Pilot and Feasibility Studies

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The implementation of EMI-Heart, a family-tailored early motor intervention in infants with complex congenital heart disease, in practice: a feasibility RCT

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Abstract

Background Children with congenital heart disease (CHD) who undergo open-heart surgery are at risk of developmental impairment, including motor delay, which contributes to parental concerns. Additionally, parents experience prolonged stress associated with their child's disease. There is a lack of early motor interventions in infants with CHD accounting for parental burdens. We developed a family-tailored early motor intervention (EMI-Heart), aiming to promote motor development in infants with CHD and family well-being. The primary aim was to evaluate the feasibility of the study design and the intervention. The secondary aim was to evaluate differences between the intervention and the control group in motor outcomes and family well-being at baseline (3–5 months), post-treatment (6–8 months), and at follow-up (12 months).

Method In this single-centre feasibility randomized control trial (RCT), infants with CHD after open-heart surgery without genetic or major neurological comorbidities were randomly allocated to EMI-Heart or the control group (standard of care). EMI-Heart's key elements promote postural functional activities and encourage parental sensitivity to infants' motor and behaviour cues. Infants assigned to EMI-Heart received nine sessions of early motor intervention at home, in the hospital, and online for a duration of 3 months by a paediatric physiotherapist. We performed descriptive statistics for feasibility and secondary outcomes.

Results The recruitment rate was 59% (10/17), all participating families completed the study (10/10), and the intervention duration was 3.9 months (±0.54), including nine intervention sessions per family. Median acceptability to parents was 3.9 (1 = not agree–4 = totally agree, Likert scale). The paediatric physiotherapist considered the intervention as feasible. The comparison of motor outcomes did not show differences between groups. However, we detected improved reliable change scores in family well-being outcomes for families of the intervention group compared to the controls.

Conclusions Our research indicates that EMI-Heart is a feasible intervention for infants with CHD after open-heart surgery. The intervention was highly acceptable both to parents and to the paediatric physiotherapist. Online treatment sessions offer a valuable alternative to home and hospital visits. This feasibility RCT provides a foundation for a future full trial.

Trial registration ClinicalTrials.gov, NCTT04666857. Registered 23.11.2020.

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Keywords Congenital heart disease, Open-heart surgery, Early intervention, Paediatric physiotherapy, Partnering with parents, Video feedback, Online intervention, Family well-being, Feasibility

Key messages regarding feasibility

- What uncertainties existed regarding the feasibility?

  We aim to perform a single-centre feasibility randomized controlled trial comparing a family-tailored early motor intervention to standard of care in infants with congenital heart disease after open-heart surgery. It is unknown if this intervention is feasible to parents and to the paediatric physiotherapist. Further, it is unclear if the outcome measures are feasible tools to assess the secondary outcomes of motor development and family well-being.
- What are the key feasibility findings?
  There was a low recruitment rate, a high adherence rate of participating families and adherence to the protocol expressed by the duration of the intervention and therapy sessions. EMI-Heart, a family-tailored early motor intervention, was feasible with a high acceptability to both parents and to the paediatric physiotherapist. The inclusion of online sessions in the intervention was highly feasible and accepted.
- What are the implications of the feasibility findings for the design of the main study?
  We demonstrated the feasibility of performing a family-tailored earl motor intervention in infants with congenital heart disease after open-heart surgery in a single-centre setting. We also showed that our study design was feasible. A full trial will need to apply a well-chosen selection of sensitive motor and family well-being outcome measures responsive to change.

Background

Congenital heart disease (CHD) is a birth defect that affects approximately 10 out of 1000 live-born children worldwide [1, 2]. Despite advances in prenatal diagnoses and medical care, a significant proportion of infants with complex CHD face an increased risk of a wide range of neurodevelopmental, behavioural, and social difficulties, including language, learning difficulties, perceptual-motor, executive function, and attention problems, some of which may be only detectable later in childhood [3–6]. The first neurodevelopmental impairment that becomes apparent in infants with CHD is motor delay. Studies show that early motor developmental abnormalities persist into adolescence and adulthood [7]. Despite robust evidence of motor impairments in infants with complex CHD [8, 9], no motor intervention has yet been specifically tailored that is specifically tailored to families with infants with a complex CHD. However, a need for early motor intervention and parental support is clearly evident [10–13]. Interventions in infants with CHD that are family-focused and delivered early in life might improve a range of outcomes, including motor and cognitive development [11, 14], compared to interventions mainly focusing on functional motor activities [15].

Early intervention in infants with CHD at risk of motor delay

There is a considerable body of literature that demonstrates the effectiveness of early intervention for infants at high risk of developmental impairments [16–18]. Both infants with CHD who undergo open-heart surgery and infants born very preterm are at high risk of neurodevelopmental impairments in various domains. However, infants with CHD tend to receive fewer therapies than preterm infants, suggesting a lack of awareness regarding the neurodevelopmental challenges they may encounter [19]. A recent systematic review found that few studies have explored early paediatric physiotherapy in infants with CHD, and these have reached inconclusive results [15]. Their findings suggest that early motor interventions may positively impact motor development in infants with CHD, but existing interventions mainly concentrated on strengthening and functional activities, but less so on postural control.

Postural activities in children with CHD

Infants often have difficulties tolerating the prone position after open-heart surgery due to factors such as discomfort following sternotomy with a potentially altered biomechanical muscle force transmission, parental concerns, and lack of opportunity [20]. In addition, infants with CHD often present with generalized muscular hypotonia [21, 22]. Earlier tolerance of the prone position has been associated with better motor skills [23]. Furthermore, early supported and independent sitting are important motor experiences with significant implications for other developmental domains, such as cognition, socialization, and language development [24, 25]. Compared to the supine position, the sitting position extends infants' point of view and enables infants to engage with their surroundings. Increased control of their body against gravity allows infants to use their arms

and hands more freely to explore objects and engage with their caregivers face-to-face [26, 27]. Studies have shown that improving postural control in infants at high risk of developmental delay can facilitate their motor and cognitive development. Additionally, caregivers are more likely to interact and provide learning opportunities to sitting infants [16, 28–30].

Family-focused interventions

Working with parents as equal partners is key for the success of early interventions for parents and their infants. This is particularly important for parents of infants with CHD as they suffer from high level of stress [31, 32]. They are at risk of post-traumatic stress disorder. Stress often persists beyond infants' hospital stays and affects the parent–infant relationship, in particular bonding and attachment [6, 33–35]. Several studies have confirmed that family factors are the most relevant predictors of later neurodevelopmental and psychosocial outcomes of children with CHD [36–38]. Family-focused interventions emphasizing parental attendance and active engagement play a significant role in improving children's outcomes and provide benefits for parental functioning and family quality of life [14, 37, 39–41].

Furthermore, motor delay in infants with CHD can add to existing parental concerns and difficulties in parent–infant attachment. Previous work by our group has emphasized the significance of involving parents as experts in their children's care and including them in decision-making [10]. These findings provided the rationale for developing the Early Motor Intervention (EMI)-Heart, a family-tailored early motor intervention that specifically addresses infants and their families with complex CHD after open-heart surgery.

Methods

We opted to include an RCT in our feasibility study to ensure that the baseline characteristics of the intervention group, EMI-Heart, and the control group, standard of care, were as similar as possible. The protocol of this feasibility study was published in 2022 [42].

Aims

The primary aim of our feasibility RCT was to evaluate the feasibility of our feasibility trial and the feasibility of the intervention EMI-Heart. We measured recruitment and adherence rates predefined by our protocol [42], acceptability of EMI-Heart to parents, fidelity, and feasibility for the paediatric physiotherapist (PT) who provided EMI-Heart in different settings.

