Reprinted from Gaden et al. 2021.



3.4 Article III: Treatment fidelity in a pragmatic clinical trial of music therapy for premature infants and their parents – The LongSTEP trial

In article III we investigated treatment fidelity (TF) in both intervention phases of the LongSTEP trial. We developed four versions of TF assessment questionnaires for music therapist self-rating and external rating of treatment delivery (TD) based on audio and video recorded MT sessions from NICU and post-discharge phases. We developed one parent questionnaire rating treatment receipt (TR) across both MT phases. For TD analysis, a total of 72 NICU and 40 post-discharge sessions of 39 unique participants were rated by 10 music therapists and 13 external raters. Treatment receipt was evaluated by 135 parents. Results from reliability analyses showed that four of the five TF questionnaires had good internal consistency measured by Cronbach's alpha (≥ 0.70). The exception was the NICU external questionnaire that scored slightly lower (α (95% CI) 0.66 (0.60, 0.73). Interrater-reliability of TD composite scores across music therapist self-rater and external rater questionnaires tested by intraclass correlation was moderate 0.43 (95% CI 0.27, 0.58). Dichotomized items (above/below threshold of satisfactory adherence ≥ 4) tested with Gwet's AC varied between 0.32 (95% CI 0.10, 0.54) and 0.72 (95% CI 0.55, 0.89). Average TD scores were satisfactory with a mean (SD) composite score across raters for NICU sessions of 4.88 (0.92), and 4.95 (1.05) for post-discharge sessions, meaning they scored between one and two Likert points away from "I completely agree" (Figure 6). Parents' TR scores were very high. The NICU group had a mean (SD) score of 5.66 (0.50), the post-discharge group's mean (SD) score was 5.65 (0.71), and the group that received MT in both phases had a mean (SD) score of 5.71 (0.40). The mean (SD) TR composite score across the three groups was 5.68 (0.53) (Figure 7). In conclusion, TF analyses indicate that music therapists adhered to essential elements of the intervention protocol to a satisfactory degree, and that MT was a uniform intervention during both NICU and post-discharge phases, as well as across the different cultural contexts of the trial. Parents also received the essential elements of the intervention, with parents who had MT in both phases having highest TR composite score. However, the differences between them and those who only had MT in one phase were small.

Figure 6 Treatment delivery average score per item and phase

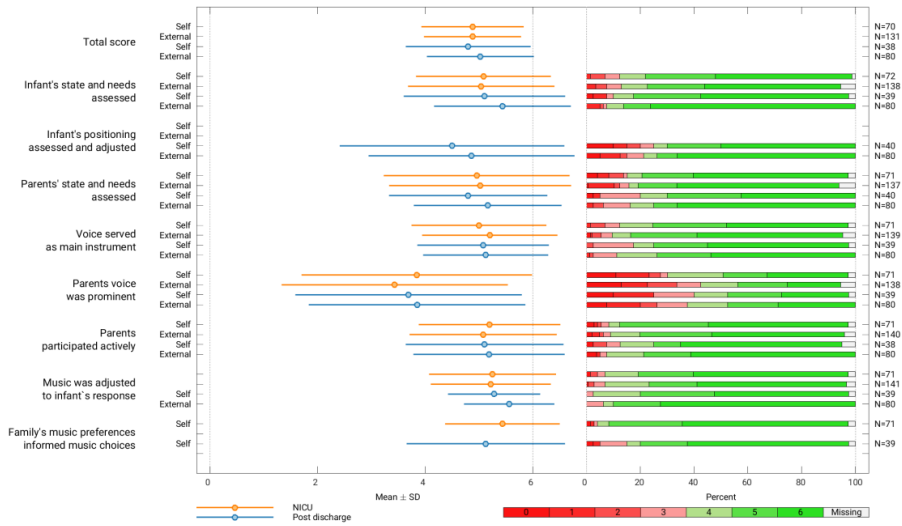
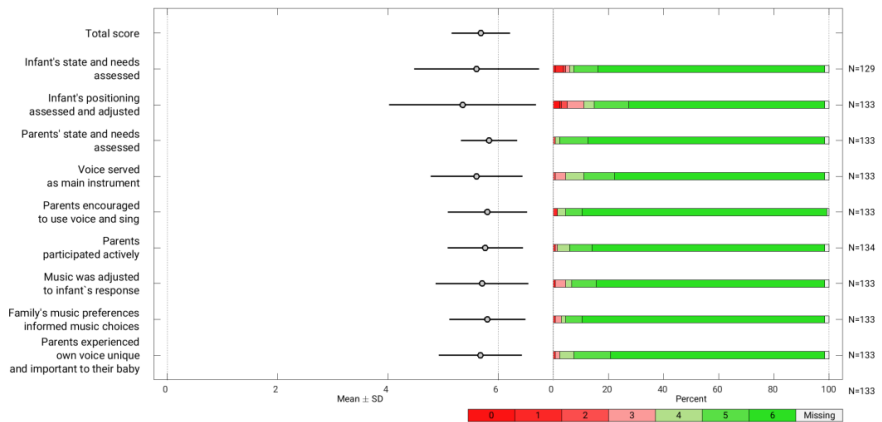


Figure 7 Treatment receipt average score per item



3.5 Additional exploratory analysis beyond the articles

3.5.1 Parent self-reported use of music

Because the treatment fidelity analysis did not include assessment of treatment enactment - the degree to which participants perform intervention-related strategies and skills between sessions/after the intervention period (Bellg et al., 2004), an exploratory analysis of a related variable from the dataset concerning parents' own use of music was conducted. At the discharge assessment parents were asked how often they sang or played music with their baby during hospitalization. In the MT group 70% (74/105) report doing so daily, whereas 39% (39/100) in the SC group did so. At the 6-month assessment they were asked how often they sang or played music or attended music groups with their baby. The results shows that daily music use post-discharge was 77% (73/95) and 67% (60/90) in MT and SC groups, respectively.

4. Discussion

The data for the three articles that make up this thesis were collected in the LongSTEP trial, enrolling 213 families with premature infants from sites in Argentina, Colombia, Israel, Norway, and Poland. We found the MT approach developed for LongSTEP to be relevant across a wide range of cultural contexts and settings where parental presence in the NICU is high. The intervention protocol's guidelines and essential elements were to a large degree feasible for music therapists to understand and follow. Its resource-oriented focus seems to be compatible with variations of family-centered standard care during NICU hospitalization across the included countries. Despite these findings, the LongSTEP RCT investigating the short-term effects of MT during NICU found no differences between SC and MT groups on the primary outcome of mother-infant bonding, nor on the secondary outcomes of parent anxiety and maternal depression. Surprisingly, the parents in our study did not show the expected levels of impaired mother-infant bonding or symptoms of anxiety and depression at baseline or discharge. In conclusion, the MT approach in our study did not lead to differences in the outcomes as far as we could detect with the chosen outcome measurement tools.

At first glance, results like these might appear disappointing. However, our findings provide opportunities for learning and guide us towards the next steps in our research on the subject. The narrow confidence intervals for the estimates of effect suggest that we can exclude both beneficial and harmful effects. Music therapy was as effective as SC in reducing impaired mother-infant bonding, symptoms of maternal depression and parent anxiety. Our results also indicate that parents in our study seem to be doing better in terms of mental health challenges following preterm birth, compared to what is reported in some of the literature, which is an inherently positive finding. To explore possible explanations for the observed lack of treatment effect, I will critically discuss the choice of primary outcome, contextual factors of the study and features of the intervention itself.

4.1 Bonding as primary outcome

In our study, a very low number (9/209, 4%) of mothers from a range of countries and socioeconomic statuses reported impaired bonding. Our findings clearly challenge the assumption of a higher risk of impaired bonding following preterm birth. Bonding was chosen as the primary outcome in LongSTEP in response to an identified gap in the literature where large-scale, rigorous studies on MTs potential benefit on long-term and parent-infant mutual outcomes were missing (Bieleninik et al., 2016; Yue et al., 2020). Given that the early parent-infant relationship is widely recognized as having long-term implications for infant development (Bowlby, 2008; Le Bas et al., 2019; Winston & Chicot, 2016), and that preterm birth might impact parent-infant bonding negatively (Kim et al., 2020; Muller-Nix et al., 2004; Trumello et al., 2018; Winter et al., 2018), bonding was also considered to be a clinically meaningful outcome variable for the LongSTEP-study.

4.1.1 Challenges in measuring bonding

The nature of bonding is assumedly a mix of biological, psychological, and sociocultural mechanisms influenced by the immediate context, the respective characteristics of the parents and baby, and their history and present situation (Cataudella et al., 2016; Kinsey & Hupcey, 2013). When bonding occurs in the context of prematurity, threats to infant survival and long-term health, parents in crisis, and the stressful environment of the NICU make bonding even more complicated (Cheong et al., 2020; Kim & Kim, 2022). In order to evaluate the effect of a particular intervention on bonding, standardized assessment tools are necessary. Such tools necessitate the operationalization of the concept of study and psychometric validation, which inevitably reduces the complexity of bonding.

In contrast to the assessment of infant-parent attachment, there is no gold standard assessment for parent-infant bonding. Various observational and self-report measures exist with different theoretical underpinnings, content, format, and purposes (Wittkowski, 2020). Observational measures are time- and resource intensive, and often require extensive training to administer and interpret. In the planning of

LongSTEP, observational tools like the Emotional Availability Scales (Biringen et al., 2008) were discussed in relation to the feasibility of their use and psychometric properties. Ultimately, observational tools were ruled out due the lack of a clearly psychometrically superior tool that would be feasible to use in NICU settings of multiple countries (Ghetti et al., 2021; Lotzin et al., 2015).

4.1.2 Appropriateness of the Postpartum Bonding Questionnaire

Self-report tools involve the risk of social desirability bias, which in turn can skew interpretation, but their advantage is that they are less costly and labor-intensive to implement (Wittkowski, 2020). In the case of bonding, self-report tools are also clinically meaningful since a complete understanding of bonding must include the parent's subjective experience of their relationship with the child (Wittkowski, 2020). After pilot testing several self-report tools, the PBQ was chosen as the primary outcome measure for LongSTEP. The PBQ is a screening tool designed to identify disorders in mother-infant bonding, but it can also be used to assess change over time (Brockington et al. 2001, 2006). The PBQ has received the strongest psychometric evaluation ratings with a high quality of evidence in a systematic review comparing parent-reported assessment measures for bonding or attachment (Wittkowski, 2020). In addition to scoring high on validity and reliability, the PBQ is free of charge and available in multiple languages. The self-report administration is quick for parents to complete. In sum, the PBQ offered many advantages for a large-scale multinational study with several assessment timepoints, like LongSTEP. To our knowledge, the PBQ has not yet been used to assess the effect of MT on bonding in other trials besides LongSTEP.

The PBQ is targeted to both healthy women and women with post-natal depression and other postpartum disorders (Brockington 2001, 2006). However, our results suggest that the PBQ may be less sensitive to detect changes in bonding in mothers of premature infants in countries with a high degree of parent participation in the NICU. In our study 96% (200/209) of the mothers scored below the cut-off (≥ 12) for indications of problematic bonding. It is possible that the strong focus on pathology

of the PBQ created a floor effect⁸ making it challenging to detect differences between mothers in the more subtle and health-oriented aspects of bonding. Another possible explanation of low PBQ scores at baseline is that mothers may experience difficulties in forming emotional bonds with their child without being willing to disclose it. The wording of some items in the PBQ can be provocative (e.g., “I have done harmful things to my baby”, “I resent my baby”). It is likely that getting parents to disclose such feelings or admit struggling with their relationship to their baby is difficult. Parents might fear stigmatization, the involvement of social services, or a potential loss of custody (Wittkowski et al., 2019). Other PBQ-items seem less relevant for the early postpartum period, particularly in the case of premature or sick infants that do not display the same behavior or tolerate the same interaction as full-term infants (e.g., “I enjoy playing with my baby”, “I feel happy when my baby smiles or laughs”). The range of languages and cultural backgrounds of mothers may also have affected how well the wording of some of the PBQ items were understood (e.g., “My baby winds me up”). While the conclusion of feasibility studies informing LongSTEP was that the PBQ appeared to be a suitable measure for impaired mother-infant bonding, parents did report some PBQ items being more provocative than the items of other tools they piloted (Ghetti et al., 2021).

The PBQ’s emphasis on pathology, measuring the degree to which bonding is impaired, stands in contrast to the MT approach in LongSTEP, which is resource-oriented and focuses on building up parents’ and infants’ strengths and innate capacities. It seems like the PBQ may not have been the most suitable outcome measure to assess changes in mother-infant bonding in the participants of our trial.

Other studies using the PBQ have also failed to confirm problematic bonding following preterm birth (Bieleninik et al., 2021). Hoffenkamp et al. (2012) found parents of preterm infants even reporting better bonding than parents of full-term infants. When comparing the average baseline PBQ total score of mothers in our

⁸ A floor effect in evaluation refers to when a large proportion of participants score very low, skewing the distribution of the scores and making it impossible to differentiate between the many individuals at the low score level (American Psychological Association Dictionary of Psychology, n.d.).

study (Mean 7.37) to that of normal mothers in Brockington (2001)⁹ (Mean 12.3), we have similar findings. The average score of mothers in our study are notably lower, suggesting better bonding. Hoffenkamp et al. (2012) hypothesize that parent-infant bonding following preterm birth might be better in affluent countries with adequate resources, while Borghini et al. (2006) suggest that parental emotional arousal during hospitalization may facilitate parental involvement, which in turn might strengthen the parent-infant-bond.

Although we did not identify significant changes in mother-infant bonding with the PBQ, this does not exclude the possibility that the LongSTEP approach to MT may impact bonding following preterm birth in ways not captured by the tool. In a qualitative interview study, parents of an Israeli cohort of LongSTEP who participated in MT in the post-discharge phase experienced that MT offered a means of expanding their relationship with their baby. Music therapy increased parents' sense of agency through building communication skills and discovering their own voice as a resource (Epstein et al., 2022). These findings are in line with previous qualitative and mixed-methods studies where parents describe an enhanced sensitivity to their baby's cues, feelings of relationship, bonding, and closeness a result of MT (Kehl et al., 2021; Kostilainen et al., 2020; McLean et al., 2019).

4.1.3 Impact of study settings

The LongSTEP MT approach was developed for use in countries where health care and social welfare policies such as parental leave schemes and economic compensation while caring for a sick child enable parents to be consistently present in the NICU. The LongSTEP NICUs were all family-centered care units without restrictions to visitation hours. Across all our sites, a very high number of the parents in both the SC (96%) and MT (92%) groups reported being present in the NICU daily. In such settings, longer periods of unwanted separation between parent and infant are prevented, which may reduce the separation-induced stress noted in some parents of premature infants (Bidzan et al., 2009). Additionally, all but one NICU

⁹ Average total score was not reported in Brockington (2001), but when adding the mean scores of each subscale reported in their Table 2 average total score of normal mothers was 12.3.

(FSFB in Colombia) had established practices to promote parent-infant bonding through KC programs. In several of the NICUs additional approaches and practices emphasizing parents' participation and unique contributions were also part of standard care (e.g., NIDCAP, NBO, breastfeeding support, 24-hour access or rooming-in with the baby). In sum, the NICUs in LongSTEP seem to have had good conditions for parent-infant bonding in place without MT, which may partially explain the low occurrence of problematic bonding in our participants. The sites have high parental presence in the NICU and seemingly good conditions for parent-infant bonding in common. However, as the descriptions of the countries and sites (pp. 26-31) illustrate, each NICU and their respective countries have unique cultural and contextual qualities, which add variance and heterogeneity to our sample and settings. Treatment fidelity assessment was hence particularly important.

4.2 Fidelity of the music therapy intervention

In line with a pragmatic approach, the MT in LongSTEP was delivered in everyday practice clinical settings as part of usual care. The MT in LongSTEP was a complex intervention (Skivington et al., 2021) as it included several different components and interactions among them (e.g., live music adapted to infants' state and needs in the moment); it also required certain expertise and skills from the music therapist (e.g., knowledge on premature infants' behavioral cues and states, musical and therapeutic skills and experience) and tailoring of the intervention (e.g., to cultural context and individual needs). The complexity of the intervention, and the aforementioned differences between countries and sites, made the systematic assessment of treatment fidelity particularly important.

Average treatment delivery (TD) scores show that music therapists across sites adhered to the elements of the intervention protocol to a satisfactory degree. Treatment receipt (TR) scores suggest that parents received the intervention as intended. Altogether, this suggests that MT was implemented in a uniform way across the eight different sites in Argentina, Colombia, Israel, Poland, and Norway. Furthermore, these findings indicate that the LongSTEP MT approach is suitable for

various cultural settings where parental presence during NICU is high, and where possibilities for providing follow-up to families after discharge from hospital exist. The high average TF scores may suggest that the non-significant results from the RCT are not related to an inconsistent implementation of the MT intervention. It is possible that a per-protocol analysis with only those with satisfactory TF scores (≥ 4 was considered satisfactory) had shown better effects. However, such an analysis was not included in the statistical analysis plan nor conducted as part of this thesis.

4.2.1 Parent use of voice and singing

Treatment fidelity findings suggest that one element of the LongSTEP approach – parents' active participation through singing, is more challenging to implement than the others. Both external raters and music therapists (self-rating) scored the TD item concerning the prominence of parental voice below the set threshold for satisfactory adherence (≥ 4 with full range of scale 0–6). The item asking if parents participated actively, however, had a satisfactory score. Parent TR scores confirm that they were encouraged by the music therapists to actively participate with singing, humming, touching, and moving their infant. However, the TR questionnaire did not include an item that specifically asked whether parents actually engaged in singing when offered opportunities. An integration of these results suggests that parents did participate actively, even if they did not sing to the extent expected by music therapists and external raters.

It is reasonable to believe that individual parents had varying expectations of what active participation and singing entailed. While some parents might have sung in MT without requiring encouragement to do so, others would not have sung without encouragement. Some parents may have perceived their own vocal engagement and participation as substantial even if they did not take on a lead role in singing, but just shared their music preferences, chose songs, hummed, or sang together with the music therapists. The TF parent questionnaire included an item asking to what degree the parents experienced that they had something unique and important to offer their baby through their voice. The average score of this item was very high, suggesting

that parents who had MT did indeed experience their voice as being important for their baby.

The feasibility studies informing LongSTEP and feedback from parents' experiences from MT post-discharge indicate that singing can be challenging, uncomfortable, and embarrassing for parents (Bieleninik et al., 2020; Epstein et al., 2022; Ghetti et al., 2021). However, parents' level of comfort with singing seems to change over time as they establish a relationship with the music therapist, get more practice and understanding of the potential of their voice, and, most importantly, after parents experience positive responses to their singing from their baby (Bieleninik et al., 2020; Epstein et al., 2022). In LongSTEP, participants had an average of ten MT sessions during NICU, and were hospitalized on average seven weeks, which might be too short a time and too few sessions for parents to get ready to take on a leading role in singing during MT sessions. However, an exploratory analysis of the data on parent self-reported music-use shows that 70% (74/105) of parents in the MT group sang or played music for their baby in the NICU daily, while 39% (39/100) of the SC group parents did the same. While parent-led singing in front of the music therapist or others can be challenging, participation in MT might have motivated parents to explore singing with their baby on their own during NICU admission.

In LongSTEP, the emphasis on parents' voices and singing is strongly related to the resource-oriented perspective informing the study (Rolvjord, 2010). Previous studies and approaches also highlight how parents' voices are the most relevant to the infant (e.g., Loewy et al. 2013; Loewy, 2015) – their “favorite music” so to speak (Shoemark & Nöcker-Ribaupierre, 2021). Parental singing can also promote intimacy and feelings of closeness, as well as support parental identity (Haslbeck, 2014; Kehl et al., 2021; McLean & McFerran, 2016; McLean et al., 2019). However, many previous studies do not report to which degree parents participated with singing compared to how often singing was primarily done by the music therapist. In LongSTEP, we proposed parent-led infant-directed singing to make parents' voices become more audible than the unfamiliar voice of the music therapist. The music therapists purposely created space for parents' voices through minimizing the use of

accompanying instruments (e.g., guitar). While accompaniment can give an added value musically and reduce the feeling of being exposed when singing alone, it may also be a barrier for parents in gaining confidence in singing (Epstein et al., 2022). In a qualitative study with Israeli LongSTEP participants a father describes how when they had to sing without accompaniment, it exposed his and his wife's ability to sing to themselves (Epstein et al., 2022). In line with the resource-oriented perspective informing LongSTEP, we intentionally challenge the dynamics of the therapeutic relationship, positioning parents as experts on what their infant communicates and needs. The parental voice is seen as a unique resource in the process of bonding with their child. The music therapist's expertise in this setting is to facilitate, support and convey knowledge in a non-directive way.

In spite of good intentions to provide space for parents' voices, it is possible that the LongSTEP approach demands too much from some parents. While no variable in our dataset captured these properties, they may be relevant to consider in future trials.

Other studies have pointed out how singing may at times be inaccessible for parents during hospitalization (McLean, 2016; Shoemark & Dearn, 2008). For example, if the baby is in a critical state or if the parents themselves feel overwhelmed by other tasks and responsibilities or their own emotions from experiencing preterm birth. In such cases, MT approaches with lower or other demands for parent participation should be considered, for example, MT approaches with a stronger emphasis on parent psychotherapeutic and trauma-informed support (Stewart, 2009a, 2009b; Loewy et al., 2021), where sessions are provided to parents alone or live music is provided in a more receptive format through music relaxation with monochord (Haslbeck & Bassler, 2020), entrainment to breathing with ocean disk (Loewy et al. 2013), or environmental MT (Rossetti, 2020).

4.2.2 Intervention implementation during the pandemic

A last aspect that affected implementation of the intervention was the Covid-19 pandemic. Recruitment of participants began in August 2018 and concluded in April 2020. When the pandemic had reached all our collaborating countries, we were in the finishing stages of data collection for the NICU phase and only a few families had

MT sessions left. For the post-discharge phase, however, several families had just started or were in the middle of the six-month follow-up. Social distancing and other precautions made delivery of MT in participants' and other health care facilities in their communities impossible. Each country operated with different governmental responses and levels of restrictions, and the continuously changing situation called for changes in the intervention protocol. We found moving all remaining MT sessions online to be the most feasible solution and made a modified MT protocol for virtual sessions, which were realized through the digital platforms that were most easily available in each country. The music therapists' feedback revealed that they were surprised about how well the sessions worked in this format, and that the families were very grateful to stay in touch and not lose the MT sessions. Virtual sessions could not fully substitute the live musical interaction of the original format. The heterogeneity of intervention delivery in the study increased, and hence virtual sessions were excluded from TF analysis. However, the shift to virtual sessions provided us a unique learning experience with how MT can be offered families who are outside commuting distance, or where MT services are not available. Based on our experiences in the LongSTEP trial, we believe virtual MT may be one way of contributing to more equal access to MT for families following preterm birth in the future.

4.3 Strengths and limitations

The LongSTEP trial is the first sufficiently powered RCT investigating the long-term effectiveness of MT on mother-infant bonding, maternal depression, and parent anxiety. We were successful in recruiting a large number of participants from a range of cultural contexts and retaining them in the study. Treatment uptake was very high, and no adverse events were reported related to the intervention, suggesting that the intervention was safe and acceptable. Completion of assessment was very high, indicating high acceptability of the outcome measures and study procedures. Where prior studies have predominantly focused exclusively on mothers and infants, or just the infant, our MT intervention and outcome measures included mothers, fathers, and

infants. The primary outcome was limited to mothers to promote homogeneity, but also because of the unique physiological and psychological changes resulting from pregnancy. Moreover, LongSTEP feasibility studies indicated that fathers were less present and available to complete intervention and assessments at some of our sites. LongSTEP is much more culturally diverse than prior large-scale studies of MT in neonatal care contexts. In our multinational trial we included countries in Europe, the Middle-East and South America, which likely increases the applicability of the findings to a broader range of settings. Collaboration across countries on large-scale studies can afford the involved partners possibilities for learning and competence building, which goes beyond the results of the research study itself.

While several limitations related to methodological factors like choice of primary outcome, reliance on self-report measures, and impact of Covid-19 have already been discussed, it is worth mentioning some additional points here. Due to the factorial design of LongSTEP, one fourth of the participants did not receive MT in either intervention phase. Participant disappointment with allocation to standard care was reported as a reason for dropping out of the study in one case (1/208. See Fig. 1 in Article II). However, at the sites that had established MT programs as part of standard care, MT was still a limited resource. Consequently, under normal circumstances all families would not have had access to MT either. At three sites MT was provided for the first time and was only accessible through participation in LongSTEP. Randomization can be seen as the fairest way to distribute MT when resources are limited.

In line with the pragmatic approach, eligibility criteria of infants and parents were intentionally broad to allow for some level of variability between sites in order to make the study feasible and stay close to usual care practice. However, one infant inclusion criterion might have resulted in a more homogeneous sample than we intended. This criterion concerned infants being “declared medically stable to start MT”. The lack of the criteria’s specificity was pointed out and discussed by study neonatologists during protocol development and planning of project implementation. A common-sense solution was to consider infants who were declared stable enough

for KC as likely to be stable enough for MT. However, some neonatologists in our study may have acted with more precaution. Such precaution could result from a lack of experience with premature infants' tolerance of MT. The sites in our study had varying degrees of experience with MT in the NICU, ranging from none to several years. Conservative interpretation of medical stability in some cases may have resulted in longer waiting times before approaching families about study participation. Thus, we may have included more mature infants than originally anticipated. Another possible result is that higher-risk or medically complex infants were excluded due to not achieving sufficient medical stability. The infant's severity of illness has been found to correlate with higher levels of maternal stress, which in turn affects maternal behavior towards the infant (Bozzette, 2007). Infants who are very preterm, have very low birth weight, or are neurologically compromised are at higher risk for later disorganized attachment (Wolke et al., 2014) Consequently, we might have missed a group of mothers with a higher risk for impaired bonding who might have otherwise been prioritized for MT in usual care practice. As a result, such the exclusion criterion moved our trial further towards an explanatory approach (Loudon et al., 2015).

The music therapists involved in the study were all trained in the intervention but differed regarding the extent of previous clinical experience and expertise, as well as personal qualities (Edwards, 2014; Elliott et al., 2018). The therapeutic alliance and therapists' skills may have affected study outcomes, despite the intervention seemingly being implemented in a uniform way. The intervention protocol was intentionally flexible. While its essential elements were operationalized into what we believed would be distinguishable and observable behaviors, the elements may have been overlapping or allowed for more variation in implementation than we were able to detect with our treatment fidelity tools and procedures.

Due to the nature of the intervention, it was not possible to blind participants and intervention providers, and the risk for bias must be taken into consideration when interpreting the estimated effects of the results. While heterogeneity is expected in a pragmatic trial it does make it statistically more challenging to identify treatment

effects as the estimates are pushed towards null. This can occur for two reasons. First, the effect itself may be reduced if the intervention works well in some cases but not so well in others, which again may be due to variations in intervention quality but also a broader mix of participants who may be more or less responsive. Second, the increased heterogeneity is associated with a larger standard error, reducing the test statistic. A strategy to accommodate for variation is to increase the sample size. In LongSTEP the sample size was slightly lower than what we aimed for (213 instead of 250 families); nevertheless, the trial was still sufficiently powered.

4.4 Implications

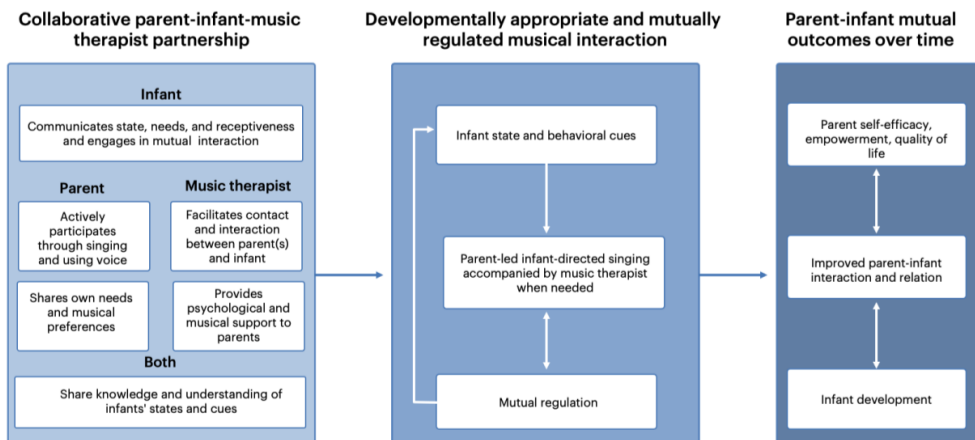
4.4.1 Adjusted model of proposed mechanisms of action

In Article I (Gaden et al., 2021) we presented a model of proposed mechanisms of action of the MT approach. We suggested that the collaborative parent-infant-music therapist relationship, together with developmentally appropriate and mutually regulated musical interaction, were two main mechanisms behind potential benefits of MT for parents, infants, and the bond between them. The model was a hypothesized model for our MT approach that was tested for the first time in the LongSTEP trial. Hence, it involved several uncertainties. With the results and experiences from the study in hand, I would like to comment on the proposed model (Figure 4) and suggest some adjustments that have implications for both practice and research.

We originally proposed that two main mechanisms might lead to change in parent-infant mutual outcomes over time: 1) the collaborative parent-infant-music therapist partnership, and 2) developmentally-appropriate and mutually-regulated musical interaction. The collaborative relationship with the unique contributions of those involved was proposed as a prerequisite for attuned musical interaction to take place. This interaction takes form through a) infants' expressing themselves through their state and behavioral cues, informing b) parent-led infant directed singing, which can lead to successful c) mutual regulation, and in turn modulate a) the infant state again.

In the new model (Figure 8), I have made some adjustments to underline the importance of music therapists providing parents with the adequate level of musical support. I also suggest changing some of the previously proposed outcomes in line with insights from the thesis' findings, as well as qualitative investigations linked to the LongSTEP trial. Other areas of the model are maintained as originally suggested.