Our secondary aim was to compare infant motor and family well-being outcomes between the intervention and control group at baseline (3–5 months of age), post-treatment (6–8 months of age), and follow-up (12 months of age). Our aim was that our results would establish a foundation for a full trial to evaluate the effectiveness of EMI-Heart in infants after open-heart surgery.

Study design and setting

This prospective, single-centre, single-blinded, two-arm parallel feasibility RCT compared EMI-Heart to standard of care in infants with CHD after open-heart surgery at the University Children's Hospital Zurich, Switzerland. Infants in need of cardiac surgery were screened between May 2021 and June 2022.

This study adhered to the TIDieR [43] and the CONSORT statement [44].

Study participants and recruitment procedure

We included infants with the following criteria: (1) diagnosed with CHD; (2) undergoing a single open-heart surgery with sternotomy and cardiopulmonary bypass (CPB) within the first 5 months of life regardless of existing motor delay; (3) born $\geq$ 37 weeks gestational age; (4) discharged home before the age of 6 months; (5) living within a 1-h-travel distance from the Children's Hospital; and (6) the informed consent of infants' parents documented by signature. We excluded infants (1) with univentricular heart defects, because they require at least one additional open-heart surgery within the first year of life, and thus therapy could not be delivered as intended; (2) with known genetic diagnoses or genetic syndromes known to be associated with adverse neurodevelopmental outcomes; (3) with large cerebral and clinically manifest lesions; and (4) whose parents had insufficient command of the German language to understand the patient information. Children undergoing CPB without a sternotomy were also excluded because we sought infants with the highest likelihood of benefitting from PT.

Members of the Department of Cardiology and the investigator and co-author EM screened and recruited infants at the University Children's Hospital Zurich between May 2021 and June 2022. The last follow-up assessment was completed in December 2022.

Infants were assigned to the intervention or the control group by a computer-generated random sequence. Group allocation was concealed. Sealed opaque and numbered envelopes were opened sequentially by members of the Children's Hospital that were not involved in this project to preserve confidentiality. Members of the Child Development Center and EM assessed all infants of the intervention group (IG) and the control group (CG) at baseline after hospital discharge (T0), at post-treatment (T1), and at follow-up (T2) at 12 months (see Table 2) to minimize inter-rater variability. All motor assessments were standardized, video recorded and

scored by assessors blinded to group allocation independently. Parents completed parental questionnaires electronically at all time-points. Parents and the paediatric physiotherapist (PT), who administered EMI-Heart were not blinded to the IG or CG. All infants received a small toy as a gift (e.g. baby rattle) for participating in the trial.

Study groups

Intervention group (IG): EMI-Heart

Besides being a tertiary care centre, our hospital also provides primary care regarding outpatient physiotherapy. Infants assigned to the IG received EMI-Heart. EMI-Heart started after hospital discharge at T0 and took place once a week or fortnightly for 45–60 min per session. The intervention comprised nine treatment sessions: three sessions at home, three at the hospital, and three online, in an alternating sequence. We incorporated online sessions as a variable option to visits at home and at the hospital.

Co-author EM contacted parents regularly via phone, text messaging, and email to maintain study adherence. EM, a senior paediatric PT of the outpatient team with extensive experience in early intervention, also provided all intervention sessions to maximize intervention fidelity. All interventions were video recorded. Co-author TD, a senior tutor and paediatric PT with extensive experience in early intervention [45, 46], discussed all videos with EM. They reflected regularly on the transactions between parents, infant, and the PT. Transactions are the process in which parents, infant, and the PT create relationships and engage with each other in dialogue to stimulate postural activities in various positions.

To ensure the quality of the intervention and to guarantee fidelity of the PT to elements of EMI-Heart, TD provided EM with regular video-based feedback using the videos of intervention sessions. The rationale and key elements of EMI-Heart were derived from our previous qualitative study examining the burdens on and needs of parents with a child with a complex CHD [10]. Parents wished to support their infants and be actively involved in their development and acknowledged as experts in their own children. EMI-Heart is tailored to each family, considers families' wishes and experiences, and is adapted to infants' motor abilities. EMI-Heart's key elements are (1) promotion of infants' postural activities and (2) partnering with the parents, as described in detail below. Both elements are closely intertwined (see Fig. 1).

The elements of EMI-Heart are described below.

Elements of EMI-Heart

1. Promotion of infants' postural activities

   The PT creates safe and playful postural activities in prone position and early sitting and supports parents in observing, exploring, and stimulating their infant's activities. Parents and PT exchange their perspectives and have face-to-face contact with the infant to stimulate the infant's interest and facilitate challenging new postural activities. The turn-taking of all the adults involved is a continuous process of dynamic transactions between infants, parents, and the PT. Parents experience iteratively how to understand and respond to infant's cues and activities joyfully in daily life. Prone position and supported sitting are adjusted to infants' needs. External support such as cushions, towels, parents' hands and/or body, furniture, and toys are used to stimulate head and trunk control, reaching, and grasping activities [42]. Parents and PT gradually decrease external support as the infant's postural control improves, such as lifting the head more easily in prone position, looking around with greater ease, showing enjoyment, and goal-directed reaching and grasping in a goal-directed way. Figure 1 illustrates the dynamic process of transactions in which parents, infant, and the PT create relationships and engage with each other in dialogue to stimulate postural activities in various positions.
2. Partnering with parents

   Partnering in EMI-Heart is a dynamic transactional process between parents and the PT to promote parental sensitivity to infants' motor and behaviour cues.

   Parents and the PT meet as equal partners, discuss their perspectives openly, and respect each other's expertise. Parents share their expertise on their infant, family history, rituals and routines, and uncertainties. The PT shares their professional knowledge and current research evidence.

   2.1. Parents' attendance and active engagement

   Parents are present during each EMI-Heart intervention in the hospital, at home, and online. Parents' attendance and active engagement provide a trustworthy and playful environment for the infant. This is a prerequisite to beginning the partnering process. Parents and PT observe, explore, and stimulate infant's postural activities together. Parents make video recordings of how they implement the intervention at home. The PT applies various strategies focused on infant's well-being, parent's confidence, and empowerment.

   2.2. Encouragement of parents' confidence and family well-being

   a Promoting parental confidence

   Following open-heart surgery, parents often feel insecure about holding and carrying their infants and are hesitant to attempt new posi-