Figure 8 Adjusted model of proposed mechanisms of action



Note: See Appendix 8 for a larger scaled version of the figure

Musical support in the collaborative parent-infant music therapist partnership

Our findings indicate that some parents hesitate in taking the lead when singing to their baby in MT. Hence, parents might need more musical support than we presumed. In the original model we proposed that the music therapist provides psychological support and integrates parents' musical preferences. Our results suggest that providing sufficient musical support should go beyond merely integrating parents' musical preferences, to providing what is needed musically for parents to feel comfortable. Although parents' potential need for musical support, and the therapists' responsibility for meeting these needs are both acknowledged in the descriptions of essential elements in the intervention protocol (Gaden et al., 2021),

the model is not as clear. Therefore, I recommend that music therapist provision of musical support should be explicitly stated and sidelined with psychological support in the model. This musical support should also be reflected in the second quadrant of the proposed mechanisms, and I have added in that the music therapist should accompany parent-led infant-directed singing as needed (e.g., through singing with them or accompanying with an instrument). This can be upon parents' request or when insecurity or discomfort with singing alone is expressed explicitly or perceived by the therapist.

Parent outcomes

In the original model's quadrant concerning parent outcomes, we used the wording "parent wellbeing" and "parental identity" while our study did not include any assessment of these outcomes per se. We measured symptoms of anxiety, depression, and stress. We did not measure or investigate aspects related to parental identity in the main trial but relied on related subprojects for insight into parent experiences and perceived benefits of our MT approach (e.g., Epstein et al. 2022). From our baseline measures, we found that parents in our study seemed to be doing quite well in terms of mental health challenges following preterm birth. These findings inform my suggestion of changing the proposed parent outcomes to less pathologic ones, such as parent self-efficacy, empowerment, and quality of life. Such outcomes would also be a better match to the resource-oriented principles behind our MT approach (Rolvsvjord, 2010).

Parent-infant mutual outcomes

We found low indications of impaired bonding in our participants, which correspond with other studies' findings of healthy bonding following preterm birth. Instead of reducing impaired bonding, I suggest that our MT approach might provide parents with competence and experiences that can help them discover innate musical resources, which in turn can contribute to improve the quality of relation and interaction between parents and infants.

Infant outcomes

Finally, the proposed outcome of infant development is retained from the original model since it will be tested at a later follow-up time point of the LongSTEP trial.

4.4.2 Implications for practice

In LongSTEP we suggested that the essential elements and guiding principles of our MT approach could be applied to sessions in both NICU and post-discharge phases. While this may still be correct, our findings indicate that some parents might need more time and support from the music therapist to be comfortable with singing. In line with parent feedback from the LongSTEP feasibility studies we recommend that sessions are done in private rooms where possible, to diminish any added stress of having other persons present. Parents voices are the most relevant ones to the infant, but we recommend music therapists to be mindful of individual differences in needs for support to build confidence with singing. For some parents this support may come in the form of instrumental accompaniment or having the music therapist singing together with them. The need for support may diminish over time as parents become familiar with singing and the positive responses singing can evoke in their baby. Some parents may not sing in sessions at all and rather prefer to try techniques demonstrated by the music therapist on their own. We also recognize that singing is not for everyone. Just like any therapeutic intervention, whether in research or in clinical practice, the MT-approach must be sensitively adapted to the client's needs and preferences and delivered according to the parents' wishes and voluntary participation. In clinical practice where MT may be a limited resource, targeting of the families that would benefit the most from MT is indicated.

We encourage music therapists to explore possibilities to offer MT after discharge from the NICU. While being discharged is a positive milestone for the families, the transition to home can also be challenging. Parent anxiety and stress may increase around the time of discharge as parents prepare for everyday life outside the hospital where they must take on the full responsibility for their child's care (Purdy et al., 2015; Zanardo et al., 2003). After discharge, some parents report isolation as their new lifestyles may be restricted due to fear of illness and health complications of

prematurity (Baraldi et al., 2021; Epstein et al., 2022). However, after discharge infants are developmentally more mature and their communicative abilities thrive (Epstein et al., 2022). Although stress and worries may still be part of parents' life, they may be less impacted by the acute crises of preterm birth and more prone to explore own caregiving resources and different ways of interacting with their baby in their home environment. For example, in our study we observed that parents' daily use of music with their baby increased in both MT and SC groups after discharge. van Wassenhaer-Leemhuis et al. (2016) recommend that support to premature infants and their parents should continue post-discharge. We suggest that MT can play a role in the continued support of infant development, parent wellbeing, and parent-infant interaction and relationships during the infants' first years of life. The period around the transition from NICU to home, and the first following months, may be of particular importance.

In LongSTEP we proposed that follow-up sessions took place in families' homes where possible. In settings where services such as early discharge through hospital at home are available, home visits may be made in collaboration with the nursing teams. Sessions can also be coordinated with follow-up visits at the hospital, or through private practice. In Norway we see a potential for collaboration with public health care centers in the municipalities. They are responsible for "The infant health care programme, age 0-5 years" which provide free healthcare for all children from birth until they start school (The Norwegian Directorate of Health, 2021). In addition to general monitoring of child development, the centers have an official mandate to support pregnant women and parents of infants and toddlers, as well as to observe and strengthen the interaction between parents and their children (The Norwegian Directorate of Health, 2022). Similar facilities in other countries can be apt settings for post-discharge MT.

4.4.3 Implications for research

Careful consideration of appropriate outcomes and outcome measures appears to be a key concern in future research. Our findings of healthy bonding in our participants, alongside with the inconclusive larger evidence base on bonding following preterm

birth, indicate that bonding as measured by the PBQ was not the most relevant outcome for our study. Bonding with the baby may not be experienced by parents as a concern at all, or not as pertinent a concern as infant survival, growth, stabilization, and development during NICU hospitalization. Measuring parent-infant bonding following MT might be more relevant if done with tools that better accommodate the situation of parents with premature infants, or if assessment is carried out in less critical stages of the NICU stay or during follow-up after discharge. One could even hypothesize that bonding is experienced differently following preterm birth (Bieleninik et al., 2021; Hoffenkamp et al., 2012), which calls for a more phenomenological investigation before outcomes and measures are chosen.

The inclusion of user-representatives in early stages of planning large-scale studies is one means to enhance the relevance of outcome measures to the population in question. A user-advisory board with parents of premature infants was consulted during the planning of LongSTEP. They confirmed that bonding was a relevant outcome. However, as described in this thesis, the term bonding can have many meanings. These parents may have had different understandings of what bonding meant based on their lived experiences. Nevertheless, this only demonstrates the importance of early involvement and careful joint exploration of the research's key ideas, concepts, and design when we as researchers consult and collaborate with the persons who are closest to the matter of our interest (Luu & Pierce, 2022).

We suggest investigation of MTs effectiveness on psychosocial outcomes that are less focused on pathology than the ones in our study. The prevailing narrative about the impact of preterm birth is a predominantly negative one, which can contribute to upholding negative stereotypes (Janvier et al., 2016; Luu & Pierce, 2022). However, as our study's findings of healthy bonding and good parent mental health following preterm birth show – the picture is more nuanced (Janvier et al., 2016). Although investigations related to positive transformations following neonatal hospitalization are scarce, descriptions of positive sides of the NICU experience can be found outside of the research literature like in first-person-reports in books, memoirs, and parent blogs (Janvier et al., 2016). Studies from domains other than medicine and pediatrics

that investigated longer-term effects of critical illness have found that positive changes often follow traumatic life-altering events, a phenomenon often-referred to as posttraumatic growth (Janvier et al., 2016; Linley & Joseph, 2004). While acknowledging the documented risks and burdens of preterm birth, aspects of growth and positive transformation should be included in the research agenda on the role of MT following preterm birth.

For future studies we recommend further investigation of MTs effectiveness on outcomes such as quality of life and wellbeing, instead of anxiety, depression, and stress. In a multinational study like LongSTEP, The Multicultural Quality of Life Index (MQLI) (Mezzich et al., 2011) could be an alternative, as well as more general wellbeing measures such as the WHO Wellbeing Index 5 (Topp et al., 2015).

During NICU hospitalization, outcomes like parent self-efficacy, agency and empowerment might be better matches to resource-oriented MT approaches – like the one in LongSTEP. One alternative could be The Parent Empowerment and Efficacy Measure (Freiberg et al., 2014) which uses a strengths-based approach to measure parent functioning. Another choice is a tool specifically tailored to the situation of parents with premature infants such as the Perceived Maternal Parenting Self-Efficacy (Barnes & Adamson-Macedo, 2007). An interesting secondary outcome could also be parent satisfaction with treatment and care during NICU, comparing MT and SC groups. The EMpowerment of PArEnts in THE Intensive Care (EMPATHIC) instrument (Latour et al., 2012; Latour et al., 2011) could be relevant for this purpose.

In further exploring the potential benefits of MT on the parent-infant relationship we recommend also using qualitative methods, including microanalysis of parent-infant interaction. Greater attention should be paid to how to support premature infants and their parents with different challenges at varying points in their lives (Luu & Pearce, 2022). We recommend further investigation of the effects of post-discharge MT, for example with an increased frequency of sessions (e.g., bi-weekly) or a longer total

course of follow-up as infants continue critical development over the first two years of life (Department of Health and Social Care, 2021; Leadsom et al., 2013).

5. Conclusions

The conclusions from this thesis, based on data collected from eight NICUs in five countries, Argentina, Colombia, Israel, Norway, and Poland, can be summarized as follows:

- The LongSTEP approach to MT for premature infants and their parents during NICU admission and follow-up emphasizes parent-led infant-directed singing, and is based on principles from developmental care, family-centered care, and resource-oriented MT. The intervention was found safe and feasible during both NICU admission and follow-up post-discharge across cultures and settings.
- No significant effect of MT on mother-infant bonding, maternal depression or parent anxiety was found at discharge from NICU. Narrow CIs on all estimates of group effects suggest that both beneficial and harmful effects could be excluded.
- Although we did not identify significant changes in mother-infant bonding with our chosen outcome measure (PBQ), this does not exclude the possibility that the LongSTEP approach to MT may impact bonding following preterm birth in ways not captured by the tool.
- Very few participants in our study had indications of impaired mother-infant bonding and poor mental health at baseline. The lack of effect may therefore also be attributed to an already good mother-infant bonding in most enrolled participants at the outset.
- Treatment fidelity analysis shows high music therapist adherence to the intervention protocol, suggesting that MT was implemented as a uniform intervention across sites and intervention phases.
- Parent evaluation of treatment receipt suggests that the MT intervention was received as intended, regardless of whether they participated in MT during NICU, follow-up, or both phases.

- The non-significant results of the RCT does not seem to result from inconsistent implementation of MT, but this should be addressed in secondary analyses and further research.
- The varying extent to which parents' voices were considered as prominent in fidelity evaluations of the MT suggest that our emphasis on parent-led singing can be challenging. Providing parents with a comfortable, private environment and sufficient musical support may help facilitate parental singing.

The research reported in this thesis has provided new knowledge on the effects and applicability of a MT approach designed to improve mother-infant bonding and parent mental health through singing. Future research should explore how this type of intervention can be further developed to target outcomes and processes relevant for premature infants and their parents during NICU hospitalization and beyond. New studies should ideally apply both quantitative and qualitative methods, as well as combinations of immediate physiological measures and a broader range of psychosocial outcomes that capture changes in long-term health and quality of life.

An important reminder provided by the parents in our trial was, that despite the unforeseen and often-dramatic circumstances surrounding preterm birth, the bond between parents and their babies is not necessarily at risk but develop normally. This finding helps to establish a broader, and more nuanced picture of parent-infant relationships following preterm birth. The strength and resilience that seems to lie within parents' love for their baby invites to further exploration of a potential role of resource-oriented MT approaches in supporting and boosting positive relational processes between premature infants and their parents.

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Article I

The LongSTEP approach: Theoretical framework and intervention protocol for using parent-driven infant-directed singing as resource-oriented music therapy

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ABSTRACT

Introduction: Despite medical advances, preterm birth and neonatal intensive care (NICU) hospitalization are demanding and pose risks for infants and parents. Various music therapy (MT) models have suggested parental singing to promote healthy bonding and development in premature infants, but evidence on long-term effects is lacking.


Method: We present the theoretical framework and intervention protocol of a resource-oriented MT approach for premature infants and their caregivers used in the international LongSTEP trial (ClinicalTrials.gov NCT03564184). We illustrate how guiding principles manifest in MT sessions, describe frames for phases of intervention, discuss prerequisites and present hypothesized mechanisms of change.

Results: The LongSTEP MT approach is resource-oriented, emphasizes parental voice and parent-infant mutual regulation, builds on family-centered care principles, and is relevant in the NICU and beyond. Essential elements include: observation and dialogue on infant and parent needs; voice as the main musical source, with parental voice as the most prominent; active parental participation; modification of music in response to infant states and cues; and integration of the family's culture and music preferences. The music therapist facilitates and supports interaction between parents and infant. Parents learn how to adapt principles in relation to infant development across NICU hospitalization and post-discharge phases.

Discussion: The LongSTEP approach is feasible in culturally diverse countries where consistent parental presence is available, but requires tailoring to local circumstances and culture, particularly in the post-discharge phase. The emphasis on parent-led infant-directed singing places a higher demand on parents than other MT approaches, and requires sufficient psychosocial and musical support for parents.

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Introduction

Each year, approximately 15 million infants are born prematurely, making preterm birth one of the world's biggest health challenges (Chawanpaiboon et al., 2019). Prematurity is associated in the long term with poorer mental health, cognitive development, and quality of life (Chawanpaiboon et al., 2019; Fevang et al., 2016). Premature infants require advanced medical care, and the preterm family will often spend weeks or months in a neonatal intensive care unit (NICU), depending on the severity of the infant's condition. Until the 1980s, NICU environments were often noisy and brightly lit places that exposed vulnerable premature infants to excessive and potentially harmful stimuli. Additionally, preterm infants and their parents faced long-term separation from each other as nurses and medical staff took on the primary caregiving roles during NICU hospitalization (Jolley & Shields, 2009). In the early 1990s, parent-driven grassroots organizations in the US demanded that parents receive easier access to information, inclusion in decision-making and more control over their children's care in the NICU (Gooding et al., 2011; Johnson, 2000; Jolley & Shields, 2009).

Following an organized collaboration between families and physicians, the establishment of *The Principles for Family-Centered Neonatal Care* (FCC) (Harrison, 1993) marked a paradigm shift in neonatal care and enabled greater integration of family-centered care within best practice (Hutchfield, 1999; Lawlor & Mattingly, 1998; MacKean et al., 2005). Early FCC initiatives resulted in models of care that involved the entire family in treatment planning, and empowered parents to take active roles in caregiving as part of the greater team. Developmental care models, such as the Newborn Individualized Developmental Care and Assessment Program (NIDCAP) (Als, 1986, 2009) and Mother Infant Transaction Program (MITP) (Rauh et al., 1988), based on Sameroff's transactional model (Sameroff, 2009; Sameroff & Mackenzie, 2003), have also contributed to improving NICU environments and care practices. Such care models provide strategies and tools to promote optimal conditions for infant neurological development and growth while also supporting parent-infant mutual processes such as the development of parent-infant bonding (Puthussery et al., 2018).

Despite these advances, preterm birth and NICU hospitalization are still demanding and often traumatic experiences that pose various risks for both premature infants and their parents. Parents of premature infants often experience high levels of stress, anxiety and fear for their infant's life (Bidzan et al., 2009), resulting in a greater risk of postpartum depression, significant stress (Sloan et al., 2008), and post-traumatic stress disorders (Korja et al., 2008; Muller-Nix et al., 2004; Winter et al., 2018). These mental health problems can in turn affect parental sensitivity and the capacity to respond appropriately towards the infant's cues (Bozzette, 2007; Field, 2010). Meeting the premature infant's physical and medical needs is a primary focus in neonatal intensive care, and although this focus is necessary and life-saving, it may alienate parents and affect the important relationship between parents and infant negatively (Bidzan et al., 2009). Parents must take on a role during NICU hospitalization that is challenging and complex, and that may compromise their ability to manage their own crisis of giving birth to a premature infant (Van Wassenaer-Leemhuis et al., 2016). The importance of addressing parents' own needs for emotional support is an emphasized part of FCC, but has been found to be the least developed aspect of FCC globally (Raiskila et al., 2016).

Although most contemporary NICUs have successfully reduced excess noise, the NICU environment and the care experienced within it still expose vulnerable infants to a range of unfamiliar and potentially overwhelming sensory input in the form of noise, smell, light and touch. The neuroplasticity of the developing premature infant's brain allows for adaptation and growth (Brummelte, 2017), but at the same time represents a risk for maladaptation in response to harmful or mismatched conditions in the environment, including mismatched parent-infant interaction (Brummelte, 2017; Flacking et al., 2012; Schore, 2001). Moreover, the premature infant's neurological immaturity results in subtle and quickly changing behavioral states and cues that can be difficult to interpret, increasing the risk for mismatched interaction and overstimulation of the infant (Bozzette, 2007; Sansavini et al., 2015). Conversely, sensory deprivation has been identified as a risk to premature infants residing in individual family rooms when consistent parental presence is lacking (Jobe, 2014).

The sound environment is particularly relevant when considering the risk of overstimulation and sensory deprivation. The sense of hearing becomes fully functional during the second trimester of pregnancy around week 25, exposing the fetus to external sounds (Graven & Browne, 2008; Hepper & Shahidullah, 1994; Moon, 2011). Experiments measuring fetal movement in response to external sounds have found fetal response as early as 19 weeks of pregnancy (Hepper & Shahidullah, 1994). The first sounds the fetus hears relate to social activities and voices, with maternal voice being dominant (Moon, 2011). Full-term newborns demonstrate a preference for their mother's voice (DeCasper & Fifer, 1980; Lee & Kisilevsky, 2014) and for infant-directed speech (Cooper & Aslin, 1990), which has implications for the role that parental voice plays. Parental voice provides a critical orienting link between the fetal auditory environment and the auditory environment experienced in the NICU (Loewy et al., 2013).

From birth, infants exhibit abilities related to perception of music that resemble those of adults (Trehub, 2001). Infants perceive communicative signals from their parents and in turn can stimulate communicative responses from them (Malloch, 1999). The cooperative and coordinated interactions that result from this communicative exchange are musical in nature. In particular, coordinated elements of pulse, quality and narrative combine to constitute *communicative musicality* (Malloch, 1999; Malloch & Trevarthen, 2009). The theory of communicative musicality provides a framework for music therapy (MT), where music's potential for connection, communication and interaction is the main focus. This theory, along with recognition of the central role of parental voice, has influenced the development of MT with premature infants and their parents.

Music therapy was first introduced to NICUs in the early 1990s through the pioneering work of Jayne Standley and colleagues (e.g. Cassidy & Standley, 1995; Standley, 1991). Their early work focused on the effects of audio stimuli on premature infants. Later clinical practice and research have incorporated live music and singing, increasingly including parents and family in MT (e.g. Cevasco, 2008b; Loewy, 2015; Shoemark, 2011b; Shoemark & Dearn, 2008). Several central contributors have highlighted the importance of song and parental voice. Shoemark (2011b) translated infant-directed singing to a strategy for hospitalized families, and emphasized empowering parents to sing and actively participate in MT (Shoemark, 2017; Shoemark & Dearn, 2008). Loewy (2015) emphasized the significance of family culture and musical preferences within her concept of *song of kin*, and evaluated its use in a clinical trial

(Loewy et al., 2013). Haslbeck (2014; Haslbeck & Bassler, 2020) contributed with an interactive, improvisational approach based on creative MT therapy, acknowledging the premature infant as an active partner and tailoring music to infant cues. Mondanaro et al. (2016) emphasized the unique contribution of fathers and the benefit of including both parents. Shoemark (2018) extended the impact of the music therapist's skillset by designing the parent education program *Time Together*, a single parent education session aimed at facilitating quality of parent-infant interaction through musical exchange.

Despite advances in practice and theory in neonatal MT, there is still a lack of experimental research exploring mutual outcomes of parents and infants, especially within a longer-term context. Informed by the aforementioned notable contributions, we propose a MT approach for contexts with a high degree of parental presence in the NICU, with an extension of MT from hospitalization through the first six months at home. The purpose of this article is to present the theoretical framework and intervention protocol for this resource-oriented MT approach for premature infants and their families. The LongSTEP MT approach is used within a multinational randomized controlled trial, and is centered around parent-driven infant-directed singing, developmentally appropriate interaction, and longer-term mutual benefits of MT for parents and infants. The LongSTEP approach to MT for premature infants and their caregivers is described in short in the trial protocol (Ghetti et al., 2019). In this article, we define and explain the theoretical underpinnings and principal guiding principles of the approach. We also illustrate how these principles manifest in MT sessions, describe frames for the phases of the intervention, discuss prerequisites required for implementing this specific approach and present hypothesized mechanisms of therapeutic change.

Context and prerequisites

The approach to MT described herein was developed for the international randomized clinical trial, LongSTEP (ClinicalTrials.gov Identifier: NCT03564184). The study is designed as a 2×2 factorial, multinational, assessor-blind pragmatic randomized controlled trial to evaluate the longitudinal effects of MT on preterm infants and primary caregivers across 12 months. Premature infants and parents were recruited from eight NICUs in Argentina, Colombia, Israel, Norway, and Poland. Randomization occurs at study enrollment and at discharge from the NICU, resulting in four conditions: (a) MT during NICU, (b) MT after discharge, (c) MT during NICU and after discharge, or (d) standard care without MT. During hospitalization, families participate in three MT sessions per week, and monthly (two sessions in the first month) for a six-month period following discharge. The main aim of LongSTEP is to evaluate the impact of MT on parent-infant bonding at six and 12 months corrected age, as measured by the Postpartum Bonding Questionnaire (PBQ) (Brockington et al., 2001). The secondary aim is to examine the effects on caregiver mental health and infant development. Eligible premature infants are born <35 weeks gestational age (GA), likely to be hospitalized at least two weeks from inclusion, and declared by NICU staff as medically stable to start MT (typically after 26 weeks postmenstrual age [PMA¹]) (Ghetti et al., 2019).

¹Postmenstrual age is defined as gestational age at birth plus the time since birth (American Academy of Pediatrics, 2004).

The LongSTEP approach is intended for level III and IV NICUs² where parental presence and participation are high. Hence, national parental leave policies that enable parents to be consistently present throughout hospitalization and for follow-up sessions after discharge are required for successful implementation. Each NICU must also support family-centered care principles of consistent parental presence and active participation in care, and parents must be willing to engage actively in MT sessions according to the LongSTEP approach through singing and using their voices, talking about infant and personal needs, and sharing musical preferences. The approach was originally developed within a Norwegian context (Ghetti et al., 2021) where infants have the legal right to parental presence at all times during hospitalization, and where family-centered care is consistently practiced (NOBAB, 2002). In the following, we specify foundational aspects of the approach and articulate the principal guiding elements that we consider essential and unique.

Guiding principles of the LongSTEP approach

The LongSTEP approach is informed by several theoretical constructs from existing MT models and research, as well as by developmental psychology and neuroscience. To promote safe uses of music with this vulnerable population, the approach includes progressive expansion of musical complexity and interaction in alignment with infant gestational age and readiness, consistent with guidelines for MT on NICUs (Standley & Walworth, 2010; Nöcker-Ribaupierre, 2013; Standley & Gutierrez, 2020), and recommendations for noise levels in NICUs (American Academy of Pediatrics: Committee on Environmental Health, 1997; White et al., 2013). The LongSTEP approach: (a) is explicitly resource-oriented, (b) is directed toward the parent-infant dyad/triad to support mutual processes between parent(s) and infant(s) and promote mutual benefits, (c) positions parental voice as the central and most important musical component of the musical interaction, (d) is aligned with family-centered and developmental care principles, and (e) aims to support parents in the evolving relationship with their developing infant so that the MT has relevance during NICU hospitalization, and also across infancy after discharge. [Figure 1](#) provides an overview of the prerequisites of the LongSTEP approach, the theoretical models and previous MT approaches informing it, as well as the approach's guiding principles, essential elements and desired outcomes.

A resource-oriented approach

A resource orientation consistent with that described by Rolvsjord (2010) is central to the LongSTEP approach and provides theoretical guideposts for the use of MT. Though Rolvsjord (2010) situates her theoretical elaboration within the context of mental health care, she also envisions the transfer of her concepts to practices in other contexts. Her sensitizing concepts that characterize a resource-oriented approach serve as guiding values in the LongSTEP approach to MT, namely: (a)

²Level III NICUs are hospital NICUs organized with personnel and equipment to provide continuous life support and comprehensive care for extremely high-risk newborn infants and those with critical illness, and level IV NICUs provide the highest level of neonatal care at a regional level with a full range of specialized services (Committee of Fetus and Newborn, 2012). In LongSTEP, seven of the included NICUs are level III and one level IV.

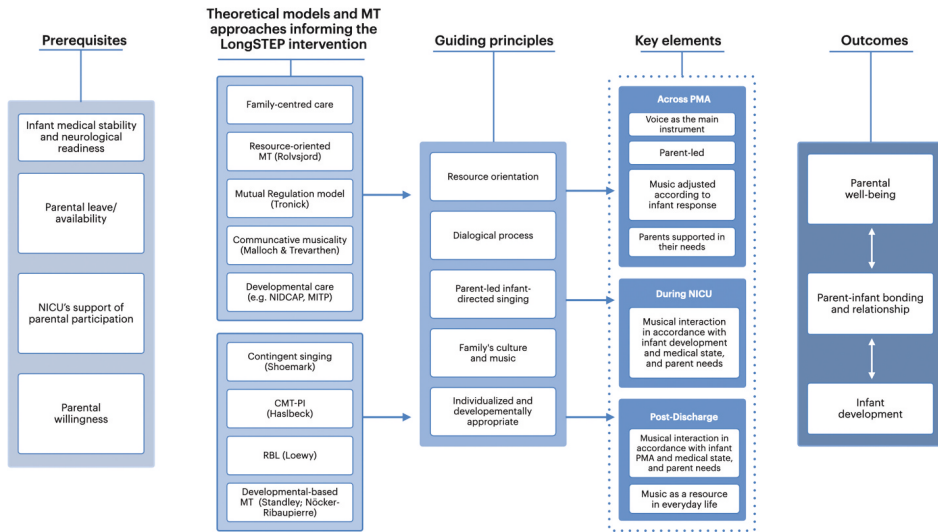


Figure 1. Theoretical framework and key elements of the LongSTEP approach

Note. The last names appearing in parentheses refer to authors associated with these approaches. References cited in the main text. Abbreviations: CMT-PI – Creative Music Therapy for Premature Infants; MT – music therapy; NICU – neonatal intensive care unit; NIDCAP – Newborn Individualized Developmental Care and Assessment Program; MITP – The Mother–Infant Transaction Program; RBL – First Sounds: Rhythm, Breath, Lullaby.

focusing on nurturing strengths, resources and potentials, (b) viewing the therapeutic encounter as a *collaboration* between client/user and therapist, (c) acknowledging and understanding the individual within their context, and (d) understanding music as a resource (Rolvsjord, 2010, p. 74). The fostering of inherent musical and parental resources is a central aspect of our approach. Parental voice, musical preferences, repertoire, culture and experiences are considered essential resources that can contribute to the formation of parental identity and self-confidence in parents and serve as tools to help them engage sensitively with their premature baby (McLean, 2016a; McLean et al., 2018). Parental voice is considered particularly important as it is an innate, accessible resource (Rolvsjord, 2004), and it provides the infant with a familiar and preferred sound that one can assume affords a sense of security in the unfamiliar NICU environment (Loewy, 2015; Shoemark, 2011b). Parents can be empowered through gaining access to and control over their resources, which may be particularly important in times of hardship and challenge (Rolvsjord, 2004).

Parents and infants possess the capacity to engage in reciprocal musical dialogue through coordinated communicative interactions (Malloch, 1999). Coordination between the communicative elements of pulse, quality and narrative combines to constitute communicative musicality (Malloch, 1999; Malloch & Trevarthen, 2009). Parental voice and the potential for communicative musicality may serve as health resources in congruence with a resource-oriented approach to MT (Rolvsjord, 2010). In our approach, parents become aware of how to attune to their infants and engage in cooperative communicative exchange. By actively carrying on these attuned musical

interactions in a variety of parent-infant situations, parents and infants actively participate in using music as a health resource.

Consistent with Rolvsjord (2010, p. 78), we view therapeutic change as occurring via a *collaboration* between the music therapist, parents and infant(s) that emphasizes equality, mutuality and participation. Though we acknowledge that the music therapist possesses expertise, we problematize the positioning of the therapist as “expert” in relation to parents. Instead, we emphasize parents’ developing expertise in knowing their infants, and support them in taking a lead as they engage in musical exchange. In our approach, both parents and music therapists share responsibility for what happens in MT, although they have different roles (Stige, 2002). Both have shared responsibility for forming a shared goal for a session, finding ways to work together and with the infant, and evaluating how these efforts have gone (Rolvsjord, 2010). Through a supportive approach and collaborative dialogue, the therapist helps parents to identify and expand their musical resources and learn to interpret and respond to their infant’s subtle signals via the musical interaction. The infant, in turn, participate with mutuality, responding as a communicative partner and making their needs known. These musical exchanges occur in response to infant states and cues as they evolve over time. Exchanges can result in musical attunement between the parent and infant (Stewart, 2009), and contribute positively to parents experiencing a relationship with their baby (Haslbeck, 2014; Ghetti et al., 2021).

A mutual process for mutual benefits

The LongSTEP approach views MT as a dialogical process that enables mutual regulation beneficial for both parents and infants. Our view of MT is, to a large degree, informed by developmental psychology, attachment theory, and research on early communication and interaction, with a particular emphasis on the significance of the human voice. From modern developmental psychology we, like several other neonatal MT approaches, adopt the view of the newborn baby, both full-term and premature, as a competent, relational being, entering the world with a capacity and need to interact with and relate to its close surroundings (Brazelton, 1978; Chamberlain, 1987; Slater, 1998). The newborn is, however, still very much dependent on available, sensitive caregivers to develop in a healthy manner. Self-regulation is a crucial capacity developed through interaction with close caregivers, recognized for its important role in promoting healthy development and wellbeing across the lifespan (Murray et al., 2015). In infants, self-regulation is exemplified when an infant intentionally disengages from an external stimulus, such as when averting their gaze (Rothbart et al., 1992). In early infancy, self-regulatory capacities begin developing through co-regulation or mutual regulation, where parents sensitively attune to infant cues and provide nurturing support through responsive interactions to calm and soothe the infant (Murray et al., 2015). Beeghly et al. (2011) argued that maternal sensitivity is best understood as a dyadic construct and as a component of the infant-caregiver communication system which is mutually regulating. The Mutual Regulation Model (Tronick, 1989) proposes that this dyadic mutually regulating communication system is made up of an infant subsystem, a parental subsystem, and the dynamic interaction between these subsystems (Beeghly & Tronick, 2011). According to this model, the success or failure of mother-infant mutual regulation depends on four reciprocal processes: (a) infants’ ability to self-organize and control their physiological

states and behavior; (b) the integrity and maturation of sensorimotor, attentional, and social-emotional elements of infants communicative system (e.g. gestures, gaze shifting, affective displays); (c) parents' ability to apprehend and correctly interpret their infant's communications; and (d) parents' motivation and capacity to respond to their infant contingently and appropriately to facilitate their infant's regulatory efforts (Beeghly & Tronick, 2011, pp. 197–198).

As premature infants are neurologically immature, and often medically compromised, they do not have the same capacity to self-organize and control their physiological states and behavior as full-term newborns (Feldman, 2009). Their behavioral states can shift rapidly and communication cues are often subtle, making them harder for parents to interpret (Richards, 2014). Developmental and family-centered care models, such as NIDCAP and MITP, involve guiding parents in understanding their baby's behavioral cues, and hence enabling them to provide developmentally appropriate and sensitive responses (Als, 2009; Rauh et al., 1988), which in turn contributes to establishing a good foundation for the parent-infant relationship (Beeghly et al., 2011; Tronick & Beeghly, 2011). Such guidance can foster mastery and self-confidence in parents (Puthussery et al., 2018).

It is, however, important to keep in mind that parents of premature infants often feel overwhelmed and are at risk of higher levels of stress and anxiety which may impair their responsivity towards the premature infant (Winter et al., 2018). It is thus important that the psychosocial needs of the parents are attended to in order to promote parental sensitivity, successful mutual regulation and interaction. Psychotherapeutic support can be offered through providing space for parents to share feelings and concerns, and receive their own musical self-care, as a way to support resilience in their caregiving roles (Loewy, 2015). For the LongSTEP approach to be successful, both infants and parents must be addressed in relation to their individual as well as mutual needs (Stewart, 2009). The LongSTEP approach is distinguished from parent education programs such as *Time Together* (Shoemark, 2018) in that sessions are conceived of as MT with therapeutic focus directed toward the parent-infant dyad/triad, and the music therapist provides psychotherapeutic support for parents during the course of MT to the extent that such support helps parents be present for their infant(s).

Parental voice is central

Parents singing for their babies is a cross-cultural phenomenon that represents an innate parental resource and act of caregiving behavior that has likely been around at all times (Dissanayake, 2009; De l'Etoile, 2006). For parents in crisis, talking to their infants might seem more natural than singing (Shoemark & Arnup, 2014). Developmental theorists have extensively investigated parent-infant interaction, and in particular the way mothers speak to their children (Beebe & Lachmann, 1988; Malloch & Trevarthen, 2009; Nakata & Trehub, 2011; Papoušek, 1996; Papoušek et al., 1991; Trainor et al., 2000; Trehub, 2001, 2009; Trehub & Trainor, 1998; Trehub et al., 1997; Trevarthen & Malloch, 2000). Maternal speech, *motherese*, or *infant-directed speech* are some of the terms used for the characteristic music-like way parents use their voices when engaging with their baby. Infant-directed (ID) speech has higher pitch, more exaggerated pitch contours, larger pitch range, slower tempo, and is more rhythmic than typical adult-directed (AD) speech (Trainor et al., 2000). Saliba

et al. (2020) found that both maternal and paternal ID speech promoted a quiet alert state in preterm infants, which is the behavioral state where the infant is most available and ready to engage (Beeghly & Tronick, 2011; Brazelton & Nugent, 1995; Nugent et al., 2007).

Infant-directed singing shares many features with ID speech, but singing additionally offers more structure through musical form, with more distinct and predictable phrasing, melodic range, and rhythmical patterns (Trehub & Trainor, 1998; Trehub et al., 1997). De l'Etoile (2006) suggests that "similarities between mother-infant interaction and ID singing provide a logical starting point for clinical interventions in music therapy" (p. 22). Furthermore, there is evidence that ID singing both captures the infant's attention and has regulating qualities delaying distress (Corbeil et al., 2013, 2015; Filippa et al., 2017; Saliba et al., 2018, 2020). Shoemark (2011a) proposed the concept of *contingent singing* to highlight the mutuality of interaction between the person singing (in her original conceptualizations, the music therapist) and the infant being sung to. The infant is not yet making his own music, but is an active participant, affecting and informing the song and interplay provided by the person singing. Filippa et al. (2020) found that contingent vocal interaction of parents, both speaking and singing, increased self-touch, eye-opening, and oral behaviors in premature infants. Infants responded differently to maternal singing than to maternal speech, however, demonstrating more rhythmical sucking behaviors and smiles to maternal singing, and more non-rhythmical mouth movements to maternal speech (Filippa et al., 2020). Singing might also offer parents a sense of both normality and intimacy, and a different way of relating than the typical NICU hospitalization experience (McLean, 2016b; McLean et al., 2018; Shoemark & Dearn, 2008).

A family-centered approach

Eight principles of patient-centered and family-centered care for newborns in neonatal intensive care were suggested by the European Science Foundation (ESF) European Research Network on Early Developmental Care in 2005, and later reviewed by Roué et al. (2017). These principles are evidence-based and are recommended as a worldwide standard (Roué et al., 2017). The principles include free 24-hour parental access, psychological support for parents, pain management, supportive environment, postural support, skin-to-skin contact, breastfeeding and lactation support, and sleep protection (Roué et al., 2017). Families are recognized as having the greatest influence on a child's health and wellbeing, and parents are ensured active participation in treatment and care. To support parents in their active role, nurses and other medical staff work in partnership with parents, demonstrating, explaining and supporting, rather than merely directing parents (Johnson, 2000). Family-centered care can shorten the total length of hospital stay for premature infants (Yu & Zhang, 2019), and is increasingly viewed as best practice in child health-care settings (Hutchfield, 1999; Lawlor & Mattingly, 1998; MacKean et al., 2005). However, FCC remains a somewhat broad concept that lacks standardization, resulting in a variety of different understandings of what comprises FCC, and how these values should be translated into action in a way that honors each individual family (Dennis et al., 2017; Kokorelias et al., 2019).

Consistent with Ullsten et al. (2020, p. 67), in the LongSTEP approach we consider and respect each family as a unique entity with its own musical history, musical

preferences, and unique intergenerationally transmitted attachment patterns. Parent participation and decision-making are essential for the collaborative and resource-oriented MT process of the LongSTEP approach, and to support parents in the development of their parental role (Broom et al., 2017).

Relevant for the infant's early development in the NICU and beyond

In a meta-analysis of randomized trials of MT for premature infants and their caregivers, Bieleninik et al. (2016) identified a substantial evidence gap related to the longer-term impact of neonatal MT, and the effects of intervention periods extending past discharge from NICU. The post-discharge phase of the LongSTEP approach addresses this gap by enabling continuity of care that bridges from hospital to home (Ghetti et al., 2019). A baby's first 1001 days, from conception until two-years-old, is increasingly considered a critical period that affects health and wellbeing throughout life (Leach, 2017; Leadsom et al., 2013). When working with families, this time window offers unique possibilities. A range of intervention programs aim to support the development of premature infants, with a majority including parents as active participants. In a meta-review, Puthussery et al. (2018) found that intervention programs with home and facility-based components for parents of preterm babies affected the quality of the maternal-infant relationship, mother-infant interactions, symmetrical and asymmetrical co-regulation, mutual attention, and maternal sensitivity and responsiveness. Van Wassenae-Leemhuis et al. (2016) reviewed randomized controlled trials of home-based family-centered intervention programs in very preterm infants aiming to improve cognitive outcomes, and concluded that research aimed at improving outcomes in premature infants must continue after discharge from the NICU. The evidence-base suggests that both infant development and parent-infant relationships develop over time, and interventions should be designed and offered accordingly. In LongSTEP, we offer MT beyond NICU hospitalization through a six-month period. In a feasibility study of the approach, parents expressed that they have found it particularly interesting to note the increasing responsiveness and interaction with their child over time (Ghetti et al., 2021). To our knowledge, LongSTEP is the first RCT to investigate the effectiveness of MT extending from NICU hospitalization through 6-months post-discharge.

Key aspects of the LongSTEP approach

The following description of the LongSTEP approach is an elaboration of the MT intervention description provided in the study protocol (Ghetti et al., 2019) tying it more clearly to the aforementioned theoretical basis. This elaboration includes a description of preparations to be made before initiating MT sessions and essential elements of the approach, as well as a discussion of aspects specific to the two phases of the intervention.

Before beginning the actual MT sessions, we propose that the therapist should meet with the family twice. First, to begin to develop therapeutic rapport, introduce basic concepts and potential aims of MT, map the family's musical preferences and experiences, and get to know other aspects that are important for the family, for example, cultural background and religious or spiritual beliefs. Second, we propose a separate session where the music therapist observes parent-infant interaction in a situation that

involves handling, for example, during diaper change, physiotherapy, or transitions from incubator/cot. Such observations can provide insights into infant development, thresholds for overstimulation, and early strategies for self-soothing. This observation also demonstrates how parents interpret and respond to their baby's cues and states.

Essential elements and fidelity assessment

Seven elements (summarized in [Table 1](#) and described in detail below) are considered essential for the LongSTEP approach, and should be manifest in each session regardless of the infant's postmenstrual age and development or the phase in which MT is provided. These elements describe the essential aspects of the approach in a form that is discretely observable for the purposes of treatment fidelity assessment. These essential elements form the basis for treatment fidelity assessment tools in the project. The descriptions and the assessment tools have been pilot tested; the results of treatment fidelity will be reported separately.

Observation and dialogue on the infant needs prior to and during MT sessions

Like the two preparation sessions, this introductory part of each session allows for an initial observation of the infant's current state and readiness for interaction. The observation should begin with some brief quiet time, where the therapist and parents watch the infant together to identify its current state and needs at that moment. This includes looking at physiological aspects such as respiratory rate and pulse, skin color, motor activity and quality of movements, sleep and awake states, and responsivity, similar to how it is carried out in newborn behavioral observation (NBO) sessions (Nugent et al., 2007). Parents are then invited to have a dialogue on their observations and thoughts on what their baby might need or benefit from at that particular moment. Additionally, adjustments are made to ensure that the infant is physically positioned to promote self-regulation. Most infants will either be held by the parent or positioned so that the infant's arms and legs are gently supported in a tucked and flexed position, with arms supported so that the infant's hands come towards midline. These principles of facilitated tucking and support for midline alignment can help to promote physical containment and autonomic regulation, enhancing general behavioral development (Hill et al., 2005; Hunter, 2010; Vergara & Bigsby, 2004). If the infant is drowsy, parents can promote quiet alertness by positioning the infant slightly more upright. The first part of the session helps to prepare the infant and parent for developmentally appropriate musical interaction and functions as an initiation into the collaborative and dialogical exchange that characterizes the therapeutic approach. Identifying the specific needs of the infant and discussing how music and voice can serve these might also add meaning to the act of singing for parents who are not motivated or feel insecure (Shoemark, 2017).

Dialogue with parents on their state and needs prior to a session

Since parents of premature infants can experience significant stress and traumatization associated with premature birth, and this intervention aims to support mutual outcomes of both infant and parents, it is important to give sufficient attention to parents' own needs and concerns. There is a consensus that parent wellbeing and mental health affect parent-infant interaction (Beeghly et al., 2011; Field, 2010; Korja et al., 2008), and parents of preterm infants are particularly vulnerable (Flacking et al., 2012; Korja et al.,

Table 1. Essential elements of the LongSTEP approach

Element	Description	Operationalization
1. Observe and dialogue on infant needs	At the beginning of the session, the music therapist and parent(s) observe and discuss the infant's current state and needs prior to presenting any music	Parent(s) and therapist attend to infant's <ul style="list-style-type: none"> ● physical positioning ● behavioral state (e.g. awake, drowsy, sleeping) ● physiological cues (e.g. skin color, breathing patterns, heart rate) Parent(s) and therapist might also discuss <ul style="list-style-type: none"> ● status of infant's development of self-regulatory capacities/strategies ● infant's tolerance and normal responses to sensory input (e.g. touch, handling, noise) ● infant's preferences for soothing ● early signs of communication/interaction (e.g. eye contact, smiles, alertness) ● current medical or other challenges/concerns regarding the infant (e.g. feeding, coming off ventilation, pain, recent or upcoming procedures)
2. Dialogue on parents' needs	At the beginning of the session (and during, as appropriate), the music therapist discusses with the parent(s) about the parents' own state, needs, and concerns	Music therapist invites parent(s) to share <ul style="list-style-type: none"> ● how they are feeling ● thoughts and concerns about the situation ● how they are doing in relating with their infant ● questions and wishes for the MT session
3. Assure voice is main instrument	Voice serves as the main instrument during the MT session	<ul style="list-style-type: none"> ● Song and voice are the most evident sources of music offered ● Other instruments (e.g. guitar, lyre) are used with moderation, preferably only when requested by parent or when considered necessary to promote/encourage continuation of parental singing. If accompaniment is provided, it is delivered in an infant-appropriate way (e.g. simple accompaniment, appropriate tempo and volume)

(Continued)

Table 1. (Continued).

Element	Description	Operationalization
4. Assure parental voice is prominent	Parental voice serves as the main instrument during the MT session	<ul style="list-style-type: none"> ● Music therapist consistently ensures opportunities for parents to take on a leading role in singing ● Parental voice is distinguishable during the MT session ● When singing together with parents, the music therapist modifies volume of own singing and takes on a supportive role musically ● If parents are hesitant or insecure about singing, the music therapist demonstrates, and supports parents in how musical qualities of the spoken voice can be used in an infant-directed way (e.g. with variations in pitch, tempo, use of pauses, vocal inflection)
5. Support parental participation	Music therapist provides opportunities for parents to participate actively during the MT session	<p>Music therapist encourages and guides parents to</p> <ul style="list-style-type: none"> ● sing or hum in an infant-directed way ● touch and move infant in response/relation to the music (when tolerated by infant) ● adjust infant's physical positioning to enable optimal conditions for interaction or containment <p>If parents are hesitant to sing, the music therapist offers other ways of participating (e.g. writing or modifying lyrics of parent-chosen songs)</p>
6. Provide music that is infant-directed and developmentally appropriate	Music is modified in response to infant's cues and responses during the MT session	<ul style="list-style-type: none"> ● Song and voice use during the session are infant-directed and attuned to cues and responsivity ● Parents and music therapist attend to and modify music according to infant's state and engagement/disengagement cues ● Music is paused or stopped in response to signs of overstimulation.
7. Integrate family's culture and musical preferences	Parents' musical preferences and abilities are integrated and accommodated into the MT session	<ul style="list-style-type: none"> ● Parents are encouraged to share preferences and familiar songs (e.g. lullabies, children's songs, pop music, music representing parents' nationality/culture/religion/belief), which are subsequently adapted and integrated within the sessions ● Music therapist accommodates parents' musical abilities (e.g. no/some experience with singing) and modifies music (e.g. models/accompanies in comfortable vocal range/key) to facilitate parental musical engagement

2008; Lasiuk et al., 2013; Loewy, 2016; Misund et al., 2013; Muller-Nix et al., 2004; Sloan et al., 2008; Stewart, 2009). Therefore, attending to parents' own need for stabilization, self-regulation, and restorative experiences forms a necessary part of the intervention, in alignment with trauma-preventive approaches to NICU MT (Loewy, 2016; Stewart, 2009). The music therapist provides space for parents to share feelings prior to and during sessions, and helps shape sessions to address parents' needs. Thus, parents' need for stabilization, self-regulation, integration of traumatic experience, and restorative experiences (Stewart, 2009) might dominate in portions of a session, and the therapist gently brings the focus back to the parent-infant interaction when appropriate.

Voice serves as the main instrument

For parents, the voice is an innate resource, accessible at all times. Parental voice forms a supportive environment for the premature infant (Roué et al., 2017), and parental singing promotes the formation of parental identity (McLean, 2016a). In the LongSTEP approach, parents are empowered to use their singing and spoken voices in an infant-directed manner, such that parental voice consistently serves as the main instrument. Singing can reduce arousal for parents and infants (Cirelli et al., 2020), enable parents to experience emotional closeness (Fancourt & Perkins, 2018), and help parents experience and contain a broad range of feelings to help them work through challenging emotions (McLean, 2016a). The stabilization and mutual regulation that occur during infant-directed singing may then enhance parents' quality of presence and sensitivity when interacting with their infant. The music therapist guides and musically supports parents in attuned use of their voice. Other musical instruments, such as guitar or monochord, are used in moderation, preferably only when requested by parents or when considered necessary to encourage continuation of parental singing (Ghetti et al., 2019). At times, particularly during initial sessions, parents can feel insecure about singing, and simple instrumental accompaniment can be of help. Such accompaniment should be delivered in an infant-appropriate way with adjusted tempo, volume, and level of complexity.

Parental voice serves as the most prominent musical voice

Acknowledging the central importance of parental voice (e.g. Filippa et al., 2017; Saliba et al., 2020), the music therapist provides adequate space and support so that parents can become comfortable, gradually taking on a leading role in singing and/or using an attuned spoken voice. The therapist assures that parental voice is clearly distinguishable during MT sessions by modifying their own singing volume to take on a supportive musical role when singing together with the parents. Through demonstration, the therapist guides parents in how to adjust musical elements such as pitch, tempo and rhythm to tailor musical interaction to infant needs and responses (Haslbeck, 2014; Nöcker-Ribaupierre, 2013; Shoemark, 2011b) so that the infant, in effect, "directs" the parents' way of singing. Later in the therapeutic process as the infant matures, touch and movement can be integrated into sessions (Nöcker-Ribaupierre, 2013).

Music therapist provides opportunities for parents to participate actively

The music therapist encourages and guides parents to sing or hum in an infant-directed way, and invites them to touch and move their infant in response/relation to the music (when tolerated). If parents are reluctant to sing, the music therapist offers

alternative ways of participating, such as writing lyrics or modifying existing lyrics of significant parent-chosen songs (Ettenberger & Ardila, 2018; Loewy, 2015) that can be used in future sessions. The music therapist creates adequate space for parents both musically and verbally, and asks questions to help parents reflect on what they do musically and how the infant responds (both inside and outside the session).

Music is modified to infant cues and responses

During introductory sessions, the music therapist introduces the concept of infant-directed singing and describes how parents can attend to and modify music according to their infant's state and engagement and disengagement cues. Parents learn how to begin with single tones or single words matched to infant breathing patterns, facial expressions and movements, and over time begin to add more complexity in terms of melodic contour, rhythmicity, phrasing and multisensory aspects of touch and rocking. Parents learn how to simplify the musical interaction when the infant shows signs of disengagement by slowing tempo, simplifying the melody or lyrics, or providing a temporary pause. However, the music therapist has overall responsibility for providing music safely for the developing infant. Thus, it is important that the music therapist using this approach has thorough training in recognition of infant engagement and disengagement cues. Such training, as provided to the therapists in this study, maximizes therapist competence in safely facilitating the approach. Further specification of our progressive sequence is found below and in the protocol (Ghetti et al., 2019).

Parents' culture and musical preferences and abilities are integrated into sessions

Parents are encouraged to share preferences and personally significant familiar songs that are subsequently adapted in an infant-appropriate manner and integrated within the session. Loewy (2015) refers to these songs as *songs of kin*, which include both lullabies and other types of music (e.g. parent-preferred popular music, music representing parents' nationality/culture/religion/beliefs, etc.). The music therapist accommodates parents' levels of musical experience and modifies music accordingly (e.g. selects a comfortable vocal range) to facilitate parental musical engagement.

Aspects specific to MT during NICU hospitalization

Families in the LongSTEP study that receive MT during NICU hospitalization commit to participating in three sessions per week. This frequency ensures: that the infant's evolving needs are addressed in a timely manner; that parents have ample opportunities for practice, repetition and constructive dialogue with the therapist; and that parents' own psychosocial needs can be met during the course of a NICU stay. We aim for a minimum of six sessions in the NICU, and conclude at a maximum of 27 in order to conserve resources and minimize heterogeneity among participants. Sessions last between 30 and 40 minutes, with time spent actively making music typically lasting from 15 to 30 minutes in accordance with infant tolerance (Standley & Walworth, 2010; Nöcker-Ribaupierre, 2013).

Music therapy can be provided during kangaroo-care, feeding or with the infant lying in the incubator or cot. In the NICU phase of the intervention, although declared medically stable for MT, infants are smaller, more neurologically immature and medically fragile than in the post-discharge phase. Following Nöcker-Ribaupierre's (2013) guidelines, MT with infants of GA 26–32 weeks should contain primarily

cautious use of singing and toned voice without accompanying instruments. Predominantly wordless singing is matched to infant responses, aiming to promote sleep or quiet alert state, depending upon infant need (Ghetti et al., 2019; Nöcker-Ribaupierre, 2013). From week 32 onwards, previous elements can be expanded upon by including dynamic touch, parent facial expressions and gesticulations, vocal inflection and phrasing to promote musical interplay, rudimentary musical dialogue and social interplay with the infant. The physical location of sessions should be considered since aspects of the physical environment can affect parental comfort with singing (Ghetti et al., 2019). If private spaces are not available, the use of folding screens can help create a more intimate atmosphere in open bay units. Music therapists should collaborate with parents and staff to ensure that sessions do not interfere with other treatment appointments or care routines. Preterm families often receive a range of services during hospitalization, and MT should not occur directly following sensory-demanding procedures or therapies due to the risk of overstimulation.

Aspects specific to MT after discharge from hospital

The sessions after discharge are provided monthly over a period of six months in the LongSTEP study, with two sessions during the first month to enable assessment and rapport building (particularly important for families who did not have MT during NICU hospitalization in the study). Providing seven sessions in the first six months enables the development of a collaborative alliance between parents and music therapist where musical interaction approaches can be tailored in alignment with infant development over time, and the changing needs of infant and family. Monthly sessions assure that parents have time to develop skills and practice in their daily lives between sessions. Infants who have been discharged home are typically more stable and more neurologically mature than those who are hospitalized. Consequently, musical interaction in the post-discharge phase can include more complexity and longer periods of active music-making, depending upon infant tolerance. Post-discharge sessions last between 45 minutes and one hour, and follow a set structure with approximately 15 minutes spent in verbal dialogue during the session (Ghetti et al., 2019). Considering that we aim for parents to use musical interactions in their everyday lives and contexts, it is ideal for sessions to occur in the family's home, but they can also be held at the hospital or other health-care settings convenient for the family. Family needs are assessed at the beginning of sessions, and parents are asked to identify particular strengths and challenges they are experiencing. Parents then discuss various approaches with the music therapist to meet these needs and can attempt such while receiving support from the therapist (Ghetti et al., 2019). Siblings who are available may be included in the sessions in an attempt to make the interactions more relevant to real-world family dynamics.

Proposed mechanisms of action

Figure 2 illustrates the proposed mechanisms of action of the LongSTEP approach. A foundation of this approach is the collaborative partnership between parents, music therapist, and infant(s), where general knowledge on premature infant development and specific knowledge of the individual infant's needs and

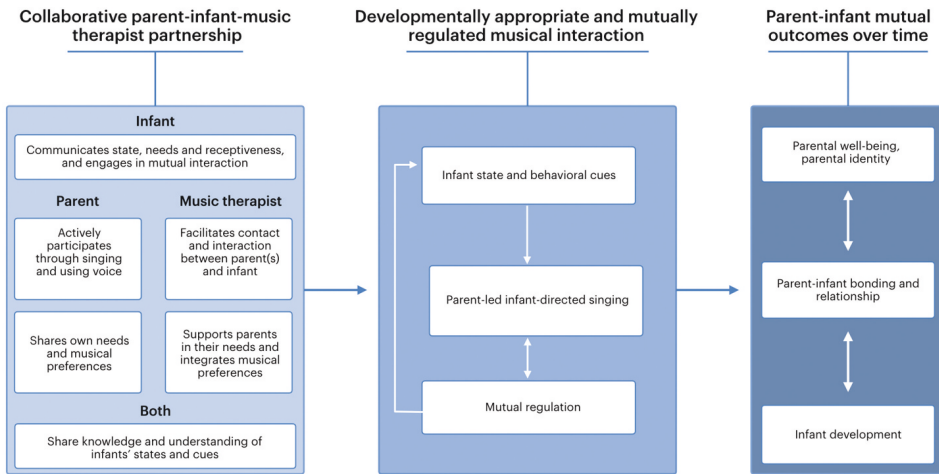


Figure 2. Proposed mechanisms of action of the LongSTEP approach

preferences are shared. In this relationship, the music therapist supports parents in regard to their immediate psychosocial needs (e.g. fear, hopelessness, traumatic stress responses), and their developing sense of parental identity and relationship with their infant. This partnership lays the foundation for parents feeling comfortable enough to explore the use of song and voice with their infant while receiving musical and emotional support from the music therapist. The music therapist facilitates interaction between parents and infant, supporting an attuned form of early dialogic communication that improves the quality of their relationship. The therapist models techniques and principles, and encourages parents to try these out with support. The infant possesses the potential for communicative musicality, and expresses either receptiveness to musical interaction or aversion to it via engagement and disengagement cues. The collaborative effort of the parents and the music therapist jointly interpreting and attending to the infant's cues helps promote attuned, developmentally appropriate musical interactions that enable beneficial parent–infant mutual regulation. Parents are empowered to use inherent musical resources, including their voices and music preferences, to develop a relationship with their infant. By singing to their infants, parents experience a reduction in arousal. As parents become regulated themselves, they can more sensitively respond to their infants musically, promoting attuned interaction with their infant and enabling mutual regulation between them. Experiencing mastery in MT through recognizing infant responses to their voices and effectively meeting their infant's needs, the parents' sense of parental identity and perception of parent–infant bond may strengthen. With a secure parent–infant bond, parents can carry forward abilities they experience in MT to future attuned musical interactions with their infant, adeptly adapting the exchange as the infant develops. By building capacity for attuned musical interaction and enabling experiences of mastery and parental identity, we hope to improve the quality of relations between parents and infants, creating a basis for better parental mental wellbeing and, ultimately, improved infant development over time.

Critical reflections

The LongSTEP approach requires contexts where parental presence is encouraged and safeguarded throughout NICU hospitalization, and thus it may not be feasible to implement it in NICU settings where parental presence is limited. The approach places high demands on parents who might already feel overwhelmed by other tasks and responsibilities during NICU hospitalization. We support Shoemark (2017) in acknowledging that singing is not a natural action for everyone, and that with today's increasingly individualized music listening technology and decreasing opportunities for active music-making in many countries, people might be less connected to their musicality. The role of the music therapist in the LongSTEP approach is to support parents in the music therapeutic process in a power-balanced, non-directive, and collaborative manner. Experiences from training music therapists and implementing the approach in the study's participating countries indicate that it may be challenging for music therapists to shift away from a familiar role of leading the music in order to provide adequate space for parents to take a lead. Although we strive for equality in the relationship between therapist and client through a resource-oriented approach, therapeutic relationships always involve power dynamics (McLean, 2016a; McLean et al., 2018; Rolvsjord, 2010; Shoemark, 2017), which must be acknowledged and actively challenged. A successful music therapist–parent partnership will be shaped by cultural and contextual factors, as well as the therapist's previous experience, personality and skills (Edwards, 2014; Loewy, 2015; Shoemark & Ettenberger, 2020). It is important, therefore, that the music therapist is able to sensitively perceive and respond to complex layers of culture, context and experience; thus, capacity for self-awareness and self-reflection is key.

Supervision provides a tool for promoting capacity for self-awareness and self-reflection. Music therapists trained for the LongSTEP trial received supervision from the trial's core team, both individually and in smaller groups. Four of our participating sites had more than one music therapist while four music therapists were alone with the responsibility of providing the intervention. Therefore, we considered it important to combine individual supervision with group sessions, aiming to support successful implementation through promoting therapists' self-awareness and providing a space where challenges could be discussed and experiences celebrated with peer support. Therapists reported that it was fruitful to share strategies from the different sites, which highlighted therapists' individual strengths and weaknesses, as well as contextual and culturally specific themes and challenges. Music therapists who engage in clinical practice in a manner consistent with the LongSTEP approach are encouraged to seek regular clinical supervision to work through their experience of this sensitive work.

Conclusion

In this article, we have developed the theoretical rationale, guiding principles, essential elements, and proposed mechanisms of action of a MT approach for premature infants and their caregivers developed for an international pragmatic RCT. We have presented and discussed prerequisites and particular considerations for carrying out this approach during NICU hospitalization and during follow-up post-discharge. We hypothesize that many parents will need repetition, practice, support and constructive dialogue in order to make lasting changes and establish new ways of relating with their

infant. However, other approaches such as *Time Together* (Shoemark, 2018) show that single parent education sessions can also have the potential to improve the quality of parent–infant interaction. Different approaches may meet families’ needs in different ways.

The LongSTEP approach is resource-oriented and family-centered, where infants and parents are supported in a dialogic MT process, aiming for mutual outcomes in the form of improved parent–infant bonding, parent wellbeing and infant development. In the LongSTEP study, we explore parental perception of parent–infant bonding. Future research could expand our understanding by exploring the impact of this MT approach on infant attachment to caregivers. LongSTEP is an international and multicultural research study. Further investigation of how cultural factors affect implementation and perception of the approach is needed.