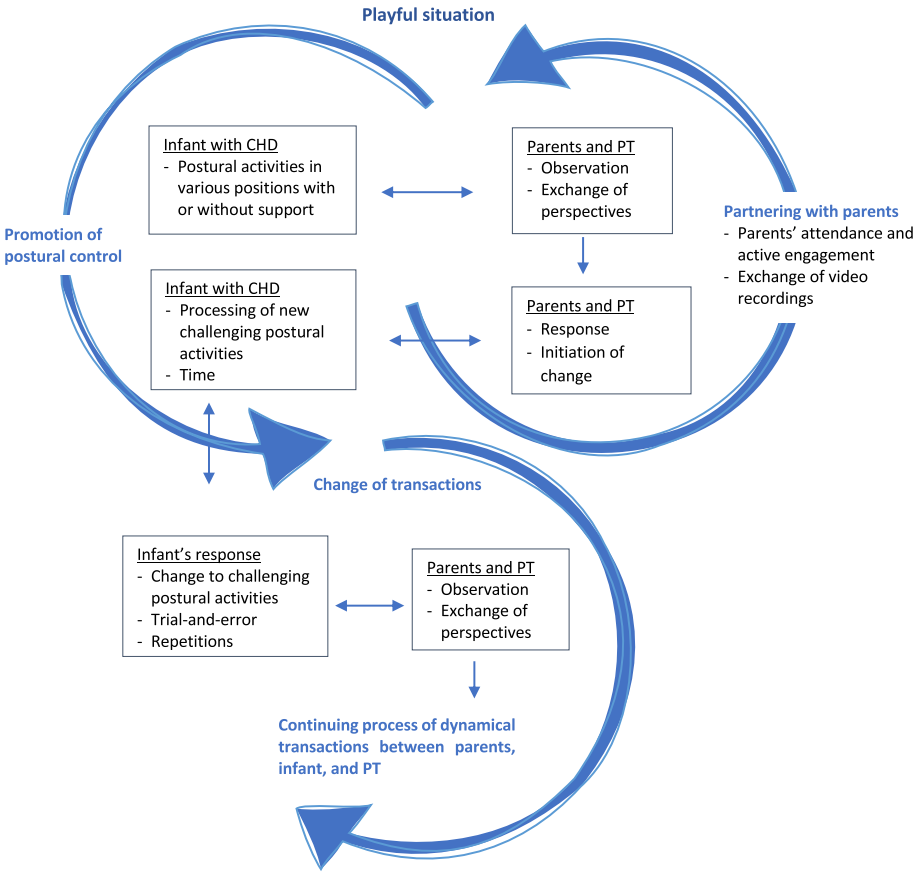

Fig. 1 EMI-Heart, a transactional model of change. The spiral illustrates the dynamic process of transactions in which parents, infant, and the PT create relationships and engage with each other in dialogue to stimulate postural activities in various positions; CHD congenital heart disease, PT physiotherapist

tions such as the prone position and early sitting [10, 15, 20]. To promote parental confidence, the PT and parents try out how they can place their infants comfortably in prone position and early sitting. The PT supports parents' activities and encourages parents to trust their infants' and their own capacities.

b Promoting the parent–infant relationship

EMI-Heart emphasizes the importance of the parent–infant relationship in the vulnerable phase of infants' illness following surgery.

Face-to-face contact in prone position and early sitting, with or without support, provokes turn-taking and a joyful interplay between parents and infants. Communication with infants during the intervention supports the parent–infant relationship. Parents use baby-talk, facial expressions such as smiling and laughing, and gestures adjusted to their own way of parenting.

c Exchange of video recordings and video feedback

The PT shares video recordings with parents that were made during the intervention. Likewise, parents are invited to exchange their own video recordings when implementing EMI-Heart during daily life. The PT gives feedback on parental video recordings. This exchange promotes active engagement and equal partnering. Reviewing video recordings enables parents and the PT to observe and reflect on their own behaviour and its effect on transactions with the infant. This provides insight into what is happening during the intervention. Additionally, parents' video recordings allow the PT to see how parents find their own creative solutions to implementing EMI-Heart in daily-life rituals and routines.

d Online sessions

Online sessions encourage parental engagement and refine communication between parents and the PT when the PT is unable to interact with the family in person. Parents

demonstrate online how creative they are in stimulating their infants in natural situations. They give the PT insight into what the parents have learnt during the intervention. The PT provides feedback and answers questions. Furthermore, the PT plays these recordings back to the parents at various points to show parents how they are doing.

Control group (CG): standard of care

After hospital discharge, infants assigned to the control group received standard of care, including cardiac surveillance, counselling, detailed neurodevelopmental assessments at the Child Development Center of the University Children's Hospital, as well as well-baby visits with their paediatrician. Usually, outpatient paediatric PT is not part of follow-up care in patients with complex CHD undergoing open-heart surgery once within the first year of life. However, some infants might receive outpatient PT if they demonstrate significant neurological abnormalities or motor developmental delay.

Outcome measures

Our primary outcome was the feasibility of the EMI-Heart trial. Feasibility was measured by clinical recruitment rate, adherence of participating families, adherence to the protocol, acceptability of EMI-Heart to parents, and feasibility for the paediatric physiotherapist providing the intervention (see Table 1). We assessed the acceptability of EMI-Heart to parents using an online questionnaire at T1. The questionnaire consisted of 18 items on a Likert scale (1 = not agree–4 = totally agree). The acceptability questionnaire was adapted from Modi et al. [47] and reflects the elements on which EMI-Heart is based (Supplementary Material 1). It was evaluated by MT, a parental stakeholder, and experts in early infant development and early intervention before the intervention started.

Our secondary outcomes were measured by infants' motor assessments and questionnaires about family well-being (see Table 2). We included multiple outcome measures to determine which might be the most feasible and sensitive ones for a future full trial.

Motor assessments consisted of the General Movement Assessment (GMA) including the Motor Optimality Score (MOS-R) [48, 49] and the Hammersmith Infant Neurological Examination (HINE) [50, 51] as baseline variables. The Infant Motor Profile (IMP) [52] and the Alberta Infant Motor Scale (AIMS) [53] were assessed at all time-points (T0, T1, T2), the Bayley motor domains of the Bayley Scales of Infant and Toddler Development (BSID III) [54] at T2. All motor assessments were video recorded and scored by assessors blinded to group allocation. Self-reported parental questionnaires evaluated parents' and infants' health-related quality of life [55–57], parental mental health [58] and stress perception [59], family empowerment [60], and parental protection [61]. Socioeconomic status (SES) was derived from a six-point scale of maternal and paternal education with a range from 2 to 12 (1 = special school–6 = higher university education) [62]. We used a survey with research electronic data capture [63, 64] hosted at the University Children's Hospital Zurich. Medical information was obtained from the hospital's electronic medical charts through its data management system.

Patient and public involvement

The Swiss Parents' Association for Children with Heart Disease provided advice and guidance for this study, with co-author MT as a parental stakeholder. The elements of EMI-Heart were developed after interviews with parents of infants with CHD who underwent open-heart surgery. These interviews identified the burdens and needs that EMI-Heart seeks to address [10]. Families were informed about the intervention's requirements and could withdraw at any time. Once

Table 1 Feasibility outcomes

Recruitment rate	**- *n* of infants eligible for recruitment**
Adherence of participating families	- *n* of infants per intervention and control group - *n* of dropouts
Adherence to the protocol	- Overall duration of intervention in months, mean (SD) - *n* of sessions per intervention family
Acceptability of EMI-Heart to parents assessed post-treatment (T1)	- Acceptability questionnaire, median (range) 18-item Likert scale (1 = not agree–4 = totally agree) - Parents' attendance per family (*n* of sessions) - *n* of video recordings sent per family, median (range)
Feasibility for the PT providing EMI-Heart	- Subjective evaluation

n number, *EMI* Early Motor Intervention-Heart, *PT* physiotherapist

Table 2 Secondary outcomes

Motor assessments		Baseline T0	Post-treatment T1	12-month follow-up T2
General Movement Assessment incl. Motor Optimality Score [48, 49]		x		
Hammersmith Infant Neurological Examination [50, 51]		x		x
Infant Motor Profile [52]		x	x	x
Alberta Infant Motor Scale [53]		x	x	x
Bayley Scales of Infant and Toddler III—motor domain [54]				x
Parental questionnaires assessing family well-being				
Infants' quality of life	Pediatric Quality of Life Inventory Infant Scales [55]	x	x	x
Parents' quality of life	Short Form Survey 36 [56, 57]	x	x	x
Parental mental health	Brief Symptom Inventory 18 [58]	x	x	x
Parental stress experience	Parental Stress Index [59]	x	x	x
Parental empowerment	Family Empowerment Scale [60]	x	x	x
Parental protection	Parental Overprotection Measure [61]	x	x	x

data analyses were finished, eligible families received both individual and general reports of the results.