Disclosure statement

Claire Ghetti is Associate Editor of the *Nordic Journal of Music Therapy*. To avoid conflict of interest, Claire Ghetti was fully masked to the editorial process including peer review and editorial decisions and had no access to records of this manuscript.

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Article II

Short-term Music Therapy for Families With Preterm Infants: A Randomized Trial

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abstract

OBJECTIVES: To evaluate short-term effects of music therapy (MT) for premature infants and their caregivers on mother-infant bonding, parental anxiety, and maternal depression.

METHODS: Parallel, pragmatic, randomized controlled-trial conducted in 7 level III NICUs and 1 level IV NICU in 5 countries enrolling premature infants (<35 weeks gestational age at birth) and their parents. MT included 3 sessions per week with parent-led, infant-directed singing supported by a music therapist. Primary outcome was mother-infant bonding as measured by the Postpartum Bonding Questionnaire (PBQ) at discharge from NICU. Secondary outcomes were parents' symptoms of anxiety measured by General Anxiety Disorder-7 (GAD-7) and maternal depression measured by Edinburgh Postpartum Depression Scale (EPDS). Group differences at the assessment timepoint of discharge from hospital were tested by linear mixed effect models (ANCOVA).

RESULTS: From August 2018 to April 2020, 213 families were enrolled in the study, of whom 108 were randomly assigned to standard care and 105 to MT. Of the participants, 208 of 213 (98%) completed treatment and assessments. Participants in the MT group received a mean (SD) of 10 sessions (5.95), and 87 of 105 participants (83%) received the minimum of 6 sessions. The estimated group effect (95% confidence interval) for PBQ was -0.61 (-1.82 to 0.59). No significant differences between groups were found ($P = .32$). No significant effects for secondary outcomes or subgroups were found.

CONCLUSIONS: Parent-led, infant-directed singing supported by a music therapist resulted in no significant differences between groups in mother-infant bonding, parental anxiety, or maternal depression at discharge.



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WHAT'S KNOWN ON THIS SUBJECT Significant positive effects of music therapy (MT) have been confirmed on premature infants' respiratory rate and maternal anxiety. Small-scale studies have suggested positive effects of MT in NICU on parent-infant bonding, but large-scale studies investigating parent-infant mutual outcomes are lacking.

WHAT THIS STUDY ADDS This randomized controlled trial investigating bonding and parental mental health demonstrated high retention of parents in music therapist-supported, parent-led, infant-directed singing with their preterm infants during NICU, despite no significant effect on parent-infant bonding. Research using salutogenic outcomes is indicated.

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Preterm birth is considered a global health challenge, as prematurity is associated with higher mortality and poorer mental health, cognitive development, and quality of life for the child.¹ Premature birth (before 37 weeks' gestation)¹ affects both infants and their families. Parents of premature infants are at greater risk for developing symptoms of anxiety, depression, and posttraumatic stress disorder than parents of full-term babies.²⁻⁶ Moreover, delays and disturbances of parent-infant bonding have been described as a consequence of the distress of experiencing preterm birth and uncertainty about the health and development of the child.^{2,3,7,8} Bonding refers to parental perception of the emergent parent-infant relationship, including feelings, thoughts, and behaviors of the parent toward the baby.^{9,10} Bonding is influenced by physical proximity between parent and infant after birth¹⁰ and may be supported by breastfeeding, co-rooming and other proximity activities such as skin-to-skin holding.¹¹ Bonding is also influenced by maternal emotional state¹² and infant ability to communicate needs to the parent.¹³ This initial parent-infant bonding contributes to the quality of the early parent-infant relationship, which may have long-term implications for infant health.¹⁴⁻¹⁶

Music therapy (MT) refers to the informed use of music, facilitated by a music therapist within a therapeutic relationship, whereby engagement in musical processes serves as a resource to promote health. MT in neonatal care is consistent with principles of family-centered care and aims to support infant development and parent-infant relationship by empowering parents in their parental roles and understanding of their infant.^{17,18} A meta-analysis of MT in neonatal care demonstrates short-term impacts on

infant respiratory rate and maternal anxiety, but evidence of long-term effects on infant and parental outcomes and effects on parent-infant mutual outcomes is lacking.¹⁹ Pilot studies and small-scale randomized controlled trials (RCTs) suggest improvements in parent-premature infant bonding after MT.²⁰⁻²⁵

Parental voice emerges as a key resource when aiming to strengthen parent-infant bonding. During pregnancy, fetal auditory capacity is fully developed around 25 gestational weeks, and fetal response to auditory stimuli has been recorded as early as 19 weeks.^{26,27} Thus, parental voice, and particularly maternal voice, can serve as a familiar and comforting link between the protected environment in utero and the often-overwhelming NICU sensory environment.^{26,28} Evidence from systematic review suggests that live and recorded maternal voice interventions are associated with physiologic and behavioral stabilization of preterm infants, with fewer cardiorespiratory events.²⁹ In mothers of full-term infants, singing is associated with increased positive affect, reduced maternal cortisol levels, improved perceived mother-infant closeness,³⁰ and faster improvement of symptoms of moderate-severe postpartum depression.³¹ Soothing singing is associated with decreased arousal levels in both mothers and infants.³² Parents of premature infants report that MT focusing on voice and singing reduces anxiety²⁵ and provides tools to enhance early communication and intimacy.³³

Investigating which interventions support bonding for families in NICU is a strategic research priority.^{34,35} Adequately powered and controlled studies are needed to investigate the impact of MT on parent-infant bonding.¹⁹ Building from the evidence base, we propose parent-

led, infant-directed singing as a resource-oriented form of MT to target early parent-infant relationship.^{36,37} Resource-oriented MT focuses on nurturing strengths, resources, and potentials of individuals, and views the therapeutic encounter as a collaboration between client(s) and therapist.³⁸

The primary aim of this study was to evaluate the effect of MT on mother-infant bonding, with secondary aims of evaluating effects on parental anxiety and maternal depression in parents of premature infants. Our hypothesis was that MT plus standard care would lead to better mother-infant bonding and improved parent mental health, compared with standard care alone.³⁶

METHODS

Design

This study is part of the Longitudinal Study of music Therapy's Effectiveness for Premature infants and their caregivers (LongSTEP) (clinicaltrials.gov NCT03564184).³¹ This part of the LongSTEP trial is designed as a 2-arm parallel, multinational, pragmatic, randomized controlled trial evaluating short-term effects of MT on parents of preterm infants at the point of discharge from NICU.³⁶

Participants and Settings

Participants were recruited from 8 NICUs in Argentina, Colombia, Israel, Norway, and Poland. Participating NICUs were equipped to provide care for complex and critically ill newborns (7 level III and 1 level IV).³⁹ The NICUs were located in countries where public support and welfare initiatives ensure high parental presence during hospitalization. Eligible sites had staff with necessary scientific and clinical expertise to conduct the

trial, including a trained music therapist. Eligible premature infants were: (1) born <35 weeks gestational age (GA), (2) likely to be hospitalized a minimum of 2 weeks from inclusion, and (3) declared by NICU staff as medically stable to start MT (typically after 26 weeks postmenstrual age [PMA]). Parent inclusion criteria were: (1) written, site-specific informed consent, (2) willingness to engage in at least 2 of 3 MT weekly sessions, (3) sufficient understanding of the respective national language(s) to answer the questionnaires and participate in MT, and (4) capacity to complete the intervention and questionnaires.³⁶ Ethics approval was granted by The Regional Committees for Medical and Health Research Ethics (2018/994/REK Nord, July 03, 2018).

Trial Procedures and Intervention

A user advisory group with parents of premature infants provided consultation before and during implementation. Intervention and outcome measures were tested in feasibility studies.^{40,41} Intervention providers were 11 music therapists with training or clinical experience with MT in NICU. Five of the 8 NICUs offered MT services before the study, while 3 implemented MT for the first time. Providers were trained in the intervention before commencing the study and received supervision from the study core team during early implementation to enhance protocol adherence. Treatment and evaluation fidelity strategies included standardized training, individual evaluation of intervention adherence, and video and audio recordings of sessions for treatment fidelity analysis that will be published as a separate article.

After providing site-specific informed consent and undergoing baseline assessments, participants were randomized to receive either MT plus standard care or standard

care alone during hospitalization. The MT consisted of parent-led, infant-directed singing supported by the music therapist. Families in the MT group were offered 3 individual MT sessions per week throughout their hospitalization, with a maximum of 27 sessions. Sessions were realized at bedside or in the family's room during skin-to-skin-time, feeding, or with the infant lying in the incubator or cot. Sessions lasted approximately 30 minutes, with time spent actively making music dependent on infant tolerance. Nonmusic session time was devoted to therapist-parent dialogue on the family's needs. For infants aged PMA ~26 to 32 weeks, MT contained cautious use of parental singing and toned voice (eg, single notes, simple melodies or short musical phrases adapted from children's songs or parent-preferred music) matched to infant state and engagement cues, such that the infant "directed" the parent's use of voice. From week 32 and onwards, this was expanded on by adding increased musical complexity and interplay.^{36,42} Accompanying instruments were used sparingly to underline the importance of parental voice. Further details about the MT intervention can be found elsewhere.^{36,37}

Standard care varied across sites but included a range of medical, nursing, developmental, and supportive care, with the exclusion of other MT approaches. All except 1 NICU included skin-to-skin time as part of standard care.

An independent data safety monitoring committee monitored safety throughout the trial.

Outcome measures

Mother-infant bonding was measured by the Postpartum Bonding Questionnaire (PBQ).^{43,44} The PBQ is a 25-item screening instrument with 4 factors (factor 1,

general impaired bonding [12 items]; factor 2, rejection and pathologic anger [7 items]; factor 3, anxiety about the infant [4 items]; and factor 4, incipient abuse [2 items]). The questionnaire consists of statements concerning the mother's feelings, experiences and attitudes toward their infant, rated on a 6-point Likert scale with responses from "always" to "never." Total score ranges from 0 to 125, with higher scores implying impaired bonding. Each factor has its own suggested cutoff point.^{43,44} This study's main outcome was PBQ total score (≥ 26 points indicate impaired bonding, ≥ 40 points indicate severe bonding disorder) with factor 1 (general impaired bonding) included as an outcome in the exploratory analysis (≥ 12 points indicate impaired bonding).⁴³ Anxiety was measured for both mothers and fathers by the Generalized Anxiety Disorder self-report tool (GAD-7) (total score ranges from 0 to 21, cutoff scores of 5, 10, and 15 indicating mild, moderate, and severe anxiety, respectively).^{45,46} Maternal depression was measured by the Edinburgh Postnatal Depression Scale (EPDS) (total score ranges from 0 to 30, >10 points indicate depression).⁴⁷ Assessments also included parent demographics and infant medical factors, including measures of common health issues for preterm infants.^{39,48-52} Discharge assessments were completed 1 to 2 days before discharge. However, assessments completed up to 2 weeks after discharge were considered valid.

Randomization and Blinding

All participants were randomized to either the control or music therapy group using a 1:1 randomization stratified by site with random block sizes of 2 or 4. Randomization occurred after informed consent and baseline assessments, using the

online randomization system www.sealedenvelope.com. The system was administered by a core team member with no involvement in clinical work. In case of multiple pregnancies, only the first-born infant was included and randomized, whereas remaining siblings received the same interventions for ethical and practical reasons. Because of the nature of the intervention, participants, providers, and data collectors were not blinded. Data analysts were kept blinded to participant allocation until data analysis was completed.

Sample Size

With an intended sample size of 250 participants (with each NICU encouraged to recruit ~50 families), the study was planned to have 80% power to detect a difference of 4 points on the PBQ (SD = 8) as the minimal clinically important difference for this study. This was based on a two-sided 5% significance level, clustered by country (ICC 0.01) and 20% attrition.³⁶

Statistical Analysis

Descriptive methods were used to characterize the sample. An intention-to-treat approach was applied by using all available data from all 213

participants as randomly assigned, regardless of the intervention actually received. Special approaches for handling missing data were deemed unnecessary because the rate of missingness was low (<10%) (Table 1). We examined effects of the intervention by testing group differences in PBQ total score at discharge by linear mixed-effects models (ANCOVA) adjusted for site because of stratified randomization (PBQ [discharge]~ group + PBQ[baseline] + 1|site). The same analyses were carried out with secondary outcomes. The quality of the models was evaluated by residual plots. Exploratory analyses of PBQ total score were conducted for prespecified subgroups (sex; GA at birth [<28 weeks, 28 to <32 weeks, and 32 to <35 weeks]; hearing status measured at discharge [normal versus abnormal]; PBQ factor 1 [impaired bonding score [≥ 12 vs <12]; parental socioeconomic status [education level]; average parental skin-to-skin care during NICU [4 or more days per week vs <4 days per week]; treatment per protocol [≥ 6 MT sessions]³⁶). Analyses were done with the statistics software R (Version 4.1.0) by using two-sided 5% significance level.

RESULTS

From August 2018 to April 2020, 213 participants were enrolled and randomly assigned to the study. Of those assigned, 208 (98%) completed assessment at discharge (Fig 1). Infant and parent demographic and medical data are similar in the two groups (Tables 1–3). Baseline scores of bonding and maternal anxiety were similar in both groups, indicating low levels of impaired bonding and mild anxiety. Paternal anxiety was slightly higher in the MT group at baseline, indicating mild anxiety. For maternal depression, both groups scored below the cutoff.

Participants assigned to MT received a mean (SD) [range] of 9.98 (5.95) [0–27] sessions. Eighty-seven of 105 (83%) received per-protocol MT (≥ 6 sessions). Mothers were present in 8.39 sessions (SD 6.01), fathers in 2.15 (SD 3.39), both parents in 2.73 (SD 3.69). Mean (SD) [range] session length was 30.8 (11.22) [0–50] minutes.

From baseline to discharge, PBQ mean total score (SD) decreased from 7.66 (9.05) to 5.51 (6.65) in the MT group, and from 7.08 (6.55) to 5.92 (6.38) in

TABLE 1 Infant Baseline Characteristics

Baseline characteristic	Standard Care		Music Therapy		All Participants	
	N	Value	N	Value	N	Value
Sex (female) ^a	108	59 (55)	105	50 (48)	213	109 (51)
Singleton pregnancy ^a	108	81 (75)	105	70 (67)	213	151 (71)
Cesarean delivery route ^a	108	89 (82)	105	83 (79)	213	172 (81)
Birth wt (g) ^b	107	1475 (445) [480, 2440]	105	1360 (421) [620, 2335]	212	1414 (422) [480, 2440]
GA at birth (wk) ^b	108	30.57 (2.77) [22.86, 34.71]	105	30.26 (2.57) [25.29, 34.29]	213	30.42 (2.67) [22.86, 34.71]
<28 wk ^a	—	17 (16)	—	21 (20)	—	38 (18)
28–32 wk ^a	—	44 (41)	—	47 (45)	—	91 (43)
32–35 wk ^a	—	47 (43)	—	37 (35)	—	84 (39)
PMA enrollment (wk) ^b	107	33.06 (1.68) [27.43, 37.29]	105	33.07 (2.21) [27.43, 44.00]	212	33.07 (1.96) [27.43, 44.00]
Apgar at 5min ^b	107	8.6 (1.27) [5, 10]	101	8.6 (1.43) [1, 10]	208	8.6 (1.35) [1, 10]
Weight at enrollment (g) ^b	104	1604 (378) [705, 2730]	105	1630 (451) [820, 3680]	209	1618 (415) [705, 3680]
Estimated severity of IVH ^{a,c}	108	—	105	—	213	—
Cranial ultrasound not indicated	—	45 (41)	—	44 (42)	—	89 (42)
None	—	52 (49)	—	50 (47)	—	102 (48)
Grade 1–2	—	10 (9)	—	9 (9)	—	19 (9)
Grade 3–4	—	1 (1)	—	2 (2)	—	3 (1)

IVH, intraventricular hemorrhage; PMA, postmenstrual age; —, not applicable.

^a N (%).

^b Mean (SD) [min, max].

^c IVH was diagnosed by cranial ultrasound and graded according to Papile et al.⁵⁰

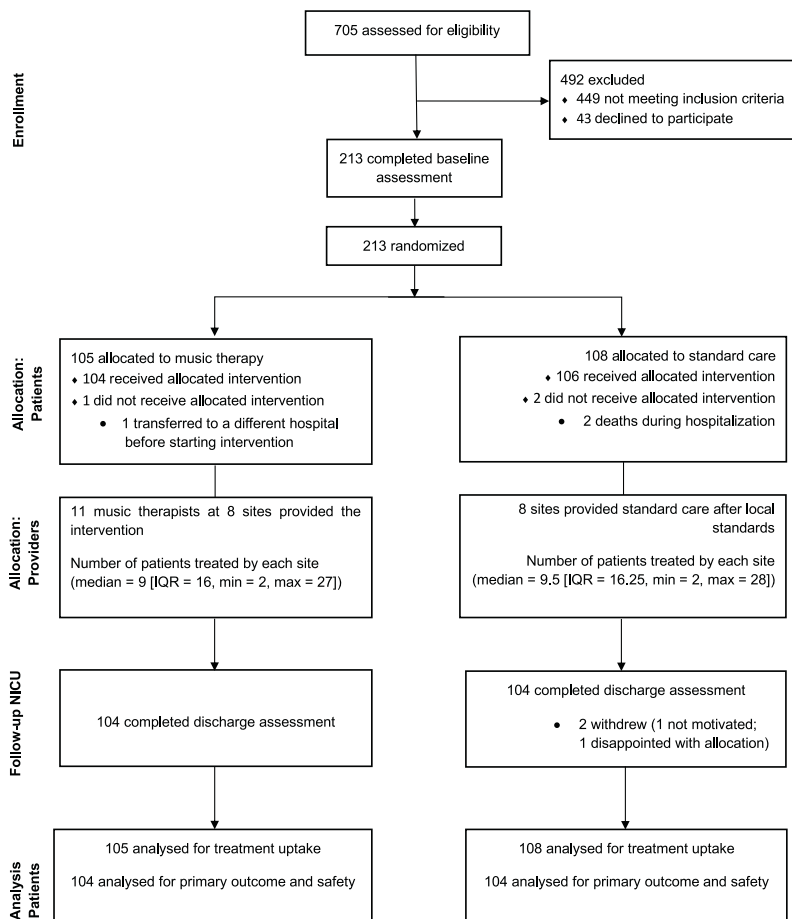


FIGURE 1
Participant flow.

the standard care (SC) group (Tables 1 and 4, Fig 2), but no significant differences between groups were found ($P = .32$). The estimated group effect (95% confidence interval [CI]) for PBQ was -0.61 (-1.82 to 0.59). Estimated group effects (95% CI) for GAD-7 was 0.14 (-0.85 to 1.12) for mothers, -0.90 (-2.03 to 0.22) for fathers, and -0.60 (-1.72 to 0.52) for EPDS. No significant between-group differences were found for secondary outcomes (Table 4, Fig 2) or predefined subgroups (Table 5). An interaction test carried out before

subgroup analysis showed no evidence of interaction.

Two serious adverse events (infant deaths) occurred during hospitalization. Both were in the standard care arm and not related to study procedures.

DISCUSSION

We investigated the effect of a MT approach using parent-led, infant-directed singing during NICU hospitalization on mother-infant

bonding and parent mental health in parents of premature infants. Our trial showed no significant effect on mother-infant bonding, parental anxiety, or maternal depression measured at discharge from NICU.

To our knowledge this is the first RCT sufficiently powered to investigate the effect of a parent-led, infant-directed singing music therapist-supported intervention on the mother-infant bond and parental mental health. Although small-scale studies have shown promising

TABLE 2 Parent Baseline Characteristics

Baseline Characteristic	Standard Care		Music Therapy		All Participants	
	N	Value	N	Value	N	Value
Maternal characteristics						
Age (y) ^a	106	32.77 (5.55) [17, 44]	104	33.11 (5.42) [21, 46]	210	32.94 (5.47) [17, 46]
Education (y) ^a	103	15.77 (2.64) [6, 21]	103	15.85 (3.35) [4, 22]	206	15.81 (3.01) [4, 22]
Usual work situation ^b	108	—	105	—	213	—
Full-time or self-employed	—	83 (77)	—	70 (67)	—	153 (72)
Other ^c	—	22 (23)	—	35 (33)	—	60 (28)
Civil status ^d	108	—	104	—	212	—
Married or living together with partner	—	96 (89)	—	98 (94)	—	194 (92)
PBQ bonding score ^a	105	—	104	—	209	—
Total score	—	7.08 (6.55) [0, 31]	—	7.66 (9.05) [0, 62]	—	7.37 (7.88) [0, 62]
Total score above cutoff (≥12) for impaired bonding	—	3 (0.3) [0, 15]	—	5 (0.5) [27, 62]	—	8 (0.4) [27, 62]
GAD-7 anxiety score ^a	105	6.10 (5.34) [0, 21]	104	6.39 (4.78) [0, 19]	209	6.24 (5.06) [0, 21]
EPDS postpartum depression score ^a	107	7.95 (5.07) [0, 20]	103	8.47 (5.17) [0, 24]	210	8.20 (5.12) [0, 24]
Paternal characteristics^d						
Age (y) ^a	101	35.37 (6.45) [22, 56]	99	36.02 (6.29) [21, 57]	200	35.69 (6.37) [21, 57]
Education (y) ^a	97	15.12 (3.21) [6, 22]	96	15.02 (3.77) [3, 25]	193	15.07 (3.49) [3, 25]
Usual work situation ^b	101	—	99	—	200	—
Full-time or self-employed	—	96 (95)	—	90 (91)	—	186 (93)
Other ^c	—	12 (12)	—	15 (15)	—	27 (14)
GAD-7 anxiety score ^a	98	4.71 (4.35) [0, 21]	98	6.30 (5.73) [0, 21]	196	5.51 (5.13) [0, 21]

GAD-7, General Anxiety Disorder-7; EPDS, Edinburgh Postpartum Depression Scale; PBQ, Postpartum Bonding Questionnaire; —, not applicable.

^a Mean (SD) [min, max].

^b N (%).

^c Other includes part-time, homemaker/stay-at-home parent, student, unemployed because of ill health or a disability.

^d Same-sex parents were invited to participate in the study but none were enrolled.

results on the impact of MT on bonding^{20–24} our study does not confirm these findings. Our failure to detect a definitive effect on bonding or parental well-being may be related to an insufficient number of sessions. Haslbeck et al noted a dose-dependent effect of creative MT on brain development in preterm infants, with no plateauing of the effect.⁵³ Their median sessions (14.87) was nearly double ours (8), which may suggest that a longer intervention period could have a positive impact on outcomes.

We used the PBQ, a screening questionnaire designed to identify disorders in mother-infant bonding.^{43,44} In spite of receiving the strongest psychometric evaluation rating in a recent systematic review,⁵⁴ the PBQ may lack sensitivity to detect changes in mothers reporting a relatively healthy bond with their premature infant. In our study, very few

mothers demonstrated impaired bonding at baseline, and although bonding scores were slightly improved at discharge from hospital, no between-groups differences were found. The strong focus on pathology within the items of the PBQ could have created a flooring effect, making it challenging to detect differences between mothers in the subtler and health-oriented aspects of bonding.

We based our hypothesis on research suggesting a higher risk of impaired bonding in mothers of premature infants.^{2,3,55,56} In our study, a very small number of mothers from a range of countries and socioeconomic statuses reported impaired bonding, which challenges this assumption. Our findings are consistent with research suggesting that mothers of premature infants may demonstrate equal, or better, quality of mother-infant interaction and attachment as

compared with mothers of full-term infants.⁸ Borghini et al argue that parental emotional arousal during hospital stay may facilitate parental involvement, which in turn might strengthen the parent-infant bond.⁵⁷ The NICUs in our study are located in countries where parents are expected to be continuously present and partake in the care of their child. Ninety-nine of 103 (96%) SC parents, and 97 of 105 (92%) MT parents, reported being present almost daily. Hence, it is possible that the parents in our study had a lower risk of impaired bonding compared with parents in other settings who experience unwanted separation.

The majority of the infants in our study were classified as very low birth weight and were hospitalized on average 7 weeks. During NICU hospitalization, instability in the baby's medical needs might be more of a concern to parents than

TABLE 3 Infant Clinical Characteristics at Discharge

Discharge characteristic	Standard Care		Music Therapy		All Participants	
	N	Value (%)	N	Value (%)	N	Value (%)
Weight at discharge, g, mean (SD) [range]	101	2464 (471) [1646, 4140]	—	2443 (464) [1640, 4320]	205	2485 (479) [1640, 4320]
Nutrition during admission, n (%)	103	—	105	—	208	—
>50% mother's breast milk	—	58 (56)	—	66 (63)	—	124 (60)
<50 mother's breast milk	—	29 (28)	—	24 (23)	—	53 (25)
Donor breast milk +/- infant formula	—	3 (3)	—	2 (2)	—	5 (2)
>90% infant formula	—	13 (13)	—	13 (12)	—	26 (13)
BPD, n (%)	104	—	105	—	209	—
None	—	58 (56)	—	55 (52)	—	113 (54)
Mild	—	30 (29)	—	27 (26)	—	57 (27)
Moderate	—	11 (11)	—	11 (10)	—	22 (10)
Severe	—	5 (5)	—	12 (11)	—	17 (8)
Discharged with O ₂ supply, n (%)	104	—	105	—	209	—
Yes	—	32 (31)	—	31 (30)	—	63 (30)
No	—	72 (69)	—	74 (70)	—	146 (70)
Systemic infection during hospitalization, n (%)	104	—	105	—	209	—
Yes	—	19 (18)	—	28 (27)	—	47 (22)
No	—	85 (82)	—	77 (73)	—	162 (78)
NEC, n (%)	103	—	105	—	208	—
No	—	99 (96)	—	104 (99)	—	203 (98)
Suspected	—	2 (2)	—	0 (0)	—	2 (1)
Confirmed	—	2 (2)	—	1 (1)	—	3 (1)
ROP, n (%)	96	—	100	—	196	—
None	—	84 (88)	—	78 (78)	—	162 (83)
Grade 1-2	—	9 (9)	—	16 (16)	—	25 (13)
Grade 3	—	3 (3)	—	6 (6)	—	9 (4)
Estimated severity of IVH, n (%)	108	—	105	—	213	—
Cranial ultrasound not indicated	—	38 (35)	—	34 (32)	—	72 (34)
None	—	59 (55)	—	55 (52)	—	114 (53)
Grade 1-2	—	10 (9)	—	14 (13)	—	24 (11)
Grade 3-4	—	1 (1)	—	2 (2)	—	3 (1)
Hydrocephalus posthemorrhage, n (%)	103	—	105	—	208	—
Yes	—	0 (0)	—	1 (1)	—	1 (1)
No	—	103 (100)	—	104 (99)	—	207 (99)
PVL, n (%)	102	—	101	—	203	—
None	—	99 (97)	—	95 (94)	—	194 (96)
Grade 1-2	—	3 (3)	—	3 (3)	—	6 (3)
Grade 3-4	—	0 (0)	—	3 (3)	—	3 (1)
Hearing test status, n (%)	71	—	77	—	148	—
Passed	—	70 (99)	—	69 (90)	—	139 (94)
Not passed	—	1 (1)	—	8 (10)	—	9 (6)

BPD was diagnosed according to the criteria of Bancalari et al.⁴⁸ Systemic infection was considered if positive blood cultures coincided with clinical signs suggesting blood infection. NEC was determined by clinical and radiologic criteria of Bell et al.⁴⁹ Only definite NEC (Bell stages II to III) was included. ROP was graded according to the international classification by the Committee for the Classification of Retinopathy of Prematurity, and was recorded as the most severe stage in either eye.⁶⁵ IVH was diagnosed by cranial ultrasound and graded according to Papile et al.⁵⁰ Hydrocephalus was diagnosed in accordance with Volpe.⁵¹ PVL was diagnosed by ultrasound after 28 d of life, with grading from 1 to 4 in accordance with De Vries and Rennie.⁵² BPD, bronchopulmonary dysplasia; IVH, intraventricular hemorrhage; NEC, necrotizing enterocolitis; PVL, periventricular leukomalacia; ROP, Retinopathy of prematurity; —, not applicable.

immediate and longer-term relationship and bonding. Interventions aiming to support parent-infant bonding might be more effective after discharge from hospital when infants are typically more stable and families have returned home.^{58,59} A qualitative study of a LongSTEP cohort supports this possibility, where parents experienced that

postdischarge MT led to positive change in their parent-infant relationships (Epstein et al, unpublished data, December 2021).

We found no significant effects of our intervention on maternal depression or parental anxiety. Our findings related to anxiety contrast with a meta-analysis reporting a reduction in maternal anxiety after

MT.¹⁹ The timepoint of assessment could explain this discrepancy. We assessed anxiety shortly before discharge, a time when anxiety and stress might peak as parents prepare for transitioning from hospital to home and taking full responsibility for their child's care.^{58,59} Kehl et al found that MT significantly reduced parental anxiety 2 weeks after birth and

TABLE 4 Observed Values at Discharge, Changes From Baseline, and LME Results

	Observed Values				Change From Baseline			Group Effect (LME) ^b		
	SC		MT		SC	MT	Difference ^a	Coefficient (95% CI)	df	P
	N	M (SD)	N	M (SD)	M	M	M (95% CI)			
PBQ total score	101	5.92 (6.38)	105	5.51 (6.65)	-1.29	-2.15	-0.85 (-2.32 to 0.61)	-0.61 (-1.82 to 0.59)	192	.318
GAD-7 mother	99	4.60 (4.67)	105	4.74 (3.78)	-1.69	-1.66	0.03 (-1.23 to 1.29)	0.14 (-0.85 to 1.12)	191	.787
GAD-7 father	92	4.54 (4.58)	95	4.24 (4.18)	-0.24	-2.03	-1.80 (-3.31 to 0.29)	-0.90 (-2.03 to 0.22)	173	.115
EPDS	101	6.2 (5.21)	104	6.02 (4.45)	-1.59	-2.47	-0.88 (-2.18 to 0.42)	-0.60 (-1.72 to 0.52)	194	.290

df, degrees of freedom; M, mean.

^a From *t* test, assuming equal variance.