Statistical analysis

The study's primary aim was to determine the feasibility of the EMI-Heart trial and establish a foundation for a larger RCT. This study was not designed for inferential analyses and was not intended to be generalizable to a wider population. We aimed for a sample size of 16 infants. This number was determined by examining clinical data from the University Children's Hospital [65] and prior intervention studies [14] and represented about 30% of infants who had had open-heart surgery within the first 5 months of life over the last 3 years. A member of our research team not involved into the trial checked data entry and descriptive data analysis. We analysed data using R (version 4.2.2) [66] and performed descriptive statistics for feasibility and secondary outcomes at all time-points. We did not perform inferential analyses for secondary outcomes due to the small sample size of the study. However, we calculated reliable change scores [67, 68] of the intervention and control group and of infants individually between T0 and T2 to determine whether scores changed sufficiently that the change was unlikely to be due to simple measurement unreliability.

Results

Study participants and recruitment

Of 88 infants with CHD assessed for eligibility, 71 did not meet inclusion criteria (see Fig. 2 flow diagram). Among the 17 eligible families, 7 families declined to participate, for a variety of reasons including long travelling times, burden of managing childcare, and psychological distress. This left 10 families who participated and were randomized.

Participants' characteristics including infant, cardiac, and parental variables are depicted in Table 3.

Table 3 presents baseline characteristics of included infants. Despite the use of an RCT design, characteristics of the two groups were different. Specifically, four out of five infants of the control group (CG) received outpatient physiotherapy which was prescribed by physicians prior to discharge. Further, two infants in the intervention group (IG) additionally underwent open-heart surgery during the EMI-Heart intervention, while the CG had no cardiac intervention. Maternal age and parental socioeconomic status were also lower in the IG compared to the CG. Additional there were differences between groups regarding median gestational age, age at CPB surgery, ICU, and hospital stays.

Results of the primary feasibility outcomes

Feasibility outcomes are depicted in Table 4. Adherence rate was 100% and acceptability of EMI-Heart to parents was very good. It is noteworthy that families in the intervention group sent on average 16 video recordings to the PT to show how they implemented the intervention.

Acceptability of EMI-Heart to parents

Results of the acceptability questionnaire (Supplementary Material 1) showed that parents in the intervention group highly appreciated suggestions about how to support their infants' motor development in everyday life (median score 3.9 (range 3 to 4)) (see Table 4). They experienced the PT's inputs as easy to understand and to implement. Parents perceived themselves as equal partners and liked sharing their ideas with the PT. Parents stated that EMI-Heart improved their understanding how to promote infants' motor and behaviour cues

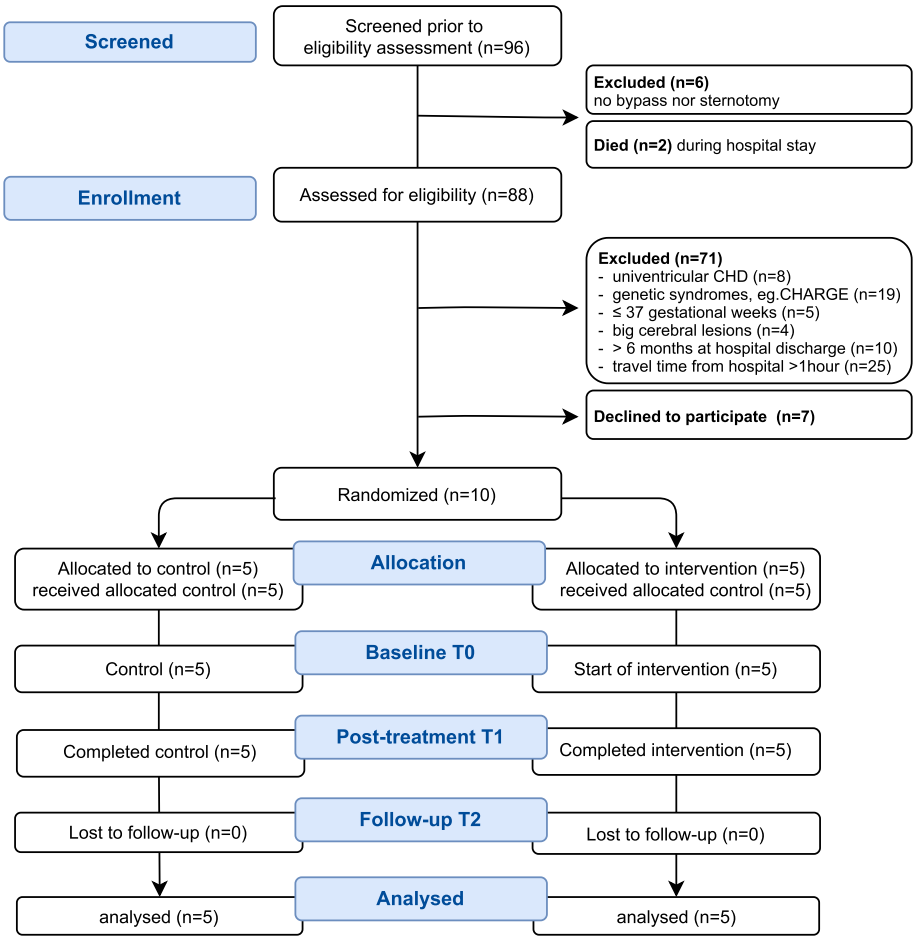

Fig. 2 Flow diagram of the study procedure according to Consort 2010

and that they still implemented elements of EMI-Heart in daily life after the end of the intervention. Moreover, they reported that they trusted their infants more, allowed them to experiment with early sitting, and that EMI-Heart had improved their infants' development. Exchanging video recordings helped parents to share their ideas and see their infants from a different perspective. All parents appreciated home visits and online sessions. Three families wished that EMI-Heart had lasted longer than nine sessions. All parents recommended EMI-Heart to other families with infants with complex CHD.

Feasibility EMI-Heart applied by the paediatric PT

After completion of all intervention sessions, the PT, co-author EM, carefully reflected on various factors involved in EMI-Heart. The findings of the PT are described in an overview below (see Table 5). Overall, the PT considered that performing EMI-Heart was feasible.

Results of the secondary outcomes

Secondary outcomes were measured by infants' motor scores and family well-being scores (see Table 2). Due to the small sample size, we performed descriptive statistics and compared our result to normative data where available. We additionally calculated reliable change scores of the intervention and control group and of infants individually between T0 and T2. Results of motor outcomes and family well-being outcomes are depicted in Tables 6 and 7.

Motor outcomes

At baseline (T0), motor outcomes of both groups were similar as measured by the General Movement Assessment (GMA) including the Motor Optimality Score (MOS-R) [48, 49], the Infant Motor Profile (IMP) [52], and the Alberta Infant Motor Scale (AIMS) [53]. Descriptive statistics of motor outcomes and reliable change scores are presented in Table 6. Overall, motor