^b Based on a linear mixed-effects model including variables mother-infant bonding, parent anxiety, and maternal depression with site as random effect.

halfway through NICU stay, but not at 2 weeks after discharge.²³ Although an RCT including mothers with moderate-severe postpartum depression found singing to give faster improvement in symptoms,³¹ no large-scale RCTs have investigated the effect of MT on depression in mothers of premature infants. Our findings of no effects on maternal depression are, however, in line with pilot studies.^{23,24} The complex caretaking role parents take

might compromise parents' capacity to manage their own crisis of experiencing preterm birth.⁶⁰ Although the study protocol includes elements of psychotherapeutic support for parents in alignment with trauma-preventive models,³⁶ the training and intervention protocol³⁷ might not have addressed this sufficiently enough for therapists to implement it in a uniform way, or therapists may have struggled to adequately meet parental needs

within the sessions. Additionally, the emphasis on parent-led singing might have placed too much responsibility on parents in a critical phase. However, qualitative results from feasibility testing of LongSTEP contrast such a notion.⁴⁰ Parents in the feasibility study reported that MT helped them get to know their infants, and that resources they developed during MT were transferrable to parent-infant interactions outside of MT.¹⁹ Further

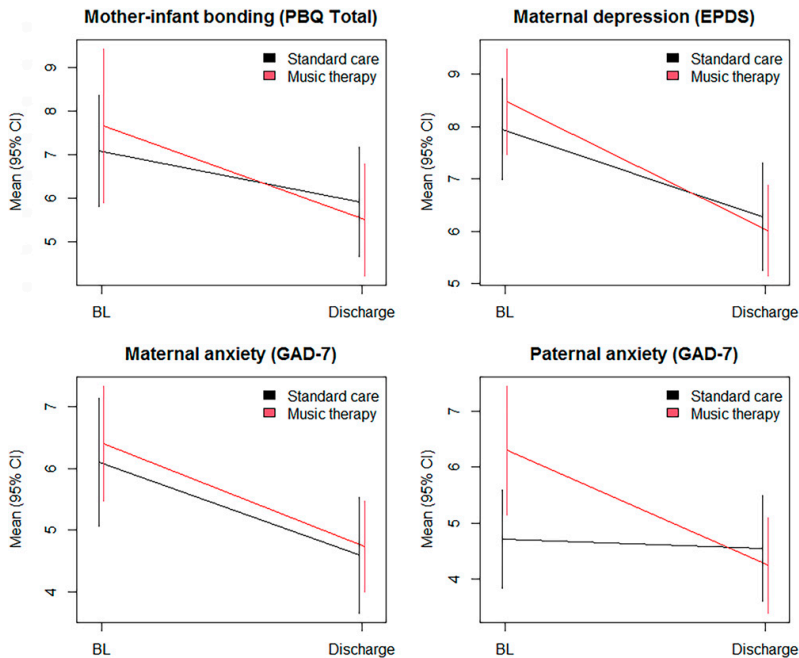


FIGURE 2 Results at discharge.

TABLE 5 LME Results for Subgroup Analysis

	Group Effect (LME) ^a		
	Coefficient (95% CI)	df	P
Gender	-0.60 (-1.81 to 0.60)	190	.33
GA at birth	-0.59 (-1.81 to 0.62)	190	.34
Hearing status at DC	-0.33 (-1.85 to 1.19)	132	.67
Maternal education level	-0.67 (-1.86 to 0.53)	186	.27
Paternal education level	-0.42 (-1.64 to 0.81)	173	.50
Hours of skin-to-skin care	-0.66 (-1.87 to 0.55)	189	.29
Indications of impaired bonding at baseline	-2.97 (-10.45 to 5.51)	2	.52

DC, discharge; df, degrees of freedom; GA, gestational age.

^aThis table shows the interaction of each subgroup with the intervention in a linear mixed-effects model including the variable mother-infant bonding, with site as random effect.

examination of the impact of MT on the parent-infant relationship and parental mental health is indicated. Since the LongSTEP study's MT approach is resource-oriented, future studies applying this or similar approaches might benefit from choosing outcomes that are less focused on pathology, such as quality of life, parent self-efficacy, self-confidence, and empowerment.

Strengths and Limitations

Low attrition in our study indicates a high acceptability of intervention and study procedures among participants in different cultural contexts. Whereas research often focuses exclusively on mothers, we included both parents in the intervention and study outcomes.⁶¹ Although we aimed to engage both parents actively in MT, our primary outcome was limited to mothers to promote homogeneity for reasons of analysis. Limitations include the reliance on self-report assessments, which may contribute to social desirability bias, and inclusion of outcome measures that may not have been sufficiently sensitive to circumstances of the included mothers. The chosen outcome measures may not have fit well enough with the focus of the intervention. In mothers of full-term infants, singing has been shown to positively impact arousal, affect, and stress hormones.^{30,32} Investigating

biological markers of anxiety instead of self-reported symptoms could augment our understandings of potential impacts on anxiety. Although music therapists in the study were trained on the intervention, they differed regarding previous clinical experience and expertise. Variation in intervention implementation might have affected outcomes, although we noted no specific effect for site. Variations in adherence to protocol will be further investigated in treatment fidelity analyses. Conclusions of this study are applicable to NICUs where consistent parental presence is feasible.

CONCLUSIONS

In this large-scale RCT we found no significant effects of music therapist-supported, parent-led, infant-directed singing on mother-infant bonding, parental anxiety, or maternal depression, although scores on all outcomes improved in both groups. Additional examination of the impact of MT interventions on parent-infant mutual outcomes and parental well-being would be valuable. We also suggest investigating the effect on outcomes that are more salutogenically focused, for example quality of life, parent self-efficacy, self-confidence, and empowerment.

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ABBREVIATIONS

- CI: confidence interval
- DSMC: Data Safety Monitoring Committee
- EPDS: Edinburgh Postpartum Depression Scale
- GA: gestational age
- GAD-7: General Anxiety Disorder-7
- IVH: intraventricular hemorrhage
- MT: music therapy
- NICU: neonatal intensive care unit
- PBQ: Postpartum Bonding Questionnaire
- PMA: post-menstrual age
- RCT: randomized controlled trial
- SC: standard care

Mangersnes and Mrs Røed coordinated data collection, and critically reviewed and revised the manuscript; and all authors approved the final manuscript as submitted and agree to be accountable for all aspects of the work.

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Article III

Treatment fidelity in a pragmatic clinical trial of music therapy for premature infants and their parents: The LongSTEP trial

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Abstract

Background

Treatment fidelity (TF) refers to methodological strategies used to monitor and enhance reliability and validity of interventions. We evaluated TF in a pragmatic RCT of music therapy (MT) for premature infants and their parents.

Methods

213 families from seven neonatal intensive care units (NICU) were randomised to receive standard care, or standard care plus MT during hospitalization, and/or during a six-month period post-discharge. Eleven music therapists delivered the intervention. Audio and video recordings from sessions representing approximately 10% of each therapists' participants, were evaluated by two external raters and the corresponding therapist using TF questionnaires designed for the study (treatment delivery (TD)). Parents evaluated their experience with MT at the 6-month assessment with a corresponding questionnaire (treatment receipt (TR)). All items as well as composite scores (mean scores across items) were Likert scales from 0 (completely disagree) to 6 (completely agree). A threshold for satisfactory TF scores (≥ 4) was used in additional analysis of dichotomised items.

Results

Internal consistency evaluated with Cronbach's alpha was good for all TF questionnaires ($\alpha \geq 0.70$), except external rater NICU questionnaire where it was slightly lower ($\alpha 0.66$). Interrater reliability measured by Intraclass correlation coefficient (ICC) was moderate (NICU 0.43 (CI 0.27, 0.58), post-discharge 0.57 (CI 0.39, 0.73)). Gwet's AC for the dichotomised items varied between 0.32 (CI 0.10, 0.54) and 0.72 (CI 0.55, 0.89). 72 NICU and 40 follow-up sessions with 39 participants were evaluated. Therapists' mean (SD) TD composite score was 4.88 (0.92) in NICU phase, and 4.95 (1.05) in post-discharge phase. TR was evaluated by 138 parents. Mean (SD) score across intervention conditions was 5.66 (0.50).

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Conclusions

TF questionnaires developed to assess MT in neonatal care showed good internal consistency and moderate interrater reliability. TF scores indicated that therapists across countries were successfully implemented MT in accordance with the protocol. Although the intervention did not lead to significant changes in study outcomes, high treatment receipt scores indicate that parents received the intervention as intended. Future research in this area should aim to improve the interrater reliability of TF measures by additional training of raters and improved operational definitions of items.

Clinical trials registry name: ClinicalTrials.gov Identifier NCT03564184

Keywords: research methods, fidelity, treatment fidelity, randomised controlled trial, pragmatic trial, multinational trial, non-pharmacological interventions, music therapy, premature infant, parent-infant bonding

Background

Fidelity refers to the degree to which the delivery of an intervention adheres to the protocol or program developed. During the last decades, assessment of fidelity has had an increasing significance for evaluations, treatment effectiveness research, and service administration.¹ In multi-site studies, fidelity criteria are essential to ensure that interventions are conducted uniformly across sites, since this affects the reliability and validity of the intervention and conclusions about intervention effectiveness.^{2,3} A non-significant result might be the result of an ineffective intervention or could be due to interventionists not adhering to the protocol.¹

In clinical trials reporting on treatment interventions, the term treatment fidelity (TF) denotes the fidelity or integrity of an intervention.² Different methodological strategies can be applied to assess TF. Bellg et al. propose a five-component model for TF assessment in behavioural studies, including the design of the study, training of providers, treatment delivery, treatment receipt, and enactment of treatment skills.² These components are mutually exclusive and failing to attend to any of the components could compromise the internal validity of the study.³ The first two components are considered in the development and preparation of a study, whereas the three last components can be assessed during and after implementation of an intervention. Training of providers is a central part of enhancing TF with behavioural interventions, as it often requires learning new skills that might differ from clinicians' existing training and experience.² Standardized training and monitoring and maintaining of provider skills are some of the recommendations of Bellg et al.² Treatment delivery (TD) refers to the extent the provider of treatment has adhered to the guidelines for the intervention and delivered treatment as intended. Treatment receipt (TR) refers to the degree to which the participant understands the treatment and their ability to perform protocol-related skills and strategies during the intervention.² Assessment of treatment enactment (TE) requires processes to monitor and improve the ability of patients to perform treatment-related strategies and skills in their daily lives between sessions or after the intervention period. In this article we report on TD and TR assessed during the implementation of the Longitudinal Study of music Therapy's Effectiveness for Premature infants and their caregivers – LongSTEP (ClinicalTrials.gov identifier NCT03564184).⁴

The intervention assessed in LongSTEP was a music therapy (MT) approach carried out in the context of neonatal intensive care units (NICUs) in different countries across Europe, Middle East, and South America. With MT we refer to “the informed use of music, facilitated by a trained music therapist within a therapeutic relationship, whereby engagement in musical processes serves as a resource to promote health”.⁵ MT was first introduced to NICUs in the early 1990s^{6, 7} with early research demonstrating positive effects on physiological and behavioural outcomes of premature infants, such as respiratory rate, oxygen saturation, heart rate, weight gain, and feeding patterns.⁸⁻¹¹ Within the last decade, MT in NICU has evolved in line with principles of family-centred care, supporting both infant development and parental well-being, including facilitating early parent-infant relationship through empowering parents in their parental roles and understanding of their infant.¹²⁻²⁴ We designed a pragmatic randomized controlled trial (RCT)²⁵ to evaluate longer-term parent-infant mutual outcomes, an identified gap in the knowledge base.²⁶ Since MT in NICU has predominately been conducted within the context of US health care,^{27, 28} we wished to contribute to knowledge development through collaboration with research partners from a broader range of cultural contexts where MT had not yet been systematically implemented in neonatal care, including Argentina, Colombia, Israel, Norway, and Poland. Given the international and multi-cultural nature of our trial, we were particularly interested in evaluating TF. Furthermore, to our knowledge no clinical trials of MT in NICU have systematically evaluated TF. In the context of the LongSTEP-trial, we developed and implemented strategies to enhance and assess TF, drawing upon experiences and examples from other MT trials with different populations.²⁹⁻³¹

The overall aim of this article is to report on TF in the LongSTEP trial, through evaluating the reliability of TF questionnaires developed for the trial, and the extent to which MT in the two intervention phases was a uniform intervention across therapists at the different research sites. Consistent with a pragmatic approach²⁵, intervention delivery included a balance of guiding principles for the intervention, combined with flexibility and openness to the clinicians’ interpretations and adaptations of the intervention to better fit usual care in their cultural context. Our research questions were: 1) What is the internal consistency and interrater reliability of the TF questionnaires? 2) To what extent did the music therapists adhere to the essential

elements of the intervention protocol? 3) To what extent did parents perceive MT to be in line with essential elements of the protocol?

Methods

Study design and participants

The participants were families with preterm infants born before 35 weeks gestational age (GA), likely to be hospitalized at least two weeks from inclusion, and declared by NICU staff as medically stable to start MT (typically after 26 weeks postmenstrual age).⁴ The included NICUs were level III and IV³² units located in Argentina, Colombia, Israel, Norway and Poland, all countries with high levels of parent presence in the NICU. Families were randomised to receive standard care, or standard care plus MT during NICU hospitalization, and/or during a six-month follow-up period post-discharge. Participants in the control group were required not to receive any music-related interventions during the intervention period, and therapists were instructed to do MT sessions in individual patient rooms, if possible, to reduce contamination.

Intervention

Our approach builds on previous models and approaches to MT in NICU^{10, 17, 27, 33-36}, and theories such as resource-oriented MT^{37, 38}, the mutual regulation model^{39, 40}, family-centred principles and developmental care models.^{41, 42} Further details on the theoretical foundation can be found elsewhere.⁴³ Seven elements are considered essential for our approach and should be present in each session regardless of infant post-menstrual age, or the phase in which MT is provided.⁴⁴ These were: 1) Observation and dialogue on infant's needs prior to and during MT sessions, 2) Dialogue with parents on their state and needs prior to sessions, 3) Voice serves as the main instrument, 4) Parental voice serves as the most prominent musical voice, 5) Music therapist provides opportunities for parents to actively participate, 6) Music is modified to infant cues and responses, and 7) Parents' culture and musical preferences and abilities are integrated into sessions (See Additional file 1). Per protocol MT during NICU hospitalization comprised three weekly 20–30-minute sessions throughout hospitalization of minimum two weeks (minimum 6, and maximum 27 sessions). Number and average length of session were tracked. Families randomised to MT during follow-up participated in seven monthly 45–60-minute

sessions over a six-month period. Follow-up sessions were carried out at home, in the hospital or at other health facilities. MT was adapted to the two phases in accordance with the intervention protocol. ⁴³

Treatment delivery

Training of providers. To monitor provider skills and delivery, all therapists submitted recordings of themselves carrying out sessions early in the implementation phase so that the core team could assess need for additional training or support. The recordings used for this quality control purpose were excluded from the TF analysis. The music therapists were also encouraged to use a tracking form to increase awareness of aims, techniques, and progress across the course of the MT sessions. Supervision was another strategy to support successful implementation and protocol adherence. All therapists participated in online group supervision at least twice, and online individual supervision at least once during the implementation period. The aim was to increase therapists' self-awareness, and to provide a space where challenges could be discussed openly, strategies could be shared, and experiences celebrated with peer support. These sessions also helped highlight contextual and culturally specific aspects of the study. Eleven music therapists were trained to deliver the MT intervention in our study. All eleven were female, masters-prepared music therapists, of which two were in the terminal stage of their degree. Six of the music therapists in the study participated in the in-person one-day training consisting of lectures and practical exercises based on the intervention protocol during the study's kick-off meeting. Five music therapists joined after study initiation and received online training sessions with the same content.

Treatment fidelity questionnaires. Five TF questionnaires were developed along with the theoretical foundation and intervention protocol ⁴³, translating the intervention's essential elements into items of behaviour we predicted would be audible or observable in the sessions (See Additional file 2). One element was not observable for all raters because of reliance on insight into the MT process and was therefore only included in the music therapist and parent questionnaires. Another element was only observable with video recordings and was included in the post-discharge tools only. The TF questionnaires were designed with Likert-scaled items (each 0-6), with anchors "I completely disagree" to "I completely agree". A threshold of ≥ 4 per item was

decided *a priori* as a satisfactory level of TF, with higher numbers indicating better therapist adherence and parent perceived receipt of the item. Four TD questionnaires were created in accordance with: a) the phase within which MT was delivered, b) which elements could feasibly be distinguished in audio versus video recordings of sessions, and c) who was completing the rating. The questionnaires were: *Treatment Delivery Questionnaire for Music Therapist Self-ratings, NICU* (seven items) and *post-discharge version* (eight items), and *Treatment Delivery Questionnaire for External Raters, NICU* (six items) and *post-discharge version* (seven items) (See Additional file 3). Each therapist's sessions were reviewed by the corresponding therapist and two external raters who understood the language spoken and were educated in MT or psychology. Raters were provided with descriptions of behaviours related to each item to look for and were instructed to listen to or watch the recorded session once in its entirety, while filling out the questionnaire. The fifth questionnaire developed was the *Treatment Receipt Questionnaire* (nine items), where parents who received MT in one or both phases were instructed to think back on their experiences with MT as a whole and evaluate the degree to which they perceived the guiding principles of the MT intervention (See Additional file 4). The TD questionnaires were pilot tested by two members of the study core team and the TR questionnaire was discussed with the user advisory group who suggested simplifying the language. Changes were made accordingly before implementation.

Data collection

Treatment delivery analysis was based on recordings from approximately 10% of each therapist's participants, evaluated by the corresponding music therapist and two external raters per therapist. Sessions during NICU stay were audio recorded, and during follow-up were video recorded for all sites, except one that could not obtain permission to video record. Music therapists were responsible for audio/video recording their own sessions. Video instructions were to aim for a frame that showed both parent(s) and infant. Participants for TD analysis were randomly selected using www.randomresult.com with the "Pick items" function. If the material from the selected participant was not possible to use (e.g., missing video/audio, participant dropped out of study) a new participant was drawn randomly. We strived to evaluate sessions that were distributed over time, avoiding the first and last session as first sessions were used to explain and demonstrate aspects of the intervention; and last sessions to sum

up content from the course of MT and dialogue about continued, independent use of music. Hence, we expected that first and last sessions would include minimal levels of interaction between parents and infants or singing. When participants received per protocol MT during the NICU phase, we analysed recordings of session three, five and seven. When participants had fewer than seven sessions, we analysed session two, four and six. If a participant received more than 10 sessions, the 11th was added with the intention of investigating drifting, however we did not have a sufficient sample for this analysis. Recordings of two sessions per participant were evaluated from the follow-up phase, either three and six, three and five, or four and six, based on useable video. The variation of session numbers in the final data material was due to missing recordings. Parent self-report ratings at the six-month assessment served as data for analysis of TR.

Analysis

Descriptive methods were applied to characterize the two participant samples for TD and TR. Categorical data were analysed with frequency and percentage, and numerical data with mean, standard deviation, and range due to normally distributed data. The internal consistency of each TF questionnaire was evaluated with Cronbach's alpha ⁴⁴ with alphas of ≥ 0.70 indicating good internal consistency. ⁴⁵ Interrater reliability (IRR) between music therapists and external raters was evaluated per item and composite score with intraclass correlation coefficient (ICC) with a two-way model, single measurement, and absolute agreement. Additionally, we calculated agreement of categorical items dichotomised to above/below threshold of satisfactory adherence (≥ 4). Because of high prevalence of single item alternatives for some items we used Gwet's AC ⁴⁶ instead of kappa, due to the known weaknesses of kappa in this case. ⁴⁷ Mean TD scores per item, therapist, and composite score (mean scores across external ratings and music therapist self-ratings) were calculated from ratings from the two intervention phases (NICU and post-discharge). Mean TR scores per item and composite score per participant, and composite score across intervention conditions (MT in NICU, post-discharge or both) were calculated from parent ratings. For these analyses it was not necessary to consider who was the first and who the second external rater. Statistical analyses were done with software R version 4.1.0 ⁴⁸ and graphics with Matlab 2021b. ⁴⁹

Results

In total, 72 NICU and 40 post-discharge sessions of 39 unique participants (Table 1) were rated by 10 music therapists and 13 external raters for TD assessment. For post-discharge sessions, we also reviewed video characteristics of who was present in sessions and their visibility in the recordings for data quality purposes. Mothers were present in all sessions, fathers in 37.5%, siblings in 41% and grandmothers in 15% of the sessions. Mothers were fully visible in 81% of the videos, fathers in 83% of the sessions they attended, while the infants were fully visible in only 53% of the videos. The same applied to music therapists who were visible in 53% of the videos. Treatment receipt was evaluated by 135 parents at the six-month assessment (Table 2).

Table 1 **Sample characteristics treatment delivery**

	NICU MT		PD MT		MT both phases	
	N	Value	N	Value	N	Value
Infant female sex, No. (%)	27	10 (37%)	30	12 (40%)	18	7 (39%)
Infant birth weight, mean grams, (SD)	27	1355 (405)	30	1365 (401)	18	1314 (358)
Infant GA at birth, mean weeks, (SD)	27	30.1(2.5)	30	30.0 (2.6)	18	29.6 (2.4)
Mother age, mean years (SD)	26	31.8 (4.0)	29	32.0 (5.2)	17	32.3 (3.9)
Mother education, mean years (SD)	27	15.8 (3.8)	30	16.1 (3.1)	18	16.4 (2.7)
Mother usual work situation, No. (%)	27	-	30	-	18	-
Full-time- or self-employed		19 (70%)		21 (70%)		12 (67%)
Other ¹		8 (30%)		9 (30%)		6 (33%)
Mother civil status, No. (%)	26	-	30	-	18	-
Married		19 (73%)		20 (67%)		13 (72%)
Living together but not married		7 (27%)		9 (30%)		5 (28%)
Other		-		1 (3%)		-
Father age, mean years (SD)	27	34.5 (4.4)	30	34.2 (5.3)	18	34.2 (5.0)
Father education, mean years (SD)	26	15.1 (3.6)	29	15.0 (3.4)	17	15.6 (3.2)
Father usual work situation, No. (%)	27	-	30	-	18	-
Full-time- or self-employed		26 (96%)		29 (97%)		17 (94%)
Other ¹		1 (4%)		1 (3%)		1 (6%)

Abbreviations: NICU, neonatal intensive care unit; MT, music therapy; PD, post-discharge; SD, standard deviation; GA, gestational age. ¹ Other includes part-time, homemaker/stay-at-home parent, student, unemployed and seeking work, unemployed due to ill health or a disability. Note: One participant was removed from sample because of language challenges between therapist and family.

Table 2 Sample characteristics treatment receipt

	NICU MT		PD MT		MT both phases	
	N	Value	N	Value	N	Value
Infant female sex, No. (%)	49	22 (45%)	41	23 (56%)	45	21 (47%)
Infant birth weight, mean grams, (SD)	49	1344 (440)	41	1423 (430)	45	1415 (414)
Infant GA at birth, mean weeks, (SD)	49	30.2 (2.7)	41	32.85 (5.47)	45	30.34 (2.57)
Mother age, mean years (SD)	49	31.9 (5.6)	40	33.07 (2.21)	44	34.07 (5.20)
Mother education, mean years (SD)	48	15.2 (3.6)	39	16.03 (2.97)	44	16.36 (2.80)
Mother usual work situation, No. (%)	49	-	41	-	45	-
Full-time- or self-employed		30 (61%)		33 (80.5%)		33 (73%)
Other ¹		19 (39%)		8 (19.5%)		12 (27%)
Mother civil status, No. (%)	48	-	41	-	45	-
Single		3 (6%)		4 (10%)		2 (4%)
Married		33 (69%)		25 (61%)		32 (71%)
Living together but not married		12 (24%)		10 (24%)		11 (24%)
Other		-		2 (5%)		-
Father age, mean years (SD)	47	35.1 (6.8)	39	35.4 (5.7)	42	36.4 (5.3)
Father education, mean years (SD)	45	14.3 (4.0)	38	15.6 (3.4)	41	15.7 (7.2)
Father usual work situation, No. (%)	47	-	39	-	42	-
Full-time- or self-employed		44 (94%)		38 (97%)		39 (93%)
Other ¹		3 (6%)		1 (3%)		3 (7%)

Abbreviations: NICU, neonatal intensive care unit; MT, music therapy; PD, post-discharge; SD, standard deviation; GA, gestational age. ¹Other includes part-time, homemaker/stay-at-home parent, student, unemployed and seeking work, unemployed due to ill health or a disability. Note: One participant was removed from sample because of language challenges between therapist and family.

Reliability of treatment fidelity questionnaires

We conducted reliability analyses of the questionnaires, assessing internal consistency and interrater reliability. Internal consistency of the scales was measured with Cronbach's alpha indicating good internal consistency (≥ 0.70) for all except NICU external rater questionnaire which scored slightly lower (α (CI) 0.66 (0.60, 0.73) Table 3)). For all questionnaires, most items appeared to be worthy of retention resulting in a decrease in or no change in alpha if removed (See Additional file 5). Based on these alpha calculations, it was decided to keep all items in all scales and to calculate composite scores as planned.

Table 3 Cronbach's alpha for treatment fidelity questionnaires

	Treatment delivery		Treatment receipt
	NICU	Post-discharge	Both phases
Music therapist self-rater questionnaire	0.75 (0.67, 0.82)	0.87 (0.84, 0.91)	-
External rater questionnaire	0.66 (0.60, 0.73)	0.80 (0.76, 0.84)	-
Parent questionnaire	-	-	0.79 (0.74, 0.83)

Note: All values are raw α (CI). Abbreviations: CI, confidence interval; NICU, neonatal intensive care unit

Interrater reliability (IRR) of the TD composite scores across music therapist self-rater and external rater versions was moderate with ICC 0.43 (CI 0.27, 0.58) (Figure 1)). Gwet's AC for the dichotomised items varied between 0.32 (CI 0.10, 0.54) and 0.72 (CI 0.55, 0.89) (Figure 1)).

Treatment delivery and treatment receipt

Mean composite TD score across raters for NICU sessions was 4.88 (0.92) and 4.95 (1.05) for post-discharge sessions, scoring between one and two Likert points away from "I completely agree" (Figure 2). Mean TD composite score per therapist ranged from 3.17 to 5.46 for NICU phase, and 3.51 to 5.65 post-discharge (Figure 3).

Treatment receipt mean scores were very high with NICU group mean (SD) of 5.66 (0.50), post-discharge group 5.65 (0.71), and 5.71 (0.40) for the group who received MT in both phases. Mean (SD) TR composite score across groups was 5.68 (0.53) (Figure 4)).