Table 3 Participants' characteristics

Study groups	EMI-Heart group	Control group
Intervention, *n*	5	5
Outpatient physiotherapy		4
Sex, female, *n*	3	4
Gestational age in weeks, median (range)	38.6 (36.4 to 40)	41.1 (36.6 to 41.9)
Birth weight in grams, median (range)	2680 (2510 to 3500)	3200 (2490 to 4100)
5-min Apgar, median (range)	9 (8 to 10)	6 (5 to 9)
Acyanotic CHD, *n*	2	3
VSD	1	2
ASD and aortic isthmus stenosis		1
ALCAPA, corona anomaly	1	
Cyanotic CHD, *n*	3	2
TGA	2	
Truncus arteriosus communis		1
Pulmonary vein malposition	1	
Pulmonary atresia		1
Age at surgery in days, median (range)	28 (3 to 144)	18 (4 to 97)
CPB time in minutes, median (range)	328 (92 to 409)	230 (106 to 326)
Length of ICU stay in days, median (range)	12 (2 to 32)	17 (4 to 44)
Length of hospital stay in days, median (range)	14 (6 to 34)	19 (9 to 97)
Postoperative need for ECMO	2	2
Additional open-heart surgery during intervention time	2	
Time in days between surgery and start of intervention, median (range)	89 (23 to 117)	
Infants' age in months, median (range) at		
Baseline T0	4.3 (4.0 to 5.6)	4.4 (3.2 to 5.5)
Post-treatment T1	9.0 (7.7 to 9.5)	8.1 (7.2 to 9.6)
12-month follow-up T2	14.3 (11.9 to 16.0)	14.5 (13.6 to 16.1)
Parental variables		
Age mother in years, median (range)	29 (24 to 32)	36 (34 to 43)
Age father in years, median (range)	31 (24 to 37)	35 (33 to 49)
SES*, median (range)	6 (6 to 10)	10 (7 to 12)

CHD congenital heart defect, *VSD* ventricular septal defect, *ASD* atrial septal defect, *TGA* transposition of the great arteries, *CPB* cardiopulmonary bypass, *ICU* intensive care unit, *ECMO* extracorporeal membrane oxygenation, *T0* baseline, *T1* post-treatment, *T2* 12-month follow-up

* Socioeconomic status (SES), maternal and paternal education

scores increased with age for infants in both groups. Accordingly, scores at 12-month follow-up (T2) were similar for the IMP, and the AIMS.

However, the intervention group (IG) had a lower AIMS score than the control group (CG).

General Movement Assessment including the Motor Optimality Score (MOS-R)

All infants showed fidgety movements at T0 and presented with a MOS-R scores of 20 or 21. Regarding the MOS-R, we observed a combination of atypical movement patterns (e.g. atypical mouth, kicking, and foot-to-foot), atypical postural patterns (e.g. body or head asymmetry, retroflexion of trunk and shoulder girdle, extension of arms, and atypical finger postures), and atypical movement characters (e.g. jerky, tremulous, and/or monotonous). Research has shown that normal fidgety movements in combination with a MOS-R of < 21 can indicate an increased risk of future adverse neurodevelopment in gross and fine motor performance and cognitive and/or language skills at school age [69, 70].

Hammersmith Infant Neurological Examination (HINE)

Two infants (one per group) could not complete the HINE assessments at T0, and one infant of the group CG at T2 due to irritability. At T0, HINE global median scores of the IG were larger compared to those of the CG. The median score of both groups at T0 was 58.4, and 68.4 at T2, corresponding to a suboptimal score according to Romeo et al. [71]. The median score of the reference population of typically developing infants at

Table 4 Feasibility outcomes

Recruitment rate	**59.9% (10/17)** **Reasons for decline: long travel-time, burden of managing childcare, psychological distress**
Adherence of participating families	IG (5/5), CG (5/5)
Drop out	IG (0/5), CG (0/5)
Adherence to the protocol	
Overall duration in months, mean (SD)	3.9 (± 0.54)
n of sessions per IG family	9* (3 × home, 3 × hospital, 3 × online)
Acceptability of EMI-Heart to parents	
Acceptability questionnaire, median (range), 1 = not agree–4 = totally agree	3.9 (3 to 4)
Parents' attendance, *n* of sessions	100% (9/9)
n of video recordings sent per family, median (range)	16 (9 to 20)
Feasibility for the PT providing EMI-Heart	See descriptive overview below

IG intervention group, *CG* control group, *n* number, *PT* physiotherapist

* 9 intervention sessions attended per family

Table 5 Feasibility for the paediatric PT

Home
i. The PT experienced that travelling was time consuming as she used public transport. One home visit generally lasted 3 h
ii. Scheduling home visits required flexibility from the PT
iii. Initially, the PT felt intrusive when visiting families at home but quickly experienced the advantages in gaining insight into families' natural surroundings. This enabled the use of the families' own furniture and infants' toys and allowed direct observation of how parents implemented EMI-Heart
iv. The PT experienced home visits as less formal and more trustful
Hospital
i. The PT did not spend time travelling
ii. This setting enabled the PT to use a range of various equipment and toys adjusted to infants. This allowed parents to gain new ideas
iii. Appointments were combined with cardiac check-ups whenever possible. The PT observed that families did not mind coming to the hospital
iv. The PT observed that most of the hospital visits were attended only by mothers
Online
1. The PT did not spend time travelling
2. It was easy to make appointments at times when both parents could be present
3. The PT and parents used digital media without difficulties
4. The PT observed that parents interacted with their infants more actively online than meeting live as the PT could not interact physically
5. The PT saw how parents stimulated and played with the infant in their natural surroundings
6. During online sessions, the PT shared video recordings with parents to discuss and reflect on interactions with their infant
Video recording, exchange of video recordings, and video feedback
i. The PT was able to video record all intervention session using a tripod
ii. Video recordings gave the PT insight on how parents applied given suggestions and about parents' own ideas in real life situations
iii. The PT received many more parental video recordings than expected
iv. The PT shared video recordings to give parents the possibility to review the intervention from another point of view

PT physiotherapist

9 months of age is 72 (65 to 78). Reliable change was not reported as there were no age-independent norm data available.

Infant Motor Profile (IMP)

Trajectories of both groups progressed similarly from T0 to T2 (see Fig. 3). Reliable improvement was detected in mean scores in both groups and in all infants individually from T0 and T2 (see Table 6). IMP scores of both groups were between the 15th and 50th percentile at T0 and around the 15th at T2 when compared to the Dutch norms [52]. This finding might be explained by the fact that all study infants had lower median scores in the performance, fluency, and

Table 6 Motor outcomes

	Median (range)		Baseline (T0)	Post-treatment (T1)	12-month follow-up (T2)	RC group	RC infant
Motor outcomes	GMA incl	IG	21 (20 to 21)	x	x	–	–
	MOS-R	CG	21 (20 to 21)				
	HINE	IG	58.6 (50 to 62)	x	69.7 (66.7 to 77)	–	–
		CG	52 (43 to 62.4)		67.5 (67 to 68.6)		
	IMP	IG	77 (76 to 78)	88 (84 to 96)	92 (85 to 98)	✓	5x ✓
		CG	75 (64 to 78)	86 (81 to 88)	90 (86 to 96)	✓	5x ✓
	AIMS	IG	8 (6 to 10)	18 (15 to 33)	47.5 (39 to 57)	✓	5x ✓
		CG	9 (6 to 10)	28 (17 to 32)	58 (40 to 58)	✓	5x ✓
	BSID III motor	IG	x	x	82 (55 to 122)	–	–
		CG			106 (64 to 116)		

GMA MOS-R General Movements Assessment incl. Motor Optimality Score, *HINE* Hammersmith Infant Neurological Examination, *IMP* Infant Motor Profile, *AIMS* Alberta Infant Motor Scale, *BSID III* Bayley Scales of Infant and Toddler Development, *IG* intervention group, *CG* control group, *RC* note: the higher the scores, the better the motor performance; – does not apply; reliable change; ✓ = RC + (reliable improved); 0 = RC0 (indeterminate change); X = RC – (reliable deteriorated)