Figure 1 Interrater reliability of treatment delivery questionnaires

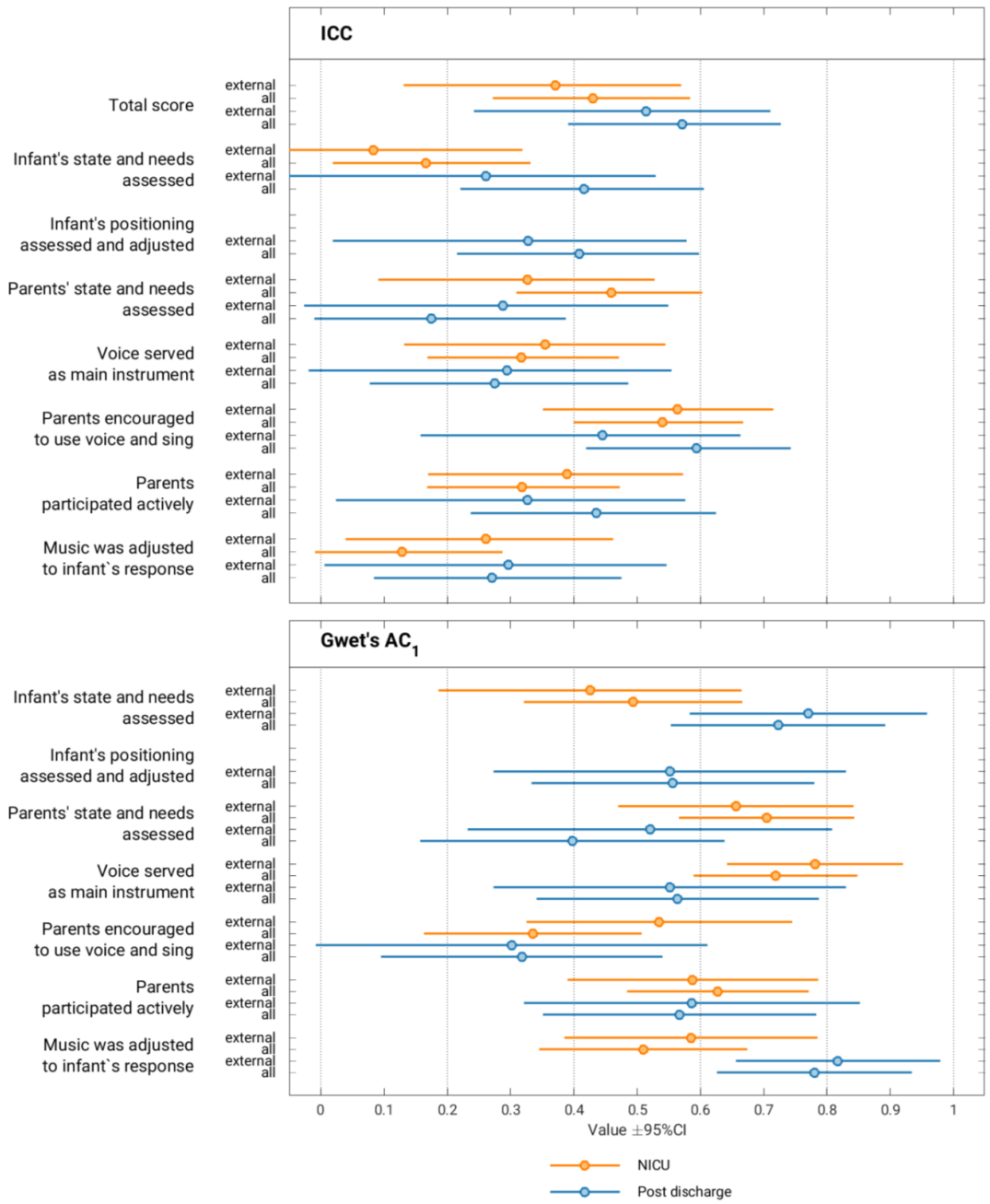


Figure 2 Treatment delivery scores per item

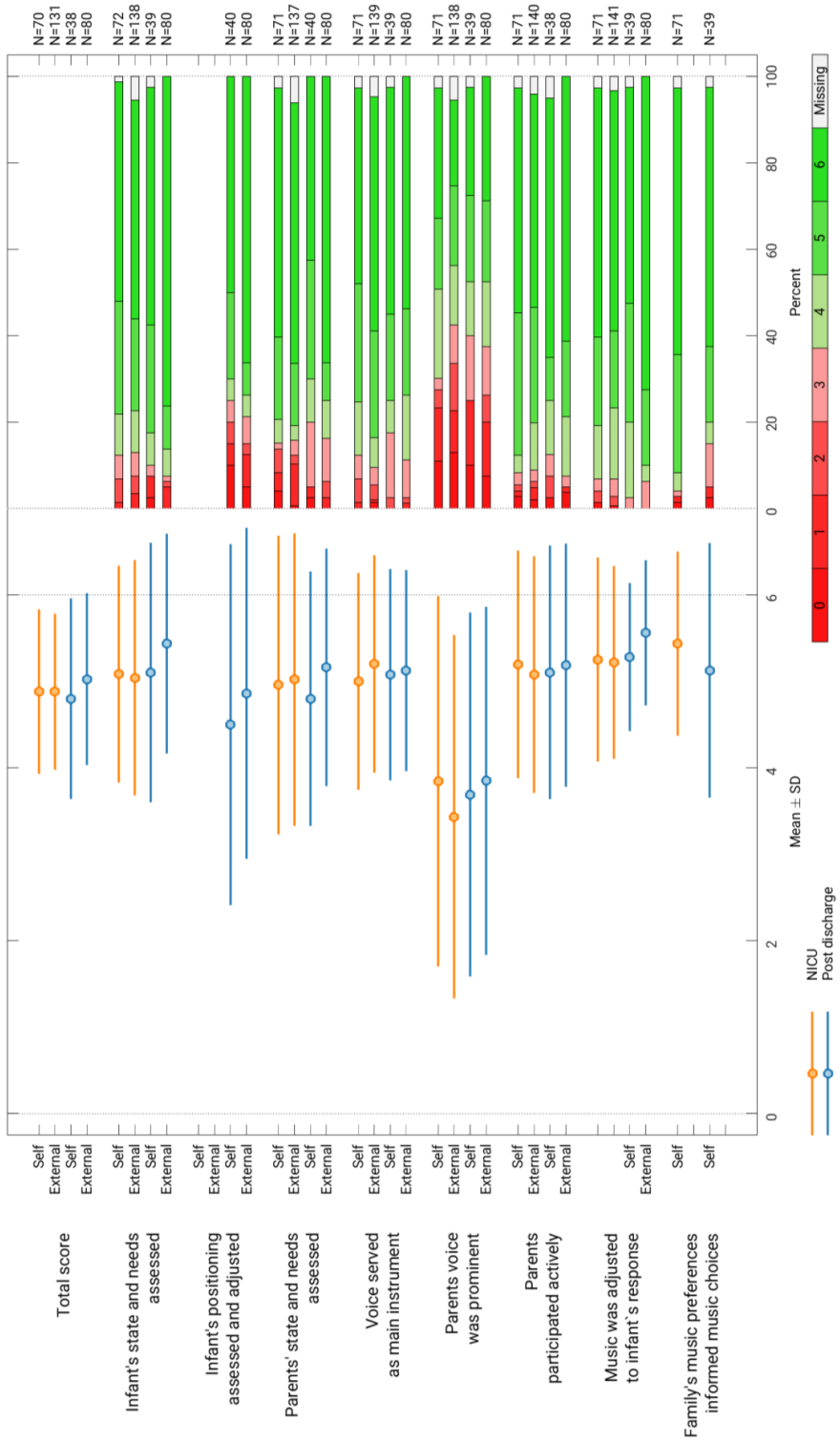


Figure 3 Treatment delivery scores per therapist

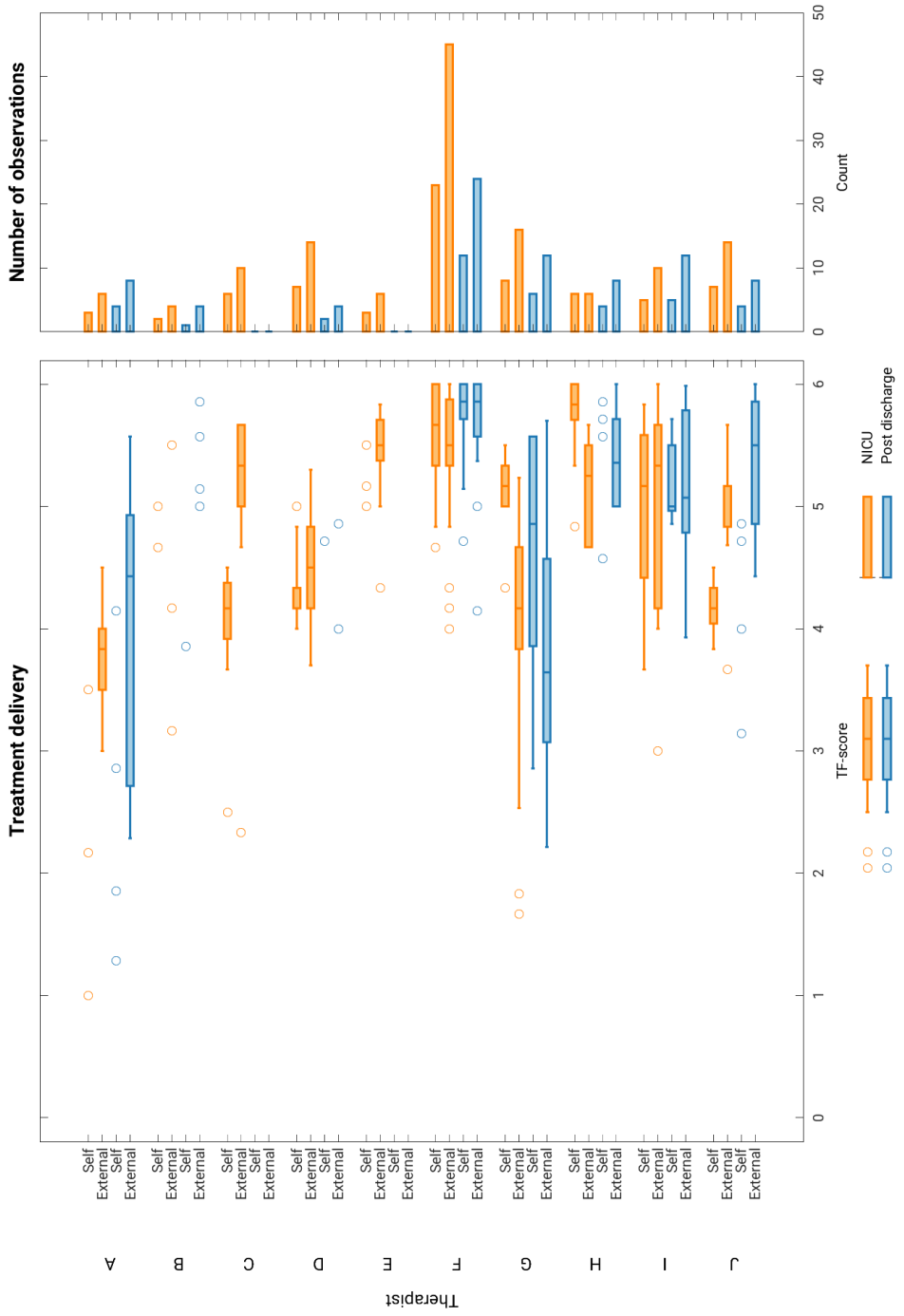
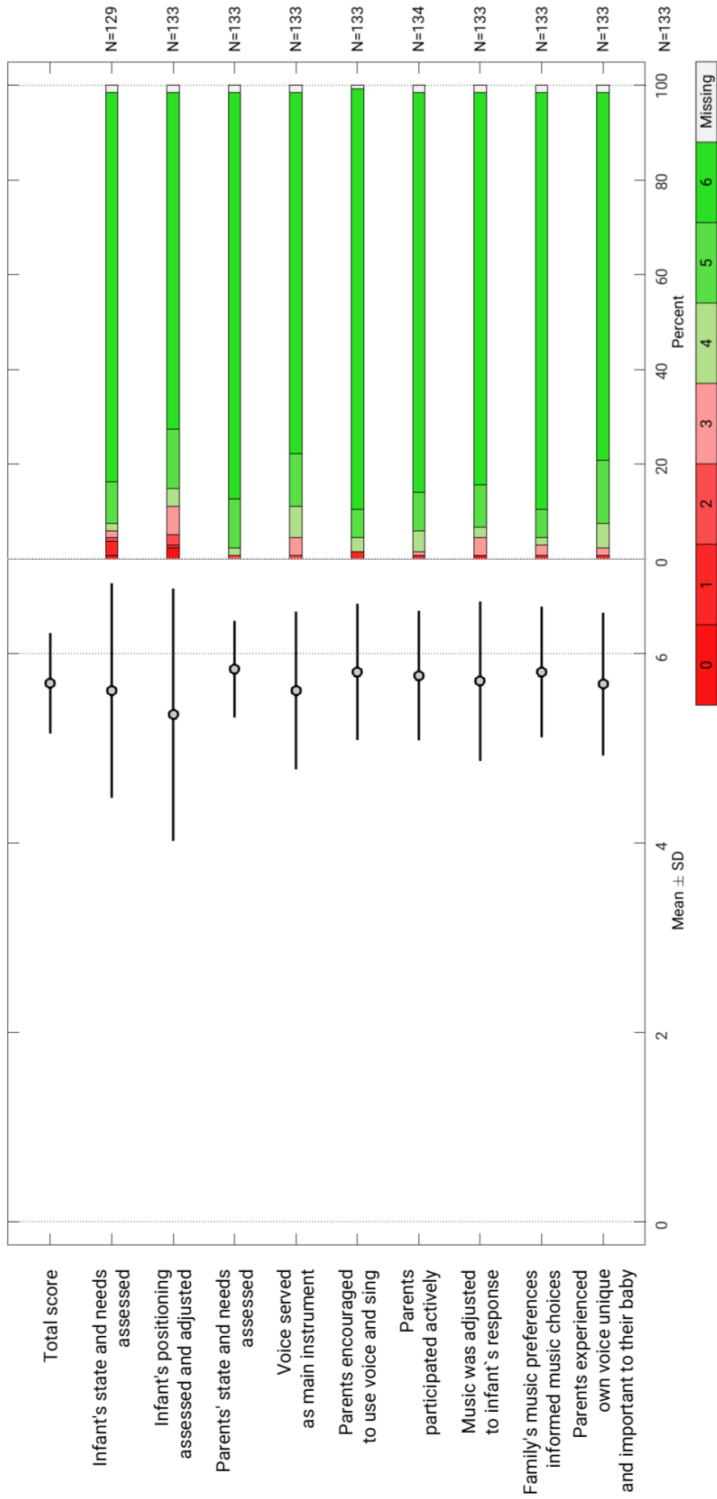


Figure 4 Treatment receipt scores per item



Discussion

We reported on treatment fidelity in a multi-nation clinical trial, LongSTEP. Average TD composite scores indicate that music therapists adhered to central elements of the intervention protocol to a satisfactory degree, and that MT was a uniform intervention during NICU stay and follow-up post-discharge. TR scores were also satisfactory, with several items scoring very high, suggesting that parents received the essential elements of the intervention. Parents who received MT during both NICU and post-discharge had the highest TR composite score, but differences between receiving MT in one or both phases were smaller than expected. Due to cultural differences between the participating countries, variation in experience of therapists, and the complex nature of the intervention, we were pleasantly surprised by these results. In line with a pragmatic approach²⁵, we were seemingly successful in our implementation of TF strategies, including the provision of guidelines for intervention delivery that left sufficient room for flexibility and individual tailoring required to fit each site's usual care across a range of cultural contexts. This indicates a high degree of clinical applicability of our MT approach outside the research context.

Treatment fidelity questionnaires had acceptable internal consistency, and moderate interrater reliability (IRR). The moderate results on IRR could be due to the complex, flexible character of the intervention. Interpreting musical interaction and subtle infant behaviours from recordings with varying quality is challenging. We also believe that rater training could have been better. Whereas the music therapists received standardized training in the intervention and supervision, the external raters received written instructions for evaluation of the sessions and written individual support when they requested it. We recommend providing more systematic training of raters, including establishing adequate interrater reliability with the intervention trainer using sample videos before commencing rating of study data. We did not complete analysis of test-retest reliability due to lack of resources but recommend such analysis. Instructions for raters should also include a narrow time window for completing ratings and we suggest that ratings be completed shortly after sessions take place, so that potential problems with recordings or other factors are discovered early in the process.

Overall TD scores were high but one item concerning parents' voices serving as prominent musical voices during MT, which scored below the threshold for satisfactory adherence (≥ 4). This finding could be explained by the fact that singing to one's baby is an intimate action that many parents can feel shy or insecure about with others present. At some sites, lack of space meant that several families shared rooms, which poses several challenges including the risk of contamination. Control group families were asked to avoid participating in any music-related intervention during the intervention period, but due to open-bay units and lack of space, intervention and control group families may at times have been in the same room. While MT was tailored individually, such that other families in the room would not have received MT per protocol, there is still a chance that they overheard tips and strategies and applied these independently. Having other families nearby might have compromised the opportunity to provide a comfortable atmosphere where parents felt safe to sing and try out new things. During supervision, several music therapists reported addressing such challenges by encouraging parents to sing while still making sure they felt comfortable and respecting their reservations and needs. A feasibility study testing our intervention found that the use of guitar was effective to support mothers' musical engagement, allowing them to feel more confident when singing.⁵⁰

It may be that expectations regarding active participation through use of voice vary considerably among external raters, music therapists and parents. Where external raters and music therapists might have expected that parents would sing often in most sessions, and hence rated this item low when singing occurred less often than expected, parents might have felt that any amount of singing was more than they would have done without MT and thus perceived their own vocal engagement as substantial. An item unique to the TR questionnaire addressed whether parents experienced their voices as being unique and important to their baby. This item had a very high (>5) score, which suggests that parents experienced their own voices as unique resources, despite music therapist and external raters rating parents low on use of voice in sessions.

While the results from the main timepoint of the LongSTEP trial are not yet analysed, results from the preliminary timepoint of discharge report non-significant effect of MT on mother-infant-bonding, maternal depression, or parental anxiety.⁵ Since our present analysis shows satisfactory levels of TF, these non-significant results do not seem to be the result of inconsistent implementation of the intervention but may rather indicate that the intervention was not well-matched for the specific outcomes measures chosen. Parents' TR scores suggest that the intervention did contribute to parents perceiving that they have something unique to offer their baby through using their voices. Through participation in MT, they also perceived essential elements about how music was adjusted to their baby's needs in the moment, which benefitted them as transferable skills they could use on their own in their everyday lives - both between sessions during NICU hospitalization and follow-up, and after the intervention period ended.

Our TF analysis has limitations. For TD evaluation, all raters knew when in the therapeutic process the session happened which might have affected raters' expectations and the outcomes of the ratings. There were large differences between the therapists' number of participants and sessions, and hence large variation in the data from which the scores were calculated. It may be that the sample of participants for TD was not representative due to our strategy of excluding participants with missing video/audio. It is also possible that poor audio/video quality in some instances made certain behaviours correspondent with the intervention's essential elements very difficult to observe. The TF questionnaires lacked an option for raters to report if the item was not possible to observe, and the degree to which raters reported poor data quality may have varied. In contrast to recordings strategically selected for TD assessment, parents who evaluated TR rated their overall experience with MT thinking back on the course of sessions over time, meaning they could base their evaluation on more sessions and probably a broader range of experiences.

LongSTEP was designed as a pragmatic trial aiming to increase applicability of study results to real world settings and usual treatment. However, through developing and implementing strategies to enhance, monitor, and evaluate TF which included development of intervention guidelines⁴³, and monitoring and supervision of music

therapists during the intervention period, one could argue that we actually moved slightly towards the explanatory end of the explanatory-pragmatic continuum.²⁵

Conclusion

Treatment fidelity questionnaires developed to assess treatment delivery and treatment receipt of MT for premature infants and their parents in the LongSTEP study showed good internal consistency and moderate interrater reliability. Treatment delivery scores indicated that music therapists across a wide range of cultural contexts were able to successfully implement the complex behavioural intervention of our MT approach, adhering to the essential elements of the intervention protocol. This indicates high clinical applicability of the LongSTEP approach to MT in NICU. Parents' high treatment receipt scores support this notion and indicate specific areas where the intervention benefitted them above and beyond the LongSTEP trial's primary and secondary outcomes. Parents experienced their own voices as unique resources in relation to their baby, and likely developed skills transferable to their daily lives.⁵¹ Future research in this area should aim to improve the interrater reliability of TF measures, for example by additional training and follow-up for raters and/or by improved operational definitions of items.

List of abbreviations

CI – Confidence interval

DC – Discharge

GA – Gestational age

ICC – Intraclass correlation coefficient

IRR – Interrater reliability

M – Mean

MD – Median

MT – Music therapy

NICU – Neonatal intensive care unit

PD – Post-discharge

RCT – Randomized controlled trial

SC – Standard care

SD – Standard deviation

TD – Treatment delivery

TF – Treatment fidelity

TR – Treatment receipt

Declarations

Clinical Trial Registration

Longitudinal Study of music Therapy's Effectiveness for Premature infants and their caregivers – “LongSTEP”. ClinicalTrials.gov Identifier: NCT03564184

Ethics approval and consent to participate

Ethics approval for the LongSTEP trial was granted by The Regional Committees for Medical and Health Research Ethics (2018/994/REK Nord, 03 July 2018). Each site also obtained ethics approvals in accordance with local and national procedures for clinical research. Informed consent from participants was obtained after written and oral explanation of project's aims, duration of involvement, expected benefits to participants and others, nature of the interventions, procedures involved in participation, and any potential risks. It was emphasized that participation in the study was voluntary and that participants could withdraw at any time from all or part of the study.

Consent for publication

All study participants were informed and consented to that the outcomes of the study were to be published, but that no details would be divulged from which the participant could be identified.

Availability of data and materials

The datasets used and analysed for this study are available from the corresponding author upon reasonable request.

Competing interests

The authors declare no competing interests with respect to the research, authorship, and/or publication of this article.

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Authors' contributions

All authors have confirmed responsibility for the reported research and approved the final manuscript as submitted. TSG drafted the initial manuscript and carried out statistical analyses. CGo, IK and CGh participated with concept and design, interpretation of data, and in drafting and revising the manuscript. JA carried out statistical analyses, made graphics, participated in interpretation of data, and in revising the manuscript. ASS and LB participated in interpretation of data, and in revising the manuscript.

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Additional files

Additional file 1: Essential elements of the LongSTEP approach to MT in NICU

1. Observation and dialogue on infant's needs prior to and during MT sessions

This element relates back to principles of developmentally appropriate care for premature infants^{1,2} and aims to ensure that each session's content is tailored to infant development, as well as their state and needs in the moment.³ Each session should begin with an initial observation together with parents of the infant's state and readiness for interaction. Physiological cues such as respiratory rate and pulse, skin colour, and behavioural cues such as motor activity and quality of movements, sleep/awake states, and responsivity help music therapist and parents to identify infant state and needs in the moment. Music therapist should articulate own observations along the way and encourage parents to do the same to promote dialogue, participation, and a shared understanding of the infant. Infant physical positioning should be observed and adjusted to support infant readiness and parent-infant mutual regulation.

2. Dialogue with parents on their state and needs prior to session

Preterm birth not only affects the infant but also parents and other family members. Parents of premature infants are at risk for significant stress⁴, post-traumatic stress disorder⁵, anxiety⁶, and depression, which in turn might negatively affect parent-infant interaction.⁷ Therefore, it is important to also check in with parents. A stressed or anxious parent might not be ready to engage in musical interaction without having a chance to express what they are experiencing and feeling. Parents should be provided time and space to express feelings and concerns, as well as needs they themselves wish the music therapist can meet.

3. Voice serves as the main instrument

The human voice is a contrast to the highly technological and often noisy environment of a NICU. In comparison to other musical instruments that have been used in NICU settings (e.g., harp, monochord, guitar)⁸⁻¹⁰ the voice is also highly available, easy to adjust in terms of volume and intensity, and as such provides an accessible resource for both therapist, parents, and others who interact with the infant. Using the voice also affects breathing patterns, which provides a potential pathway to actively work with regulating stress, emotions, and anxiety in parents.¹¹

4. Parental voice serves as the most prominent musical voice

The voices most relevant and powerful for the infant are the parents' own voices. The foetus develops the capacity for hearing during the second trimester meaning that sound is part of their sensory world long before they are born.¹² Parents' voices, and particularly the pregnant mother's voice, are therefore familiar to new-borns^{13,14} and may provide premature infants a sense of security and a link back to the safe and predictable environment of the womb.¹⁵ Focusing on parent voice also relates back to principles of resource-oriented music therapy where personal resources and empowerment are central.¹⁶

5. Music therapist provides opportunities for parents to actively participate

In line with principles of family-centred care we propose that parents should be supported to participate actively and eventually take on a leading role in the music therapy. Parents' unique insight and knowledge about their baby is a resource in this parent-infant relational work, and the music therapist brings musical competence to this collaborative therapeutic relationship. To facilitate parent musical engagement, the music therapist should accommodate parents' levels of musical experience and modify music accordingly (e.g., select comfortable vocal range). Parents should be supported in identifying and expanding their musical resources and through this learn to interpret and respond to the infant's subtle signals in a new manner which is transferable to daily life settings when the music therapist is not present.

6. Music is modified to infant cues and responses

We believe premature infants possess potential to be communicative partners with their parents through their communicative musicality.¹⁷ Although their communication cues are often subtle, and they cannot yet make their own music, premature infants can be active participants in MT through affecting and informing the song and interplay provided by the person singing.¹⁸ However, premature infants are easily overstimulated which can be harmful to their development.¹⁹ Therefore, it is crucial that all interaction is tailored to infants' states and responses along the way. As in the initial observation, infant state and behaviour should be carefully monitored and impact the way musical interaction is carried out throughout the session, following guidelines for safe uses of music with premature infants expanding musical complexity and interaction progressively in line with infant GA and readiness^{10, 20, 21}.

7. Parents' culture and musical preferences and abilities are integrated into sessions

In the same way that parents' voices are the most relevant ones, parents' own music preferences, culture, and musicality are a crucial part of the intervention, and should be integrated into the sessions. Parents are invited to share their favourite songs, or songs that might be particularly meaningful to them in the situation they are in, and the therapist demonstrates how these can be adapted in an infant-appropriate way. This is in line with Loewy's concept "song of kin"⁸ which includes everything from lullabies, parent-preferred popular music, and music that is representative of the family's nationality, religion, or cultural background. Because our study includes five different countries, we emphasize the importance of the music therapists' ability to sensitively perceive and respond to complex layers of cultures, contexts, and experiences of the families.²²

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Additional file 2: Wording of items in treatment fidelity questionnaires

Music therapist self-ratings, NICU

1. In the beginning of the session, music therapist and parent(s) observe and dialogue on infant's current state and needs prior to presenting any music
2. In the beginning of the session (and during, as appropriate), music therapist dialogues with parent(s) about parent's own state, needs, and concerns
3. Voice serves as the main instrument during the music therapy session
4. Parental voice serves as a prominent musical voice during the music therapy session
5. Music therapist provides opportunities for parents to actively participate during the music therapy session
6. Music is modified in response to infant's cues and responses during the music therapy session
7. Parents' musical preferences and abilities are integrated and accommodated into the music therapy session

Music therapist self-ratings, post-discharge

1. In the beginning of the session, music therapist and parent(s) observe and dialogue on infant's current state and needs prior to presenting any music
2. In the beginning of the session (and during, as appropriate), music therapist dialogues with parent(s) about parent's own state, needs, and concerns
3. In the beginning of the session (and during, as appropriate), infant's physical positioning is attended to and adjusted to support infant self-regulation and enhance conditions for parent--infant interaction
4. Voice serves as the main instrument during the music therapy session
5. Parental voice serves as a prominent musical voice during the music therapy session
6. Music therapist provides opportunities for parents to actively participate during the music therapy session
7. Music is modified in response to infant's cues and responses during the music therapy session
8. Parents' musical preferences and abilities are integrated and accommodated into the music therapy session

External raters, NICU

1. In the beginning of the session, music therapist and parent(s) observe and dialogue on infant's current state and needs prior to presenting any music
2. In the beginning of the session (and during, as appropriate), music therapist dialogues with parent(s) about parent's own state, needs, and concerns
3. Voice serves as the main instrument during the music therapy session
4. Parental voice serves as a prominent musical voice during the music therapy session
5. Music therapist provides opportunities for parents to actively participate during the music therapy session
6. Music is modified in response to infant's cues and responses during the music therapy session

External raters post-discharge

1. In the beginning of the session, music therapist and parent(s) observe and dialogue on infant's current state and needs prior to presenting any music
2. In the beginning of the session (and during, as appropriate), music therapist dialogues with parent(s) about parent's own state, needs, and concerns
3. In the beginning of the session (and during, as appropriate), infant's physical positioning is attended to and adjusted to support infant self-regulation and enhance conditions for parent--infant interaction

4. Voice serves as the main instrument during the music therapy session
5. Parental voice serves as a prominent musical voice during the music therapy session
6. Music therapist provides opportunities for parents to actively participate during the music therapy session
7. Music is modified in response to infant's cues and responses during the music therapy session

Parent ratings of treatment receipt

1. Before the music started, the music therapist and I looked at my baby and discussed my baby's current state (for example asleep, awake, calm, distressed), and what my baby needed in the moment
2. Before the music started, the music therapist and I checked if my baby was laying (or sitting) in a way that helped my baby be ready for music, and if needed we adjusted the baby's position
3. Before the music started, the music therapist asked me about how I was feeling, and whether I had any particular concerns, as well as any wishes or needs for the music therapy session
4. We used singing and our voices more than any other instrument during music therapy sessions
5. The music therapist encouraged me to use my voice and singing to connect with my baby in the music therapy sessions
6. I actively participated in the music therapy sessions. For example, by singing, humming, touching and/or moving my baby to the music, and/or making up songs and lyrics
7. The music was adjusted according to my baby's reaction to it. For example, we changed tempo, volume, or melody to interact with my baby, or paused and made the music simpler if the baby seemed to need a break
8. We used music that my family and I like in the music therapy sessions
9. I experienced in the music therapy sessions that I have something unique and important to offer my baby through my voice

Examples of discrete behaviors to look for

- 1. In the beginning of the session, music therapist and parent(s) are observing and dialoguing on infant's current state and needs prior to presenting any music**
Music therapist and parents observe and talk together about infant's state in the moment, physical positioning, infant strengths/resources and challenges that parents have observed lately e.g. development in self-regulatory capacities/strategies, tolerance to sensory input (e.g. touch, handling, noise etc.) communication/interaction (eye-contact, smiles, alertness) and current medical or other challenges/concerns (e.g. feeding, coming off ventilation, pain etc.).
- 2. In the beginning of the session (and during, as appropriate), music therapist dialogues with parent(s) about parent's own state, needs, and concerns**
Music therapist invites parents to share how they are doing, as well as any thoughts, concerns, and questions they might have prior to the session, and during the session if appropriate (e.g. if a parent starts crying during the session, seems stressed).
- 3. Voice serves as the main instrument during the music therapy session**
Song and voice are the most evident sources of music offered. Other instruments (guitar, lyre) are used with moderation, preferably only when requested by parent or when considered necessary to promote/encourage continuation of parental singing (e.g. if parent is not comfortable with singing without support), and is delivered in an infant-appropriate way (e.g. simple accompaniment, appropriate tempo and volume).
- 4. Parental voice serves a prominent musical voice during the music therapy session**
Music therapist makes room for the parent to take on a leading role in singing. The parental voice is distinguishable during the music therapy session. When singing together with parents, the music therapist modifies volume of own singing and takes on a supportive role musically. If parents are hesitant or insecure about singing, music therapist demonstrates and supports parent in how spoken voice can be used in an infant-directed way with musical qualities (e.g. with variations in pitch, tempo, use of pauses, vocal inflexion etc.).
- 5. Music therapist provides opportunities for parents to actively participate during the music therapy session**
Music therapist encourages and guides parents to sing or hum in an infant-directed way, invites them to touch and move infant in response/relation to the music (when appropriate). If parents are resistant to singing, music therapist offers other ways of participating such as writing or modifying lyrics of parent-chosen songs. Music therapist creates adequate space for parents, asks questions, and enables parents to share experiences they have had with their baby so far (e.g. asking what strategies have been successful in soothing the child, how the baby prefers to be held, touched etc).
- 6. Music is modified in response to infant cues and responses throughout the music therapy session**
Song and voice use within the session seems to be infant-directed, and attuned to infant cues and responsivity. Parent(s) and music therapist is observed to attend to and modify music according to infant's state (e.g., changes in alertness, responsivity) engagement/disengagement cues, and pauses/stops music in response to signs of overstimulation.