Table 7 Family well-being outcomes

	Median (range)		Baseline T0	Post-treatment T1	12-month follow-up T2	RC group	RC infant
Family well-being outcomes	PedsQL the higher, the better	IG	68.1 (57.6 to 80.6)	78.5 (63.2 to 87.5)	77.8 (57.6 to 86.1)	0	1x ✓ 4 × 0
		CG	70.1 (49.3 to 93.6)	68.8 (57.6 to 99.3)	72.9 (63.9 to 93.1)	0	1x ✓ 4 × 0
	SF-36-MH the higher, the better	IG	51.0 (8.0 to 51.6)	41.6 (26.1 to 59.1)	52.7 (33.0 to 54.9)	0	1x ✓ 4 × 0
		CG	47.2 (38.6 to 60.3)	38.1 (7.6 to 57.5)	42.7 (16.1 to 53.0)	0	3 × 0 2 × X
	BSI-18 the lower, the better	IG	7 (3 to 34)	9 (0 to 10)	3 (0 to 16)	✓	3x ✓ 2 × 0
		CG	4 (0 to 9)	2 (0 to 33)	6 (0 to 35)	X	3 × 0 2 × X
	FES the higher, the better	IG	52 (46 to 60)	54 (40 to 60)	53 (48 to 57)	–	–
		CG	51 (39 to 59)	47 (36 to 55)	45 (43 to 60)		
	PSI the lower, the better	IG	119 (155 to 139)	118 (78 to 158)	103 (89 to 172)	0	2x ✓ 2 × 0 1 × X
		CG	123 (65 to 166)	126 (80 to 154)	128 (76 to 151)	0	4 × 0 1 × X
	POM the lower, the better	IG	36 (21 to 55)	39 (10 to 54)	40 (19 to 52)	–	–
		CG	42 (25 to 48)	36 (23 to 47)	33 (23 to 40)		

PedsQL Pediatric Quality of Life Inventory Infant Scales, *SF-36-MH* Quality of Life Short Survey-Mental Health, *BSI-18* Brief Symptom Inventory, *FES* Family Empowerment Scale, *PSI* Parental Stress Index, *POM* Parental Overprotection Measure, *IG* intervention group, *CG* control group, *RC* reliable change; ✓ = reliable improved; 0 = indeterminate change; X = reliable deteriorated; – does not apply

variation domain of the IMP than the Dutch norms. Research has shown that lower IMP scores in low-risk infants were associated with lower IQ scores at 4 years of age and neurological cognitive, and behavioural function at school age [72].

Alberta Infant Motor Scale (AIMS)

Both groups started at a similar level and improved until T2. However, scores of the IG increased less compared to scores of the CG from T0 to T2. Reliable improvement was detected in mean scores in both groups and in all infants individually from T0 and T2 (see Table 6). Median scores of all infants were at the 10th percentile at T0 compared to the Canadian norms [53]. At T2, the median score of the intervention group was ≤ 10th percentile and > 75th percentile of the control group. AIMS scores of two infants (one per group) could not be included due to irritability at T2.

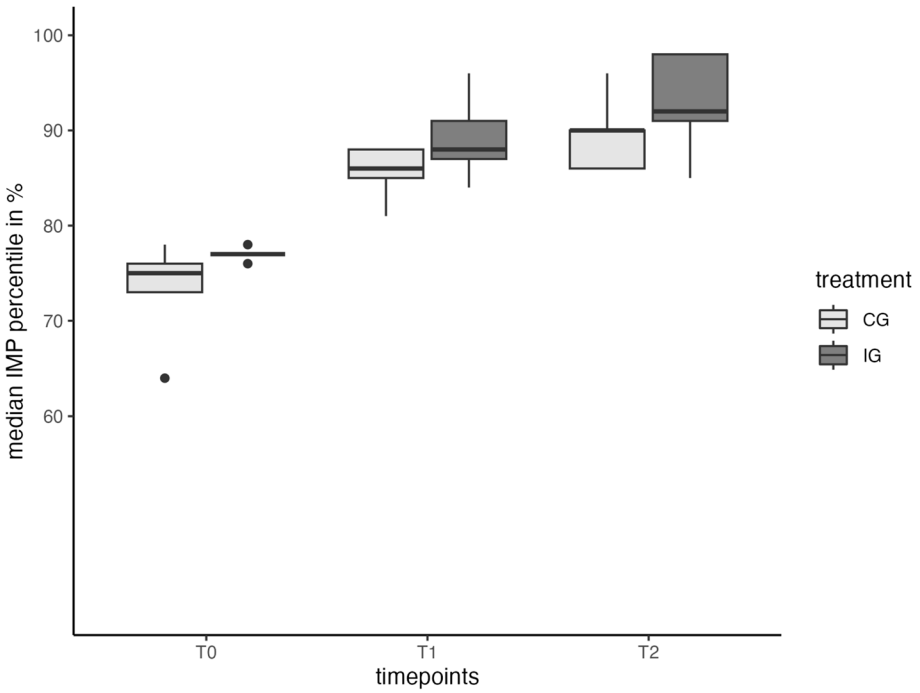

Fig. 3 Results of the Infant Motor Profile for the intervention (IG) and control (CG) groups

Bayley Scales of Infant and Toddler Development (BSID III)

Median BSID-III motor composite scores of the intervention group were slightly below the norm (median 82, range 55 to 122), while the control group (median 106, range 64 to 116) ranged slightly above the test norm mean of 100 (SD of 15) [54]. Motor scores of the control group were higher compared to results of other studies in patient group that were performed in our hospitals with a larger sample size. Feldmann et al. [73] and Meuwly et al. [74] reported a mean motor composite score of 90.36 (SD 13.94) and of 92.7 (SD 15.4) respectively at 12 months for infants with CHD and open-heart surgery. We only assessed the BSID III once; thus, we did not calculate reliable change.

Family well-being outcomes

Family well-being outcomes are depicted in Table 7. At T0, groups had similar outcomes representing family well-being measured by the Pediatric Quality of Life Inventory Infant Scales [55], Parental Stress Index [59], and Family Empowerment Scale [60].

We calculated reliable change of the intervention and control group and of infants individually between T0 and T2 (see Table 7).

Pediatric Quality of Life Inventory Infant Scales (PedsQL)

Median PedsQL scores of the IG showed a larger increase compared to the CG from T0 to T2. However, there was no reliable change in mean scores in either of the two groups from T0 to T2. Reliable improvement was detected in one family of each group.

Quality of Life Short Form-Mental Health (SF-36-MH)

The SF-36-MH scores of parents of the IG remained similar between T0 to T2. In contrast, scores of parents of the CG decreased. Scores of both groups dropped at T1 and increased again at T2. There was no reliable change in mean scores in either of the two groups from T0 to T2. Whereas reliable improvement was detected in one family of the IG, reliable deterioration was detected in two families of the CG. Compared to Swiss norms of a mean score of 77.47 [57], SF-36-MH in our sample were clearly below at all time-points.

Brief Symptom Inventory (BSI-18)

BSI scores of parents of the IG decreased from T0 to T2, whereas scores of parents of the CG increased (see Fig. 4). Importantly, the range of BSI scores was large, indicating a large variability in parental stress symptoms. Reliable improvement was detected in mean scores in the IG, whereas reliable deterioration detected in mean scores in the CG from T0 to T2. Reliable improvement was detected in three families of the IG; reliable deterioration was detected in two families of the CG. Compared to the German norms [58] with a mean score of 4.66, 50% of the total sample had a higher score.