Examples of discrete behaviors to look for

- 1. In the beginning of the session, music therapist and parent(s) are observing and dialoguing on infant's current state and needs prior to presenting any music**
Music therapist and parents observe and talk together about infant's state in the moment, physical positioning, infant strengths/resources and challenges that parents have observed lately e.g. development in self-regulatory capacities/strategies, tolerance to sensory input (e.g. touch, handling, noise etc.) communication/interaction (eye-contact, smiles, alertness) and current medical or other challenges/concerns (e.g. feeding, coming off ventilation, pain etc.).
- 2. In the beginning of the session (and during, as appropriate), music therapist dialogues with parent(s) about parent's own state, needs, and concerns**
Music therapist invites parents to share how they are doing, as well as any thoughts, concerns, and questions they might have prior to the session, and during the session if appropriate (e.g. if a parent starts crying during the session, seems stressed).
- 3. Voice serves as the main instrument during the music therapy session**
Song and voice are the most evident sources of music offered. Other instruments (guitar, lyre) are used with moderation, preferably only when requested by parent or when considered necessary to promote/encourage continuation of parental singing (e.g. if parent is not comfortable with singing without support), and is delivered in an infant-appropriate way (e.g. simple accompaniment, appropriate tempo and volume).
- 4. Parental voice serves a prominent musical voice during the music therapy session**
Music therapist makes room for the parent to take on a leading role in singing. The parental voice is distinguishable during the music therapy session. When singing together with parents, the music therapist modifies volume of own singing and takes on a supportive role musically. If parents are hesitant or insecure about singing, music therapist demonstrates and supports parent in how spoken voice can be used in an infant-directed way with musical qualities (e.g. with variations in pitch, tempo, use of pauses, vocal inflexion etc.).
- 5. Music therapist provides opportunities for parents to actively participate during the music therapy session**
Music therapist encourages and guides parents to sing or hum in an infant-directed way, invites them to touch and move infant in response/relation to the music (when appropriate). If parents are resistant to singing, music therapist offers other ways of participating such as writing or modifying lyrics of parent-chosen songs. Music therapist creates adequate space for parents, asks questions, and enables parents to share experiences they have had with their baby so far (e.g. asking what strategies have been successful in soothing the child, how the baby prefers to be held, touched etc).
- 6. Music is modified in response to infant cues and responses throughout the music therapy session**
Song and voice use within the session seems to be infant-directed, and attuned to infant cues and responsivity. Parent(s) and music therapist is observed to attend to and modify music according to infant's state (e.g. changes in alertness, responsivity) engagement/disengagement cues, and pauses/stops music in response to signs of overstimulation.
- 7. Parents' musical preferences and abilities are integrated into the music therapy session**
Parents are encouraged to make share preferences and familiar songs that in turn, are integrated and accommodated within the session in adapted forms. "Preferred songs" refers to both lullabies and other types of music (e.g. pop music, music representing parents' nationality/culture/religion/beliefs etc.). Music therapist accommodates to parent's musical abilities (e.g. no/some experience with singing) and modifies music (e.g. models/accompanies in comfortable vocal range/key) to facilitate musical engagement of the parent. The importance of the parental voice for the infant is emphasized, and spontaneous infant-directed speech, and vocalizations from the parent are encouraged/welcomed.

LongSTEP Treatment Delivery Tool C

Post-discharge phase, external rater version

Rater initials:

Site:

Music therapist initials:

OC Participant ID:

Session number:

Please specify who are visible in the video frame and your access to observe facial expressions and body movements

Fully visible= Facial expressions and body movements easily observed

Partly visible= Facial expressions and body movements somewhat possible to observe

Limited visibility= Difficult or not possible to see facial expressions, and limited view of body movements

Not visible, but audible= Present and audible, but not visible

Not visible, nor audible = Present, but neither visible nor audible

Place an "X" in the applicable cell for each participant present in the session

	Infant	Parent 1 (mother)	Parent 2 (partner, father)	Music therapist	Other (specify)
Not present					
Fully visible					
Partly visible					
Limited visibility					
Not visible, but audible					
Not visible, nor audible					

Examples of discrete behaviors to look for

- 1. In the beginning of the session, music therapist and parent(s) are observing and dialoguing on infant's current state and needs prior to presenting any music**
Music therapist and parents observe and talk together about infant's state in the moment, physical positioning, infant strengths/resources and challenges that parents have observed lately e.g. development in self-regulatory capacities/strategies, tolerance to sensory input (e.g. touch, handling, noise etc.) communication/interaction (eye-contact, smiles, alertness) and current medical or other challenges/concerns (e.g. feeding, coming off ventilation, pain etc.).
- 2. In the beginning of the session (and during, as appropriate), infant's physical positioning is attended to and adjusted to support infant self-regulation and enhance conditions for interaction between infant and parent(s)**
Music therapist and parent discuss how infant is currently positioned, as well as any infant preferences for positioning and containment that the parent is familiar with. If appropriate, music therapist may demonstrate containment strategies (e.g., static cupping of feet and top of head, static touch on infant's back, gathering of infant's hands, holding infant in slightly more upward position to promote alertness) for the parent to use during sessions.
- 3. In the beginning of the session (and during, as appropriate), infant's physical positioning is attended to and adjusted to support infant self-regulation and enhance conditions for interaction between infant and parent(s)**
Music therapist and parent discuss how infant is currently positioned, as well as any infant preferences for positioning and containment that the parent is familiar with. If appropriate, music therapist may demonstrate strategies to facilitate interaction for the parent to use during sessions (e.g., gathering of infant's hands, holding infant in slightly more upward position to promote alertness)
- 4. Voice serves as the main instrument during the music therapy session**
Song and voice are the most evident sources of music offered. Other instruments (guitar, lyre) are used with moderation, preferably only when requested by parent or when considered necessary to promote/encourage continuation of parental singing (e.g., if parent is not comfortable with singing without support) and is delivered in an infant-appropriate way (e.g., simple accompaniment, appropriate tempo and volume).
- 5. Parental voice serves a prominent musical voice during the music therapy session**
Music therapist makes room for the parent to take on a leading role in singing. The parental voice is distinguishable during the music therapy session. When singing together with parents, the music therapist modifies volume of own singing and takes on a supportive role musically.
- 6. Music therapist provides opportunities for parents to actively participate during the music therapy session**
Music therapist encourages and guides parents to sing or hum in an infant-directed way, invites them to touch and move infant in response/relation to the music (when appropriate). If parents are resistant to singing, music therapist offers other ways of participating such as writing or modifying lyrics of parent-chosen songs.
- 7. Music is modified in response to infant cues and responses throughout the music therapy session**
Song and voice use within the session seems to be infant-directed and attuned to infant cues and responsivity. Parent(s) and music therapist is observed to attend to and modify music according to infant's state (e.g., changes in alertness, responsivity) engagement/disengagement cues, and pauses/stops music in response to signs of overstimulation.

LongSTEP Treatment Delivery Tool D

Post-discharge phase, self-rating version

Site:

Music therapist initials:

OC Participant ID:

Session number:

Please specify who are visible in the video frame and your access to observe facial expressions and body movements

Fully visible= Facial expressions and body movements easily observed

Partly visible= Facial expressions and body movements somewhat possible to observe

Limited visibility= Difficult or not possible to see facial expressions, and limited view of body movements

Not visible, but audible= Present and audible, but not visible

Not visible, nor audible = Present, but neither visible nor audible

Place an "X" in the applicable cell for each participant present in the session

	Infant	Parent 1 (mother)	Parent 2 (partner, father)	Music therapist	Other (specify)
Not present					
Fully visible					
Partly visible					
Limited visibility					
Not visible, but audible					
Not visible, nor audible					

Examples of discrete behaviors to look for

1. In the beginning of the session, music therapist and parent(s) are observing and dialoguing on infant's current state and needs prior to presenting any music

Music therapist and parents observe and talk together about infant's state in the moment, physical positioning, infant strengths/resources and challenges that parents have observed lately e.g. development in self-regulatory capacities/strategies, tolerance to sensory input (e.g. touch, handling, noise etc.), communication/interaction (eye-contact, smiles, alertness) and current medical or other challenges/concerns (e.g. feeding, coming off ventilation, pain etc.).

2. In the beginning of the session (and during, as appropriate), music therapist dialogues with parent(s) about parent's own state, needs, and concerns

Music therapist invites parents to share how they are doing, as well as any thoughts, concerns, and questions they might have prior to the session, and during the session if appropriate (e.g., if a parent starts crying during the session, seems stressed).

3. In the beginning of the session (and during, as appropriate), infant's physical positioning is attended to and adjusted to support infant self-regulation and enhance conditions for interaction between infant and parent(s)

Music therapist and parent discuss how infant is currently positioned, as well as any infant preferences for positioning and containment that the parent is familiar with. If appropriate, music therapist may demonstrate strategies to facilitate interaction for the parent to use during sessions (e.g., gathering of infant's hands, holding infant in slightly more upward position to promote alertness).

4. Voice serves as the main instrument during the music therapy session

Song and voice are the most evident sources of music offered. Other instruments (guitar, lyre) are used with moderation, preferably only when requested by parent or when considered necessary to promote/encourage continuation of parental singing (e.g., if parent is not comfortable with singing without support) and is delivered in an infant-appropriate way (e.g., simple accompaniment, appropriate tempo and volume).

5. Parental voice serves a prominent musical voice during the music therapy session

Music therapist makes room for the parent to take on a leading role in singing. The parental voice is distinguishable during the music therapy session. When singing together with parents, the music therapist modifies volume of own singing and takes on a supportive role musically.

6. Music therapist provides opportunities for parents to actively participate during the music therapy session

Music therapist encourages and guides parents to sing or hum in an infant-directed way, invites them to touch and move infant in response/relation to the music (when appropriate). If parents are resistant to singing, music therapist offers other ways of participating such as writing or modifying lyrics of parent-chosen songs. Music therapist creates adequate space for parents, asks questions, and enable parents to share experiences they have had with their baby so far (e.g., asking what strategies have been successful in soothing the child, how the baby prefers to be held, touched etc.).

7. Music is modified in response to infant cues and responses throughout the music therapy session

Song and voice use within the session seems to be infant-directed and attuned to infant cues and responsivity. Parent(s) and music therapist is observed to attend to and modify music according to infant's state (e.g., changes in alertness, responsivity) engagement/disengagement cues, and pauses/stops music in response to signs of overstimulation.

8. Parents' musical preferences and abilities are integrated into the music therapy session

Parents are encouraged to make share preferences and familiar songs that in turn, are integrated and accommodated within the session in adapted forms. "Preferred songs" refers to both lullabies and other types of music (e.g., pop music, music representing parents' nationality/culture/religion/beliefs etc.). Music therapist accommodates to parent's musical abilities (e.g., no/some experience with singing) and modifies music (e.g., models/accompanies in comfortable vocal range/key) to facilitate musical engagement of the parent. The importance of the parental voice for the infant is emphasized, and spontaneous infant-directed speech, and vocalizations from the parent are encouraged and welcomed

TR_6	We used singing and our voices more than any other instrument during music therapy sessions:
------	--

I completely agree

--	--	--	--	--	--	--

 I completely disagree

TR_7	The music therapist encouraged me to use my voice and singing to connect with my baby in the music therapy sessions:
------	--

I completely agree

--	--	--	--	--	--	--

 I completely disagree

TR_8	I actively participated in the music therapy sessions. For example, by singing, humming, touching and/or moving my baby to the music, and/or making up songs and lyrics:
------	--

I completely agree

--	--	--	--	--	--	--

 I completely disagree

TR_9	The music was adjusted according to my baby's reaction to it. For example, we changed tempo, volume, or melody to interact with my baby, or paused and made the music simpler if the baby seemed to need a break:
------	---

I completely agree

--	--	--	--	--	--	--

 I completely disagree

TR_10	We used music that my family and I know and like in the music therapy sessions:
-------	---

I completely agree

--	--	--	--	--	--	--

 I completely disagree

TR_11	I experienced in the music therapy sessions that I have something unique and important to offer my baby through my voice:
-------	---

I completely agree

--	--	--	--	--	--	--

 I completely disagree

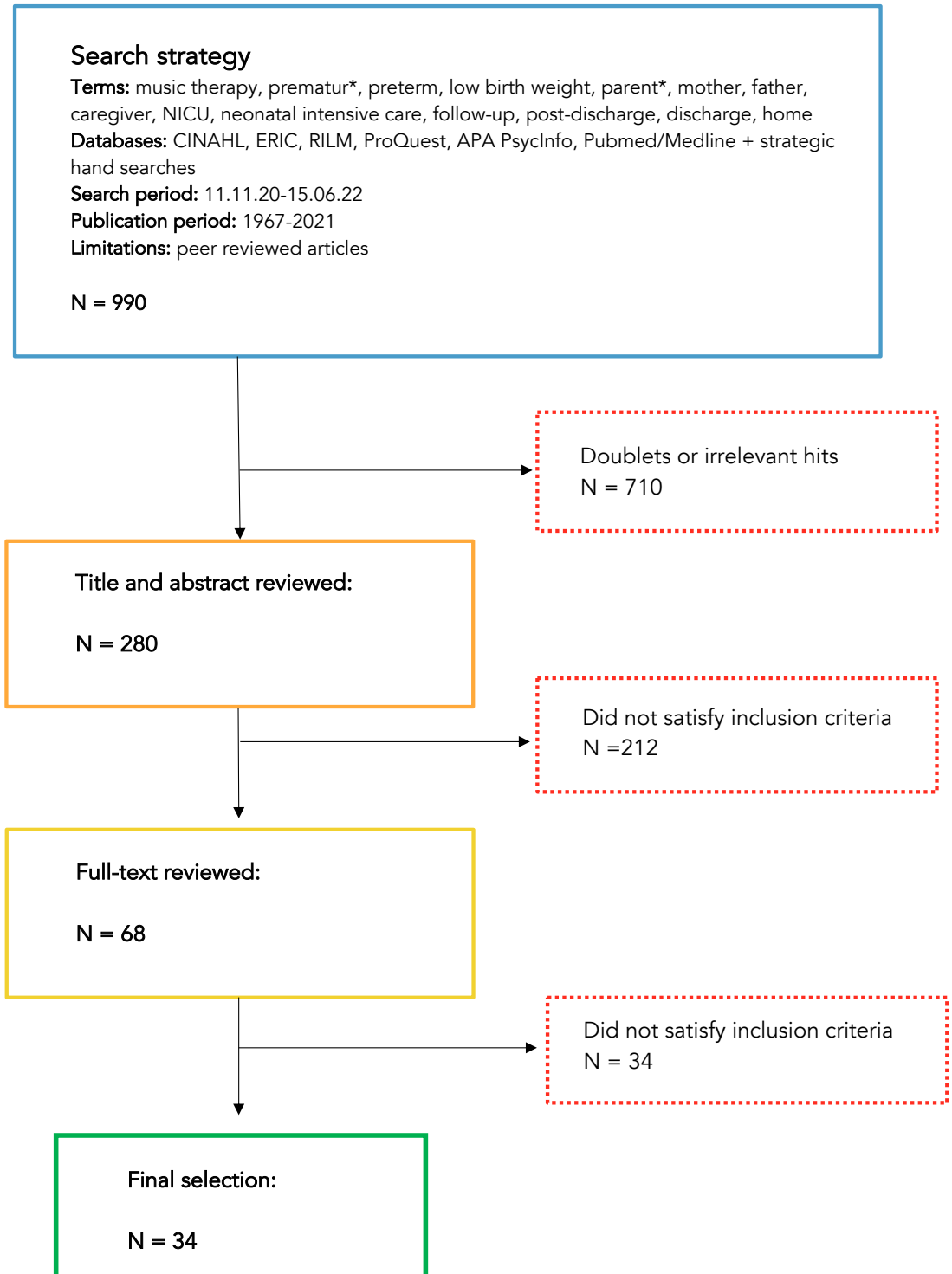
Thank you for completing this questionnaire! We appreciate your time and effort.

Appendices

1. Search strategy and inclusion criteria
2. Reviewed literature
3. Ethics approval (REK)
4. Participant information and consent form
5. Table 2 Parent baseline characteristics, corrected version
6. Conceptual model of the theoretical framework and key elements of the LongSTEP approach
7. Model of proposed mechanisms of action of the LongSTEP approach
8. Adjusted model of proposed mechanisms of action of the LongSTEP approach

Appendix 1

Search strategy and inclusion criteria



Inclusion criteria

Literature was included if it met the following criteria:

1. Participants

Infants born prematurely, defined by the World Health Organization as born before 37 completed weeks of gestation, or fewer than 259 days since the first day of the woman's last menstrual period, and their parents/caregivers.

2. Intervention

MT carried out by, or in consultation with, a trained music therapist, conducted in a neonatal unit, or neonatal intensive care unit, or during follow-up after discharge from hospital with parents as participants in the sessions, using live music.

3. Outcomes

Reports at least one outcome of the following domains as either primary or secondary outcomes: parent-infant-bonding, attachment, communication/interaction, parental sensitivity/reflective functioning, mutual-co-regulation, parent mental health and well-being, e.g., symptoms of postpartum depression, stress, anxiety, or post-traumatic stress.

4. Types of articles

Primarily individual, empirical articles were included. Some theoretical and conceptual articles of particular interest (e.g., novel areas of research or underrepresented perspectives) were included. Study protocol and literature reviews were excluded. Systematic reviews and meta-analyses who included trials with parent outcomes were included.

Appendix 2

Reviewed literature									
Authors	Year	Title	Journal	Design	Country	Participants	Outcomes	Key findings	
Epstein et al.	2022	Israeli Parents' Lived Experiences of Music Therapy with Their Preterm Infants Post Hospitalization	Journal of Music Therapy	Qualitative interview study	Israel	7 parents of premature infants born ≤ 32 weeks GA	Parent's lived experiences from MT post-discharge	Parents participating in MT post-discharge in accordance with the LongSTEP MT protocol experienced MT as a potential means of transformation in communication skills, resourcefulness and sense of agency, and a way of expanding their relationship with the infant post-discharge. However, singing was perceived as embarrassing or unnatural at first.	
Coombes & Al-Muzaffar	2021	The Singing Unit—a pilot study investigating the efficacy of a music therapy singing intervention in a local neonatal unit to support parent/infant bonding and reduce parental anxiety	Journal of Neonatal Nursing	Mixed-methods pilot study including within-subjects measures and parent questionnaires	Wales, United Kingdom	Parents of infants born ≤ 36 weeks GA	Parent anxiety, mother-infant bonding, parent wellbeing, parent perception of workshop and usefulness of intervention	Low-intensity MT consisting of a 1-hour workshop was experienced as positive and useful for the rest of the NICU stay by parents, and improved parental wellbeing significantly. The study offered a cost-effective, low intensity intervention that was well-accepted among families and staff.	
Ghetti et al.	2021	Longitudinal study of music therapy's effectiveness for premature infants and their caregivers (LongSTEP): Feasibility study with a Norwegian cohort	Journal of Music Therapy	Mixed-methods, non-randomized feasibility study	Norway	3 families with premature infants born ≤ 35 weeks GA	Feasibility, acceptability, and suitability of the treatment arm of the LongSTEP trial	LongSTEP study procedures, outcome measures, and MT approach were accepted and feasible in a Norwegian cohort. Parents were willing to participate in MT emphasizing parental voice in both NICU and post-discharge phases, recognizing parental voice as a central means of building relation with their infants. However, singing within a hospital context was perceived by some as new and a little embarrassing.	
Haslbeck et al.	2021	Music Is Life—Follow-Up Qualitative Study on Parental Experiences of Creative Music Therapy in the Neonatal Period	International Journal of Environmental Research and Public Health	Qualitative, retrospective interview study	Switzerland	Parents of 13 extremely premature infants < 29 weeks GA who had participated in feasibility	Parents experiences of CMT during NICU and possible longitudinal influence	Parents expressed positive, formative impact of CMT on their infants, themselves, and bonding in the stressful NICU setting and beyond. Some parents called for MT to continue post-discharge.	

Janner et al.	2021	Implementing music therapy in a Norwegian neonatal intensive care unit	Nursing Children and Young People	Report from exploratory implementation project, including parent survey	Norway	Parents of premature infants, NICU nursing staff	Parent experiences, and staff feedback about MT	Parent experience with MT was overall positive, reporting benefits for themselves and their infants. Nurses observed that parent-infant interaction increased following MT, but also reported challenges in recruiting parents to MT in a time of uncertainty, anxiety, and grief. Additional efforts are required to integrate MT into routine care provided by the NICU multidisciplinary team.
Alvarez et al.	2021	The impact of music therapy in late-moderate premature infants, on their parents and their environment, in a Spanish neonatal intermediate care unit	Music & Medicine	Quasi experimental study without control group, parent interviews	Spain	44 late-moderate premature infants 32-36 weeks GA) and their parents	Infant HR, RR, SO ₂ , and comfort level, parents' experiences with MT, noise level and bustle of environment	Significant effects on infant O ₂ saturation, and HR was found post-intervention, increased behavioral signs of comfort (sucking behavior, smiles). Parent experiences in interviews that MT calmed them. A decrease of tension in environment was observed.
Kehl et al.	2021	Creative music therapy with premature infants and their parents: A mixed-method pilot study on parents' anxiety, stress and depressive symptoms and parent-infant attachment	International Journal of Environmental Research and Public Health	Mixed-methods pilot study, including RCT and parent interviews	Switzerland	16 Parents of premature infants born ≤32 weeks GA	Parent anxiety, stress and depressive symptoms, parent perception of attachment	CMT + Kangaroo care did not have a significant effect on parent anxiety, stress and depression compared to standard care. However, between the first two weeks after birth and approximately halfway through the NICU stay the CMT + KC group showed a significant reduction of anxiety suggesting that CMT might be particularly useful during the first weeks in the NICU. Parents expressed that the MT helped them relax and experience intimacy and closeness with their baby.
Kobus et al	2021	Parents' Perception of Family-Centered Music Therapy with Stable Preterm Infants	International Journal of Environmental Research and Public Health	Quantitative parent survey, sub-study of RCT	Germany	32 parents of stable preterm infants born ≤32 weeks GA	Parent perception of MT	MT was perceived by parents as a positive, supportive offer for themselves and their infants during NICU. However, several parents did not experience increased confidence leading to singing more to their infants, nor developed joy in singing during the course of MT.
Loscalzo et al.	2021	Premature Triadic Music Therapy (PT-MT) for	Journal of Reproductive	Experimental feasibility study	Italy	6 preterm infants with	Infant HR, blood perfusion and	The MT intervention emphasizing the participation of both parents was found

		babies and their parents: a pilot study	and Infant Psychology	with pre-post-test design		GA 33-36 weeks and their mothers and fathers	blood saturation, parent symptoms of post-partum depression	feasible for the NICU setting where it was tested. Significant decrease of HR and increase of blood saturation was found during PT-MT but the changes were not present at the end of sessions. No significant effects were found for parent depression.
Menke et al.	2021	Family-centered music therapy—Empowering premature infants and their primary caregivers through music: Results of a pilot study	PloS one	Pilot study, in preparation of larger RCT	Germany	65 premature infants born <30 weeks GA	Infant physiological development and parent stress	From pre-to-post-intervention, parents showed a significant reduction in stress factors. However, there were no significant differences between groups. Infants in the MT group showed descriptively shorter durations of therapy, tube feeding, and length of hospitalization compared control group, but the difference between groups were not significant.
Palazzi et al	2021a	NICU music therapy effects on maternal mental health and preterm infant's emotional arousal	Infant Mental Health Journal	Pre-experimental study with control group	Brazil	33 premature infants <37 weeks GA and their mothers >18 years	Maternal anxiety, postnatal depression, stress, infant weight gain, length of hospitalization, HR and SO ₂	Mothers' anxiety, depression and stress levels was significantly lower in the MT group at discharge. A large effect was found pre-post MT session on anxiety. No significant effects were found on infant weight gain, length of hospitalization, HR or SO ₂ .
Palazzi et al	2021b	Music therapy enhances preterm infant's signs of engagement and sustains maternal singing in the NICU	Infant Behavior and Development	Quasi experimental study with quantitative video and audio analysis	Brazil	30 premature infants and their mothers	Infant signs of engagement (eye opening, smiling), maternal vocalizations	MT significantly increased infant eye-opening frequency during mother-infant interactions when infants were in awake state when interaction was initiated. Non-religious mothers sang significantly more in MT group than in CG. MT may promote infant signs of engagement and sustain maternal singing.
Ullsten et al.	2021	Parents as nurturing resources: A family integrated approach to music therapy for neonatal procedural support.	Music & Medicine	Theoretical article	Sweden, Finland, Norway	Parents of premature infants	Parents' role in neonatal pain management	Parents represent a valuable but often overlooked resource in neonatal pain management. Parents need and want to participate actively to reduce their infant's pain. In line with family-centered care, parents should be supported in actively engaging in MT as procedural support. Parents' infant-directed singing to calm and soothe can be an apt format. Music therapists are advised to

van Dokkum et al.	2021	Feasibility of Live-Performed Music Therapy for Extremely and Very Preterm Infants in a Tertiary NICU	Frontiers in Pediatrics	Single center feasibility study	Netherlands	18 extremely and very premature infants and their parents	Participation rate, signs of infant overstimulation, parent- and nurse perceived effects	take on a supportive, collaborative role, to ensure that the parent-infant relationship is the primary focus. Participation was high, parents reported high satisfaction with MT and perceived improvements in infant and own breathing. Live-performed MT was feasible and well tolerated. No signs of overstimulation due to MT was observed. Nurses perceived NICU environment as quieter post-MT. A limited number of parents were present in sessions.
Yue et al.	2021	Effect of music therapy on preterm infants in neonatal intensive care unit: Systematic review and meta-analysis of randomized controlled trials	Journal of Advanced Nursing	Systematic review and meta-analysis	China	13 RCTs involving 1093 participants	Effectiveness of MT on preterm infants in NICU	MT had significant positive effects on infant HR, RR, oral feeding volume, stress level, and maternal anxiety. Heterogeneity among studies was moderate to high.
Bielenik et al.	2020	Evaluating feasibility of the LongSTEP (Longitudinal study of music therapy's effectiveness for premature infants and their caregivers) protocol with a Polish cohort	Nordic Journal of Music Therapy	Mixed-methods feasibility study	Poland	6 families with premature infants born ≤ 35 weeks GA	Feasibility, acceptability, and suitability of the treatment arm of the LongSTEP trial	LongSTEP study procedures, outcome measures, and MT approach were accepted and feasible in a Polish cohort. Fathers were less present than mothers during NICU admission. Some parents expressed insecurity about singing in sessions. Providing instrumental accompaniment and doing MT sessions in private rooms where possible were recommended.
Epstein et al.	2020	Preterm infants with severe brain injury demonstrate unstable physiological responses during maternal singing with music therapy: a randomized controlled study	European Journal of Pediatrics	Randomized controlled trial	Israel	40 preterm infants born ≤ 32 weeks GA with severe brain injury and their mothers	Infant physiological and behavioral stability, maternal anxiety	Maternal singing during skin-to-skin care, together with MT delivered by a trained music therapist induced physiological and behavioral instability in infants, and increased mothers' anxiety during NICU hospitalization. A unique MT intervention should be tailored for the population.
Kostilainen et al.	2020	Effects of maternal singing during kangaroo care on maternal anxiety.	Nordic Journal of Music Therapy	Mixed-methods study with cluster-	Finland	36 mothers of premature infants born	Maternal anxiety, wellbeing, and perceived quality	Maternal anxiety decreased significantly in the group combining MT and Kangaroo Care, compared to control group mothers. In the

McLean et al.	2019	Parents' musical engagement with their baby in the neonatal unit to support emerging parental identity: A grounded theory study	Journal of Neonatal Nursing	Qualitative grounded theory study, based on parent diaries	Australia	9 Parents of premature infants born <35 weeks GA	Parent participation in MT and contributions to parental identity	of early mother-infant relationship	survey the mothers described that singing supported their mother-infant relationship by promoting emotional closeness and creating early interaction moments.
Eitenberger & Ardila	2018	Music therapy song writing with mothers of preterm babies in the Neonatal Intensive Care Unit (NICU)-A mixed-methods pilot study	The Arts in Psychotherapy	Mixed-methods pilot study	Colombia	15 mothers and medically stable premature infants	Bonding, maternal depression, anxiety, and wellbeing	Parents highlighted the importance of their baby's response during musical interaction to their emerging sense of parental identity. The quality of their baby's responsiveness significantly shaped their own responses and commitment to fostering musical interactions. Favorable effects were found on all outcome measures, but the sample was too small to achieve statistically significant results. Findings suggest that MT songwriting might be particularly useful for mothers with anxiety, depression, or risk for impaired bonding. The qualitative analysis showed that songwriting allowed for creative expression of feelings and thoughts.	
Shoemark, H.	2018	Time Together: A Feasible Program to Promote parent-infant Interaction in the NICU	Music Therapy Perspectives	Qualitative feasibility trial	Australia	13 mothers of infants in NICU	Feasibility and acceptability of parent educational program	Though not defined as MT, the Time Together program builds on aspects of music therapist-delivered contingent singing, which is demonstrated and practiced with parents. In just one session parents could improve their understanding of infant behavioral interaction cues.	
Eitenberger, M.	2017	Music therapy during end-of-life care in the neonatal intensive care unit (NICU): Reflections from early clinical practice in Colombia	Voices: A World Forum for Music Therapy	Report with case studies	Colombia	Parents with infants receiving end-of-life care	Potentials and challenges of MT during end-of-life care	Family-centered care, rituality and memory-making are central themes during end-of-life care. Context and culture influence attitudes and practices of end-of-life care. Adequate training, supervision and self-care practices are recommended for music therapists working in this novel area of practice.	
Eitenberger et al.	2017	Family-centred music therapy with preterm infants and their parents in the Neonatal Intensive	Nordic Journal of Music Therapy	Mixed-methods study with RCT and interviews	Colombia	36 medically stable premature	Infant HR, SO ₂ , weight gain, length of hospitalization	MT combined with Kangaroo Care resulted in significant improvements in maternal anxiety and infant weight gain. Qualitative analysis showed MT was important for parent well-	

		Care Unit (NICU) in Colombia – A mixed-methods study							infants and their parents	and re-hospitalization rate, parent anxiety, bonding	being bonding, and coping with the hospital environment.
Hamm et al.	2017	A parent-infant music therapy intervention to improve neurodevelopment after neonatal intensive care	Acta Paediatrica	Retrospective case control study	USA	USA	10 parents and infants with former NICU admission	Infant neuro-development	Participation in a weekly developmental MT class was significantly associated with infant cognition, communication, and motor domain scores. As the number of sessions increased, so did communication and cognition scores.		
Bieleninik et al.	2016	Music therapy for Preterm Infants and Their Parents: A Meta-analysis	Pediatrics	Meta-analysis	Norway	Norway	14 RCTs involving 964 infants and 266 parents	Effect of MT on preterm infants and their parents during NICU and post-discharge	A large favorable effect was confirmed on infant RR and maternal anxiety.		
McLean	2016	Exploring Parents' Experiences and Perceptions of Singing and Using their Voice with their Baby in a Neonatal Unit: An Interpretative Phenomenological Analysis	Qualitative Inquiries in Music Therapy	Qualitative interview study	Australia	Australia	9 parents of premature infants (2 fathers, 7 mothers)	Parents' experiences and perceptions of singing and using their voice in a neonatal unit	Singing and using voice was experienced as validating parental identity, providing opportunities for emotional catharsis, as well as self-soothing, and a way to build of connection with their baby. The music therapist was perceived as a partner supporting parents in their process of understanding the role of their voice to support connection to their baby.		
Mondanaro et al.	2016	Mars rising: Music therapy and the increasing presence of fathers in the NICU	Music & Medicine	Essay with case examples	USA, Colombia	USA, Colombia	Fathers of premature infants	Fathers' role and benefits of NICU MT	MT is well suited to meet the needs of fathers in the NICU through providing a culturally sensitive and normalizing environment where fathers can connect to their child, their own emotions, and take-on an active role in the NICU environment		
Loewy	2015	NICU music therapy: song of kin as critical lullaby in research and practice	Annals of the New York Academy of Sciences	Subgroup analysis of randomized clinical multisite trial	USA	USA	272 premature infants aged 32 weeks	Infant HR, RR, SO ₂ , activity level, feeding, sleeping, caloric, parent perceived stress level	Culturally-based, parent-selected, personalized musical tunes - "songs of kin" (SOK) were compared to standard popular lullaby "Twinkle, Twinkle", performed by music therapist. Lower HR was recorded during SOK. Standard lullaby had a stronger effect on infant oxygen saturation than the SOK, but difference was not significant.		

Ettenberger et al.	2014	Music therapy with premature infants and their caregivers in Colombia: A mixed methods pilot study including a randomized trial	Voices: A World Forum for Music Therapy	Mixed methods pilot study with RCT	Colombia	19 medically stable premature infants and their parents	Infant HR, SO ₂ , weight gain, cephalic perimeter, length of hospitalization, maternal anxiety, bonding	Parents stress was significantly lower after SOK. No significant effects on study outcomes were detected. Trends and qualitative data suggest MT had potential to help both children and parents to relax, strengthen their relationship, and offer premature infants a calming environment.
Haslbeck, F.	2014	The interactive potential of creative music therapy with premature infants and their parents: A qualitative analysis	Nordic Journal of Music Therapy	Qualitative observational study with video analysis	Switzerland		Interactive potential of CMT	CMT can contribute to better interaction between parents and infant. CMT may also increase parents' self-confidence, well-being and contribute to parent empowerment and coping.
Loevy et al.	2013	The Effects of Music Therapy on Vital Signs, Feeding, and Sleep in Premature Infants	Pediatrics	Randomized clinical multisite trial	USA	272 premature infants aged 32 weeks	Infant HR, RR, SO ₂ , activity level, feeding, sleeping, caloric, parent perceived stress level	Parent-preferred melodies and entrained live rhythm and breath sounds lowered infant HR and influenced sleep patterns significantly. Parent perceived stress level was significantly reduced.
Walworth, D.	2009	Effects of Developmental Music Groups for Parents and Premature or Typical Infants Under Two Years on Parental Responsiveness and Infant Social Development	Journal of Music Therapy	Quasi-experimental matched subjects study with posttest only	USA	56 caregiver-infant dyads with both premature and typically developing full-term infants under 2 years old	Parent responsiveness, infant social behaviors, parent perception of infant general development	Both premature and full-term infants attending 3 group developmental MT sessions engaged in significantly more social toy play behaviors than infants not attending MT. Parents attending MT were more positively engaged with their children, but not significantly so. Premature infants attending MT showed more social behaviors and less alone behaviors than full-term control infants.
Shoemark & Dearn	2008	Keeping Parents at the Centre of Family Centred Music Therapy with Hospitalised Infants	Australian Journal of Music Therapy	Essay based on authors' clinical experience	Australia	Parents of hospitalized infants	Potential benefits of family-centered MT for parents	While the evidence-base concerning infant benefits of MT in NICU is established, addressing parents' needs and role in NICU MT is necessary to truly align with family-centered care models.
Whipple, J.	2000	The effect of parent training in music and multimodal stimulation	Journal of Music Therapy	Experimental/control group design study	USA	20 parents and their premature	Parent-infant interaction, infant stress behaviors,	Signs of stress in the premature infant were significantly lower in the experimental group, and the parents in the experimental group

		on parent-neonate interactions in the neonatal intensive care unit				low birth weight infants.	infant weight gain, length of hospitalization	responded more appropriately to infant responses and spent more time with the infant in the ward.
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Abbreviations: NICU – neonatal intensive care unit, MT – music therapy, GA – gestational age, KC – kangaroo care, SSC – skin-to-skin care, CMT – creative music therapy, FCC – family-centered care, SOK – song of kin, HR – heart rate, RR – respiratory rate, SO₂ – oxygen saturation

Appendix 3



Region: REK nord	Saksbehandler: Veronica Sørensen	Telefon: 77620758	Vår dato: 03.07.2018	Vår referanse: 2018/994/REK nord
			Deres dato: 28.06.2018	Deres referanse:

Vår referanse må oppgis ved alle henvendelser

Claire Ghetti
Griegakademiet

2018/994 Musikkterapi for premature barn og deres foreldre - hovedstudie (LongSTEP)

Forskningsansvarlig institusjon: Uni Research AS, Helse Bergen HF - Haukeland universitetssykehus, Akershus universitetssykehus HF

Prosjektleder: Claire Ghetti

Vi viser til søknad om forhåndsgodkjenning av ovennevnte forskningsprosjekt.

Prosjektleders prosjekttale

Hovedstudien LongSTEP tar sikte på å evaluere effekten av musikkterapi på det emosjonelle båndet mellom premature barn og deres foreldre under sykehusinnleggelse og etter utskrivning fra sykehuset. Studien bygger på en forundersøkelse av LongSTEP utført i Norge, og er forankret i kunnskapshullene som ble identifisert i en tidligere meta-analyse utført av vårt forskerteam, nemlig å vurdere langtidseffekter av musikkterapi.

Deltakere i denne internasjonale randomiserte kontrollerte studien vil bli randomisert til å få: 1) standardbehandling med eller uten musikkterapi under nyfødttintensivbehandling, og 2) standardoppfølging med eller uten musikkterapi i de første 6 månedene etter utskrivning fra sykehuset. Musikkterapitilbudet vil bygge på og stimulere foreldrenes deltakelse, der musikkterapeuten vil gi støtte og veiledning som kan gjøre det mulig for foreldrene å bruke sine egne stemmer og ressurser for å fremme utviklingsmessig tilpasset og samregulert samspill med barna sine.

Vurdering

Vi viser til skjema for tilbakemelding av 28.06.18, vedlagt revidert informasjonsskriv.

REK anser at tilbakemelding er i tråd med de merknader komiteen hadde, samt at informasjonsskrivet har tilstrekkelig og nødvendig informasjon.

Etter fullmakt er det fattet slikt:

Vedtak

Med hjemmel i helseforskningsloven §§ 2 og 10 godkjennes prosjektet.

Sluttmelding og søknad om prosjektendring

Prosjektleder skal sende sluttmelding til REK nord på eget skjema senest 01.10.2023, jf. hfl. §

12. Prosjektleder skal sende søknad om prosjektendring til REK nord dersom det skal gjøres vesentlige endringer i forhold til de opplysninger som er gitt i søknaden, jf. hfl. § 11.

Klageadgang

Du kan klage på komiteens vedtak, jf. forvaltningsloven § 28 flg. Klagen sendes til REK nord. Klagefristen er tre uker fra du mottar dette brevet. Dersom vedtaket opprettholdes av REK nord, sendes klagen videre til Den nasjonale forskningsetiske komité for medisin og helsefag for endelig vurdering.

Med vennlig hilsen

May Britt Rossvoll
Sekretariatsleder

Veronica Sørensen
seniorrådgiver

Kopi til: ingvild.graff@uni.no; britt.torunn.skadberg@helse-bergen.no; britt.nakstad@medisin.uio.no

Appendix 4

Music therapy for premature infants and their caregivers (LongSTEP) – August 2018



REQUEST TO PARTICIPATE IN A RESEARCH PROJECT

«LONGITUDINAL STUDY OF MUSIC THERAPY'S EFFECTIVENESS FOR PREMATURE INFANTS AND THEIR CAREGIVERS (LongSTEP)»

This is an invitation to participate in a research study that is part of an international collaboration between [name of LongSTEP partner], [name of participating NICU, if different from LongSTEP partner], NORCE Norwegian Research Centre AS and University of Bergen, Bergen, Norway. The study aims to provide evidence of the effects of music therapy on bonding between preterm infants and their parents during neonatal hospitalization and during the first six months following discharge. The study will also follow the infant's development across the first two years of life. Within the LongSTEP project, the project team will explore how to support parents in using interaction with their premature infant to help regulate the infant, reduce stress, and promote healthy parent-infant bonding. Some parents will receive support from a music therapist, while others receive support from typical health personnel.

As a parent of a premature infant, you are invited to participate in this study. The LongSTEP study targets premature infants (born prior to 35 weeks gestational age) who are medically stable and will likely be hospitalized in the neonatal intensive care unit longer than 2 weeks from the time of recruitment, and their parents. We seek parents who: 1) are willing to participate in music therapy with their infant, 2) can read and speak either [national language 1] or [national language 2], and 2) live within reasonable commuting distance from the treating NICU.

WHAT DOES THE PROJECT INVOLVE?

If you would like to participate in the study, your participation will vary depending upon the group to which you are assigned. Assignments are random, like flipping a coin. One group of participants will receive standard care, and another group will receive standard care plus music therapy, during your infant's period of hospitalization. Once your infant is discharged to go home, you will again be randomized assigned to one of two groups with different types of follow-up in the first 6 months after you return home from the hospital. One group receives standard follow-up procedures provided by the hospital, and the other receives standard follow-up procedures provided by the hospital plus music therapy.

All participants in the study will receive the full scope of services normally available as a standard part of care in the NICU, and as well as standard follow-up services once they return home.

Participants who are assigned to receive music therapy during hospitalization will participate in short music therapy sessions 3 times per week for approximately 30-40 minutes per session (depending upon your infant's tolerance). It is not necessary to have previous musical experience or abilities. Sessions will consist of the music therapist supporting you to use your voice and body to promote your infant's regulation and support a positive relation. You will learn how to adjust simple melodies in a way that corresponds with your infant's tolerance and contributes to calming your infant.

Participants who are randomized to receive music therapy once they return home, will receive an offer of music therapy sessions 7 times within the first six months upon returning home (two times in the first month

once per month thereafter), for approximately 45 minutes per session. Sessions will occur at home, at a follow-up clinic, or at a location associated with [*insert name of collaboration partner*]. Music therapy sessions will consist of talking briefly about the infant's progress and needs, engagement in simple musical interactions, suggestions for how to use musical interaction to address infant needs, and planning for the next session.

As a participant, you will be asked to anonymously complete questionnaires about your own levels of perceived stress, mental well-being (depression and anxiety), and your perceived relationship to your infant; and answer some basic questions about socioeconomic status. There will also be questions related to your infant's development and abilities. The questionnaires take about 15 minutes to fill out. You will receive 1) questionnaires to complete when you first agree to participate in the study, 2) questionnaires to complete 1-2 days before you are discharged home, and 3) questionnaires to complete when your infant reaches 6 months and 12 months of corrected age. In addition, your infant will receive a developmental assessment at 24 months of corrected age (a process that takes approximately 2 hours), or we will obtain this information from medical records if your child already has completed this evaluation as a part of standard follow-up procedures at your hospital. [*Partner sites may choose to include additional outcomes here, such as qualitative interviews, as part of additional sub-projects*]. We might also contact you at a later point to inform you about the possibility of participating in a follow-up study (for example, when your child reaches 5 years of age) to see how your child has developed over time. You can decide then if you wish you participate in this new study or not.

Participants who are randomized to receive standard care during hospitalization will be asked not to participate in any music-based therapies or music therapy groups or sessions during the hospitalization period. Participants who are randomized to receive standard follow-up services once they have returned home are asked not to participate in any music therapy services (individualized treatment using music with a trained music therapist) during the infant's first year of life. Parents can participate in various infant/parent music groups or otherwise sing or play music with their children.

POSSIBLE BENEFITS AND RISKS

This study can support parents to use their own inherent resources to improve their relationship with their infant through musical interaction. These are techniques and concepts that parents can use over time to promote better relationships with their infant, even when the music therapist is not present.

Premature infants are neurological immature, and therefore there is a risk that any kind of sensory stimulation can overstimulate them. Considering this, the use of music therapy will be carefully adapted to the infant's tolerance for stimulation in the moment, and stimulation will be simplified or stopped depending upon the infant's behaviors and condition. The intervention follows the principles of individual-tailored, developmentally-appropriate care so that stimulation is adapted to the infant's capacity.

Some parents are not comfortable using their singing voices around other adults. It is possible that some parents do not feel comfortable singing to their infants while they are in the hospital.

VOLUNTARY PARTICIPATION AND POSSIBILITY TO WITHDRAW CONSENT

It is voluntary to participate in this project. If you would like to participate with your infant in this project, you can sign the consent statement on the last page of this form. At any time, and without giving any reason, you can withdraw your consent. This decision will not affect the further treatment of your infant. If you withdraw from the project, you may request that collected data is deleted, unless such information is already included in analyses or used in scientific publications. If you later wish to withdraw from the study or have questions about the project, please contact the site investigator [*name of partnering site's site investigator, phone, email*] or principal investigator Claire Ghetti, PhD, +47 465 08 095, claire.ghetti@uib.no.

WHAT HAPPENS WITH YOUR COLLECTED INFORMATION?

We will record certain personal information about your infant from the infant's medical chart (birthdate, birthweight, gender, diagnoses, medication, and information regarding your infant's birth, and at 24 months of age a developmental assessment if applicable). The information that is recorded about you and your infant will only be used for study purposes as described herein.

The developmental assessment at 24 months of corrected age will be videorecorded for quality assurance of the scoring, and if you receive music therapy, your music therapy sessions may be videorecorded for purposes of research quality assessment. These videorecordings are only used by the researchers for research purposes, and will be deleted at the end of the study. In addition, you have an option to consent to the videorecording of music therapy sessions for educational and training purposes. If you consent to optional videorecordings for educational/training purposes, these videos will continue to be used after the study has ended, but your name and the name of your child will not be linked to the recordings. See the specific consent item on the last page.

The information that is collected from you and your infant will be used as described in the study procedures herein. You have the right to see what information is collected about you and your infant, and the right to correct any errors in the information that is registered.

All information will be processed without names and personal identification numbers or other directly identifiable information. A code will link you to your personal information via a separate namelist. This means that your personal information is de-identified. Only authorized study personnel will have access to the list of names which ties you to the gathered study information, and both the primary investigator and the co-researchers are required to maintain confidentiality. It will not be possible to identify individual participants in the results of the study when they are published.

Before discharge, we will ask to record your name and contact information for follow-up purposes (phone number, home address and email-address), and will store this information securely and separately from other collected information.

Information about you and your infant will be de-identified and the namelist will be deleted no later than five years after the end of the project. The exception to this will be if parents have consented to the ongoing use of videorecordings from music therapy sessions for educational or training purposes. Deidentified individual participant data of the main variables of this study will be archived and made available in the Norwegian Centre for Research Data (NSD) public repository. The principal investigator is responsible for the day-to-day running of the research project and that information about you is treated in a confidential and secure manner.

INSURANCE (APPLICABLE ONLY TO NORWEGIAN STUDY SITES)

[Name of participating health institution] ensures that the study participants are covered by insurance under the Patient Injury Act.

ECONOMY

The project is financed by the Research Council of Norway (RCN, project number 273534). Supplemental funding for the project is provided by [Name of hospital affiliated with the participating NICU and name of the LongSTEP partner, if applicable]. Participants will not be paid for their participation in the project.

APPROVAL

The project as a whole has received approval by the Regional committees for medical and health research ethics (REK Nord), Norway [2018/994 REK Nord]. [Sites add their local ethics review board clearance here]

CONSENT TO PARTICIPATE IN THE PROJECT

I AM WILLING TO PARTICIPATE WITH MY CHILD IN THIS PROJECT

Place and date

Participant's signature

Participant's name in capital letters

In addition to videorecording used as part of research study purposes, I consent to the use of videorecordings from music therapy sessions for educational and training purposes.

For project staff to complete:

I confirm that I have given information about the project to the potential participant

Place and date

Signature

Role in the LongSTEP project

Appendix 5

Table 2 Parent Baseline Characteristics

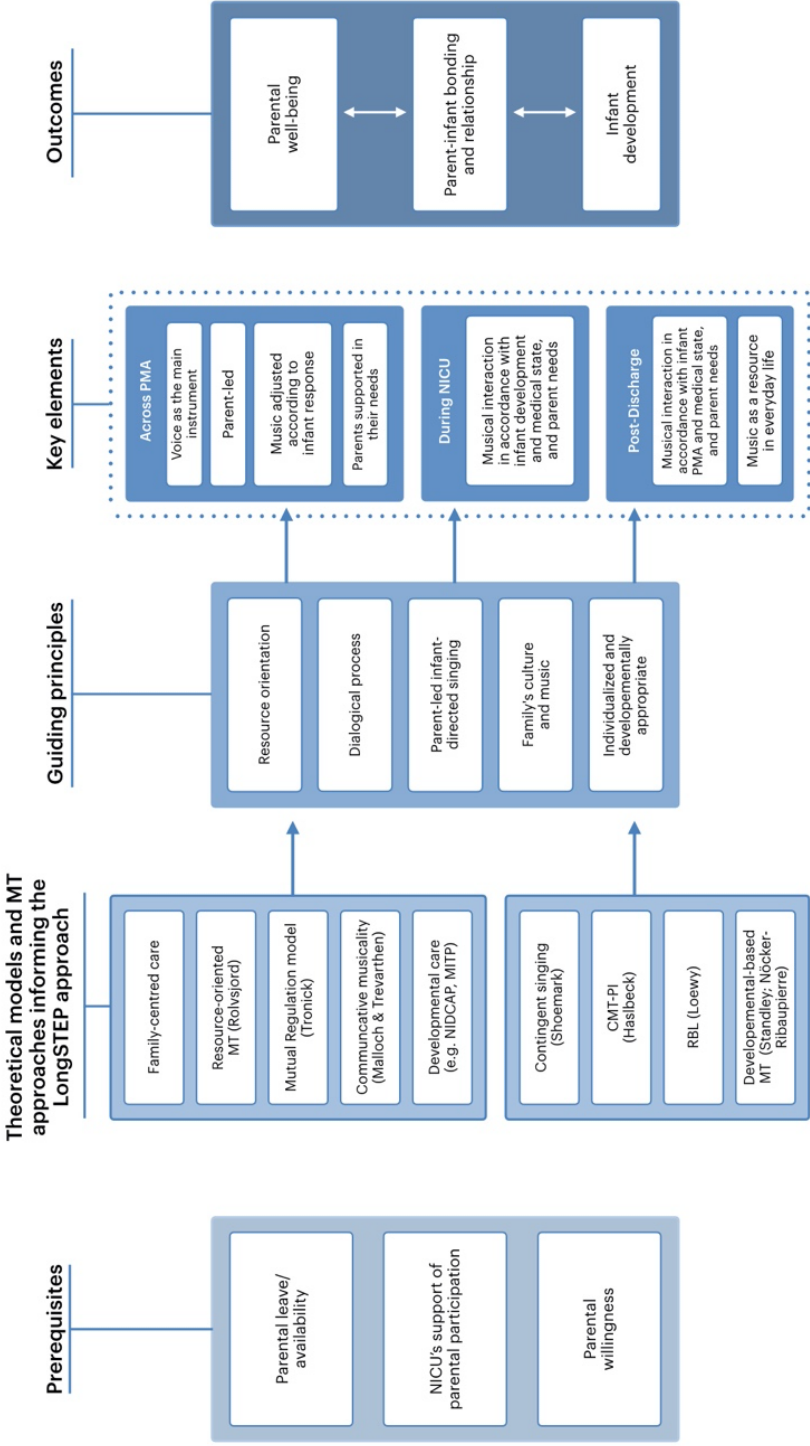
	Standard care		Music therapy		All participants	
	N	Value	N	Value	N	Value
Maternal characteristics						
Age (years) ¹	106	32.77 (5.55) [17, 44]	104	33.11 (5.42) [21, 46]	210	32.94 (5.47) [17, 46]
Education (years) ¹	103	15.77 (2.64) [6, 21]	103	15.85 (3.35) [4, 22]	206	15.81 (3.01) [4, 22]
Usual work situation ²	108		105		213	
Full-time or self-employed		83 (77%)		70 (67%)		153 (72%)
Other ³		22 (23%)		35 (33%)		60 (28%)
Civil status ²	108		104		212	
Married or living together with partner		96 (89%)		98 (94%)		194 (92%)
PBQ bonding score ¹	105		104		209	
Total score		7.08 (6.55) [0, 31]		7.66 (9.05) [0, 62]		7.37 (7.88) [0, 62]
Above factor 1 threshold (≥12) for impaired bonding ²		4 (4%)		5 (5%)		9 (4%)
GAD-7 anxiety score ¹	105	6.10 (5.34) [0, 21]	104	6.39 (4.78) [0, 19]	209	6.24 (5.06) [0, 21]
EPDS postpartum depression score ¹	107	7.95 (5.07) [0, 20]	103	8.47 (5.17) [0, 24]	210	8.20 (5.12) [0, 24]
Paternal characteristics⁴						
Age (years) ¹	101	35.37 (6.45) [22, 56]	99	36.02 (6.29) [21, 57]	200	35.69 (6.37) [21, 57]
Education (years) ¹	97	15.12 (3.21) [6, 22]	96	15.02 (3.77) [3, 25]	193	15.07 (3.49) [3, 25]
Usual work situation ²	101		99		200	
Full-time or self-employed		96 (95%)		90 (91%)		186 (93%)
Other ³		12 (12%)		15 (15%)		27 (14%)
GAD-7 anxiety score ¹	98	4.71 (4.35) [0, 21]	98	6.30 (5.73) [0, 21]	196	5.51 (5.13) [0, 21]

Abbreviations: SD, standard deviation; PBQ, Postpartum Bonding Questionnaire; GAD-7, General Anxiety Disorder-7; EPDS, Edinburgh Postpartum Depression Scale

¹ Mean (SD) [min, max]. ² N (%). ³ Other includes part-time, homemaker/stay-at-home parent, student, unemployed due to ill health or a disability. ⁴ Same-sex parents were invited to participate in the study, but none were enrolled

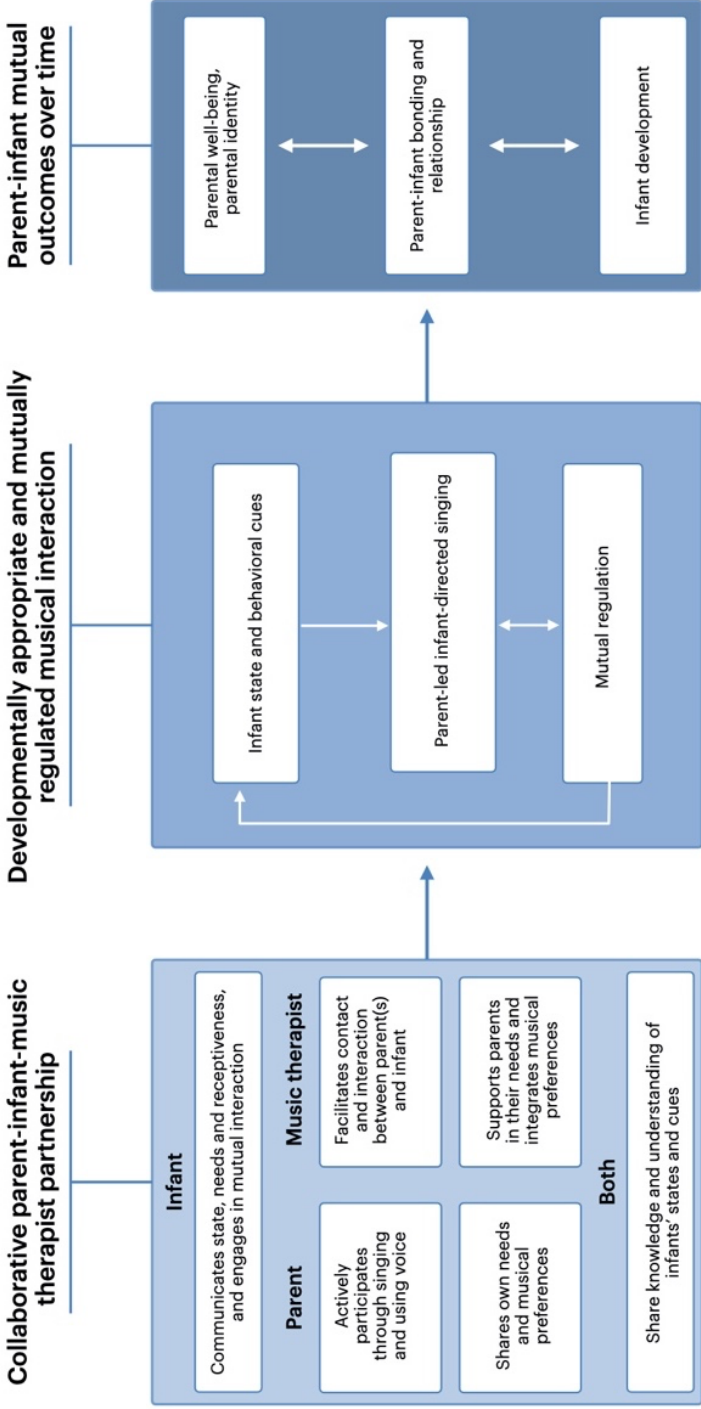
Appendix 6

Conceptual model of the theoretical framework and key elements of the LongSTEP approach, reprinted from Gaden et al., 2021



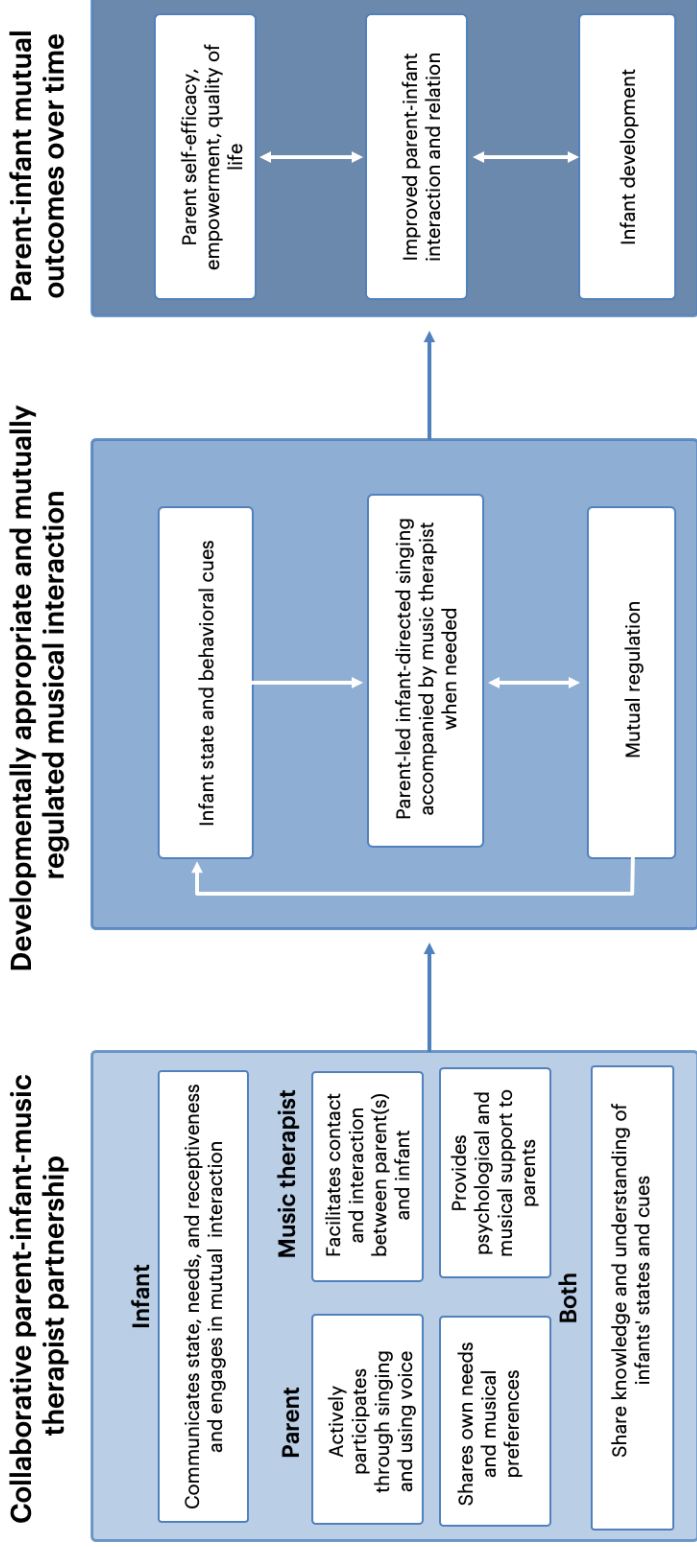
Appendix 7

Model of proposed mechanisms of action of the LongSTEP approach, reprinted from Gaden et al., 2021



Appendix 8

Adjusted model of proposed mechanisms of action of the LongSTEP approach





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