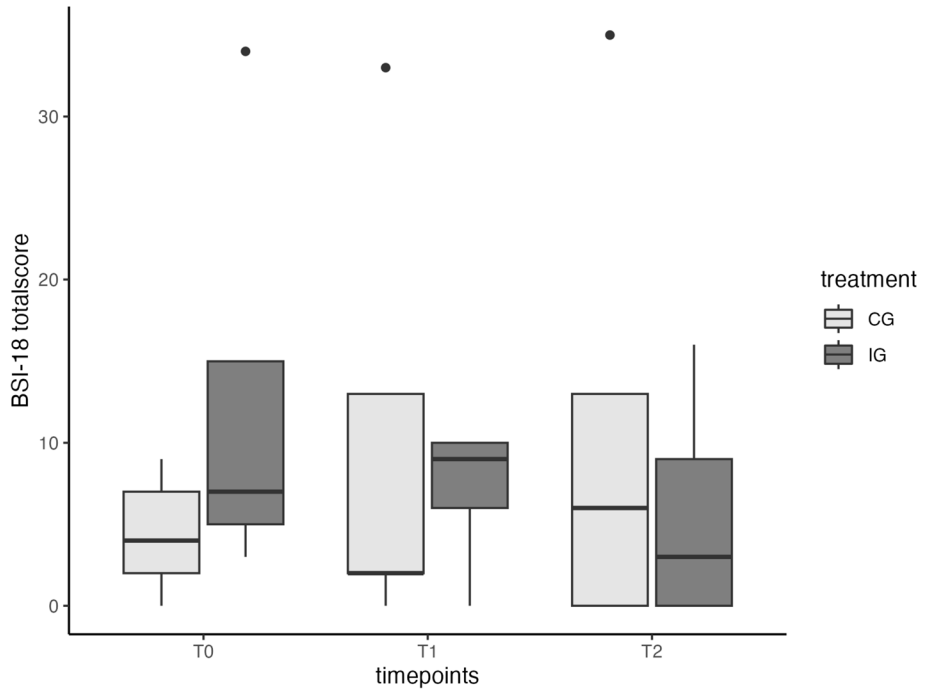

Fig. 4 Results of the Brief Symptom Inventory for the intervention (IG) and control (CG) groups

Family Empowerment Scale (FES)—family subscale

FES scores of parents of the IG remained similar from T0 to T2, whereas scores of parents of the CG decreased to T2. This indicates that parents of the IG felt empowered during the timepoints, whereas parents the CG felt less empowered over time, with an increase in the gap between both groups. Reliable change was not reported as there were no norm data available.

Parental Stress Index (PSI)

PSI median scores of parents of the IG decreased from T0 to T2, whereas median scores of parents of the CG increased, indicating that some parents of the CG might have experienced increased stress over time. However, there was no reliable change in mean scores in either of the two groups from T0 to T2. Whereas reliable improvement was detected in two families of the IG, reliable deterioration was detected in one family of the IG and two families of the CG.

Median scores of both groups were above 60 at T0 and T2. This indicated that families in our study perceived more stress than German norms [59].

Parental Overprotection Measure (POM)

In parents of the IG, POM scores increased from T0 to T2, whereas parents of the CG showed a decrease. Reliable change was not reported as there are no norm data available.

In summary, the results of motor outcomes did not show differences between groups; however, we detected improvements of reliable change scores in some family well-being outcomes for families of the intervention group compared to families receiving standard of care.

Feasibility of secondary outcome measures

Regarding motor outcomes measures, the GMA, IMP, and AIMS were easy to implement. The HINE, however, was difficult to perform as some infants were very irritable during the examination. The motor domain of the BSID III did not prove to be a good assessment tool for measuring small or qualitative changes associated with the intervention. Regarding family well-being outcomes measures, especially in the SF-36-MF, the BSI and PSI reliable change in scores of individual families could be detected.

Discussion

The major aim of our study was to assess the feasibility and acceptability of the study design, study procedure, and EMI-Heart, a family-tailored early intervention for infants with CHD after open-heart surgery. The results of this feasibility and RCT pilot of EMI-Heart including 10 infants showed that (a) the intervention was feasible and acceptable to both parents and the PT, (b) online treatment sessions were easier to attend for both parents and a practical alternative to home and hospital visits, and (c)

some of the secondary outcome measures require modification when used in a full trial. Specifically, the Hammersmith Infant Neurological Examination may not be used shortly after surgery since infants did not tolerate the neurological examination. Further, the third edition of the Bayley Scales of Infant Development may not be a good assessment tool for measuring small or qualitative changes associated with an intervention, as it measures milestones. In addition, the Parental Overprotection Measure may not be a valid tool to use during infancy, where a certain degree of 'overprotective parenting' is considered as good parenting.

Although, our sample size was small, the recruitment rate of 60% was satisfactory, and no participants dropped out of either the intervention or control groups throughout the study period. The recruitment process was hampered by the COVID-19 pandemic during which surgeries had to be cancelled and postponed. Thus, infants who were initially eligible but who then became too old for inclusion were lost from the study as we only included infants until the age of 5 months. Because we aimed for a homogeneous sample, we had to exclude a large number of infants with CHD, including those with a univentricular CHD, those also diagnosed with a genetic comorbidity, and those born premature. These infants should be included in a future full trial as they are at highest risk of neurodevelopmental impairments [12, 75] and therefore should be eligible for early motor interventions.

The adherence to the protocol in regard to study design and procedure was successfully maintained by the researcher (EM) and was delivered to all participating families as planned. The intervention dose of nine sessions per infant, a combination of home, hospital, and online settings, and the 4-month duration of the intervention was provided as intended.

Acceptability to parents was very high. This was underlined by results of the acceptability questionnaire and parental attendance in all intervention sessions. Parents attended all sessions regardless of the setting which indicates excellent feasibility. One interesting finding was that the presence of mothers, fathers, or both depended on the setting: whereas the hospital sessions were attended almost exclusively by mothers, online sessions were more frequently attended by fathers or both parents. This experience is in line with recent literature discussing the benefit of online health services, which include reductions in travel and waiting times, parking costs, and pressure to take time off work and manage childcare. Online sessions have been reported to refine communication between therapists and parents and leave time and space for parents to find their own solutions [76–78]. We therefore aim to continue using online sessions in our practice as a valuable alternative to face-to-face interventions.

An additional key confirmation of the intervention's acceptability to parents was the exchange of video recordings provided by parents and those of the recorded intervention sessions. We introduced this approach in EMI-Heart because research has indicated that video feedback promotes parental engagement and improves their ability to read and respond to their children's cues [79, 80]. An unexpected finding was the large number of video recordings each family sent to the PT: on average 16 recordings per infant throughout the intervention period. This demonstrates parents' active engagement, a key element in EMI-Heart. These video recordings enabled the PT to observe a range of parent–child interactions in daily life, such as early supported sitting while bathing, in a high-chair at the piano, at the dining table, and in the kitchen. The PT observed that exchanging videos added value to parents' reports in providing insight into how parents incorporated EMI-Heart in daily life. It also allowed the PT to provide direct feedback and promote parental confidence in themselves and their infant.

Furthermore, we plan to assess the fidelity of EMI-Heart to determine whether the content of the intervention was provided as intended. All video recordings of intervention sessions and those provided by the parents were analysed with a content-structured content analysis by a master student using MQXDA 2020. The fidelity of EMI-Heart will be analysed at a later timepoint.

Our secondary outcomes provided several aspects to be discussed. We included many outcome measures to determine which might best determine the effect of the intervention on infant motor and parental well-being and would be sensitive enough for use in a full trial. We decided to keep the Infant Motor Profile and the Alberta Infant Motor Scale as motor outcomes. They complement each other, can be assessed together, and thus do not impose any additional burden on infants. We encountered difficulties performing the Hammersmith Infant Neurological Examination. Infants were often very irritable during the neurological examination. This irritability may be due to sensitivity in the chest region following surgery and aversion to manipulation following treatments associated with the cardiac disease. We used the Bayley Scales of Infant and Toddler Development III at 1 year of age because this test is widely used to assess at-risk children's performance in three main developmental domains: motor, language, and cognition. However, the Bayley motor domain may not be a good assessment tool for measuring small or qualitative changed associated with an intervention is not a sensitive tool for measuring intervention, as it measures milestones. The fourth edition may be more sensitive to change as it provides a polytomous scoring of motor performance. Because the patients included in this feasibility RCT study were in

the less severe part of the CHD spectrum and thus had better motor outcomes than infants with more severe CHD, it was unlikely that the intervention would lead to improved motor outcomes. As mentioned above, the group of infants with univentricular CHD should be included in a future full trial for this comprehensive, family-tailored intervention.

Although using the General Movement Assessment (GMA) restricted our sample regarding the age range of eligible patients as a baseline assessment, we would emphasize the usefulness of the motor optimality score (MOS-R). In a full trial, the GMA could be used as a predictor variable but should not be used in such a way that all infants would require a GMA assessment, thus being an inclusion criteria. In our sample, all infants had MOS-R scores ≤ 21, and research has shown that normal fidgety movements in combination with a MOS-R score of ≤ 21 can indicate an increased risk of difficulties in motor performance, working memory, and executive function at school age [69, 81]. Lower MOS-R scores may indicate poorer quality of motor behaviour at T0, which might be associated with lower scores in the performance, fluency, and variation domains of the IMP at T2.

Family well-being outcomes were assessed with the Pediatric Quality of Life Inventory Infant Scales (PedsQL), Parental Stress Index (PSI), Family Empowerment Scale (FES), Quality of Life Short Form Survey (SF-36), and Brief Symptom Inventory (BSI-18) questionnaires. The PSI, the SF-36, and the BSI-18 seemed to be sensitive for measuring intervention changes as we detected reliable change, implying that changes between baseline and follow-up were unlikely to be due to measurement unreliability. We used both the SF-36 and the BSI-18. Compared to the SF-36, the BSI-18 did not only detect reliable change in individual families but also between mean scores of both groups. We concluded that the BSI-18 would be sufficient to assess mental health for a future full trial as its content overlaps with the SF-36. Even though there were no norm data available for the FES to calculate reliable change scores, its descriptive data suggest differences between the intervention and the control group.

From the results of our qualitative study and of other studies [82, 83], we decided to use the Parental Overprotection Measure (POM), because this is the only questionnaire available that assesses parental overprotection. It was developed for preschool children, 3–5 years of age, and assesses parental overprotection in situations that involve perceived risk or threat to the child [84]. It became clear that the questions on the POM focusing on parental overprotective behaviour in preschool children (e.g. 'I keep a close watch on my child at all times') were not valid for use in the infant age group, as this is considered good parenting not overprotective parenting.

Several limitations need to be considered regarding the design of this study. Our inclusion criteria were strict and hampered recruitment, which led to a small sample. This prevented the application of statistical analyses to examine individual factors within the sample. By using an RCT design, we had hoped to achieve similar baseline characteristics between groups. However, this was not the case, most likely due to the small sample size. Further, four out of five infants in the control group received outpatient PT. This might have been because the socioeconomic status (SES) of parents of the control group was higher than that of parents of the intervention group. The association of parental SES with early child development is well-known [34, 85, 86]. Alternative controlled study designs such as pragmatic randomized clinical trials and randomized crossover designs might offer practicable alternatives to classical RCTs. They have less restrictive inclusion criteria and still allow randomization at individual and cluster levels (e.g. within hospitals, regions). Pragmatic randomized clinical trials are used to understand how treatments work in real-world scenarios with heterogenous populations to optimize the generalizability of trial results [87].

Furthermore, the travel distance to and from the hospital was a limiting factor for enrollment. In relation to the small size of Switzerland, a 1-h journey constitutes a significant distance. Even when insurance companies agree to pay for home visits, only a fixed fee is paid independent of travel time. Additionally, a reduced fee is paid for online sessions. This issue requires discussion between physiotherapy associations and health insurance companies. In our study, both parents and the PT appreciated that the intervention took place in three different settings.

The systematic review by Kaeslin et al. [15] described the influence of early physiotherapy in infants with CHD after open-heart surgery within the first year of life. The focus of early physiotherapy described was mainly on strengthening and performance of functional activities. Even though no positive effects could be shown, a trend towards improvement in motor development could be observed. However, family-focused programmes like the Congenital Heart Disease Intervention programme (CHIP) [14] focusing on maternal adjustment demonstrated significant gains on mental scale of the Bayley and improvements of maternal anxiety and coping. Other relation-based interventions focusing on infant feeding [88] or skin-to-skin contact [89] showed improved infant weight gain, better automatic regulation, and shorter hospital stays compared to interventions that focused on functional improvement like oral motor exercises [90]. These results confirm the findings of secondary outcomes in our study with a tendency for families of the intervention group to perceive better family well-being than those of the control group.

While the general aim of early intervention programmes for infants at increased risk of motor impairments is to advance the function of the motor system [91], EMI-Heart focuses on empowering parents to support their infants' development and to trust in themselves and their infant. EMI-Heart promotes parental sensitivity and positive responsiveness to infant's actions and interactions such as parent-infant attunement, turn-taking behaviour, experience of joyful play, and infant empathy during daily care activities. The physiotherapist uses her therapeutic ideas and experience as a means of promoting parent–infant attachment and thus nurturing the distressed infant and parents. This is of particular importance as family factors are the most relevant predictors of later neurodevelopmental and psychosocial outcomes of infants with CHD [36].

Conclusion

Our research demonstrates the feasibility of our study design and indicates that EMI-Heart is a feasible intervention for infants with CHD after open-heart surgery that was highly acceptable to both parents and the paediatric physiotherapist. Online treatment sessions offer a valuable alternative to home and hospital visits. Parental video recordings can provide paediatric physiotherapists with additional information about how parents practise an intervention in daily life. Further research in a full trial using a pragmatic RCT may provide evidence to support the widespread use of EMI-Heart, a family-tailored early motor intervention for infants with CHD and their families.

Abbreviations

CHD	Congenital heart disease
EM	Elena Mitteregger
TD	Tineke Dirks
MT	Manuela Theiler
BL	Beatrice Latal
OK	Oliver Kretschmar
EMI-Heart	Family-tailored early motor intervention in infants with complex congenital heart disease
IG	Intervention group
CG	Control group
GMA MOS-R	General Movements Assessment including Motor Optimality Score
HINE	Hammersmith Infant Neurological Examination
IMP	Infant Motor Profile
AIMS	Alberta Infant Motor Scale
BSID III	Bayley Scales of Infant and Toddler Development
PedsQL	Pediatric Quality of Life Inventory Infant Scales
SF-36	Quality of Life Short Survey 36
BSI-18	Brief Symptom Inventory 18
FES	Family Empowerment Scale
PSI	Parental Stress Index
POM	Parental Overprotection Measure

Supplementary Information

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Additional file 1: Supplementary Material 1 Acceptability questionnaire.

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Authors' contributions

EM, TD, and BL conceptualized this study. EM and TD wrote the manuscript, and BL critically revised it. MT gave her advice and feedback as a parental stakeholder. OK contributed his medical expertise and advised on recruitment. All authors discussed, read, and approved the final manuscript for publication.

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Availability of data and materials

The datasets used and analysed are available from the corresponding author on reasonable request.

Declarations

Ethics approval and consent to participate

This study was registered at ClinicalTrials.gov, NCTT04666857 on 23.11.2020. Written informed consent was obtained from all parents by EM.

Consent for publication

Participants gave consent for the publication of the results of this feasibility RCT. Findings from this study will be presented at national and international conferences, to parent organizations, and to healthcare stakeholders for widespread dissemination of the results.

Competing interests

The authors declare no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

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