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## Supporting Parenting at Home – Empowering Rehabilitation through Engagement (SPHERE): Study protocol for a randomized control trial

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# Supporting Parenting at Home – Empowering Rehabilitation through Engagement

## (SPHERE): Study protocol for a randomized control trial

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## ABSTRACT

**Introduction.** Infants with neurodevelopmental disabilities (NDs) show emotional, cognitive, and socio-interactive dysregulation dramatically impacting on caregiving behavior. Early video-feedback interventions (VFIs) are effective in promoting sensitive parenting, that in turn supports infants' development, even in case of ND. In the light of limited resources of the healthcare systems, technological advances in telemedicine may facilitate the delivery of VFI to a greater number of families of infants with ND. To date, no study has implemented a telemedicine VFI (TVFI) for families of infants diagnosed with ND. **Methods and analysis.** The Supporting Parenting at Home – Empowering Rehabilitation through Engagement (SPHERE) project is a randomized controlled trial aimed at assessing the effectiveness of an early family-centered TVFI parenting support on dyads with infants diagnosed with ND. It includes two arms (TVFI vs. Booklet Psychoeducational Intervention) and three assessment phases: T0, baseline; T1, immediate post-intervention; T2, 6-month follow-up. **Ethics and dissemination.** This study is funded by the Italian Ministry of Health and was approved by the Ethics Committee (Pavia). Results will be published in peer-reviewed journals and presented at national and international scientific conferences.

**Keywords:** family-centered intervention; maternal sensitivity; neurodevelopmental disability; parenting; telemedicine; video-feedback.

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2 27 **Strengths and limitations of this study**  
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- 5 28 • This RCT will develop, deliver, and test a scalable video-feedback intervention to  
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7 29 improve parental skills and developmental outcomes of infants with neurodevelopmental  
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9 disabilities.  
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12 31 • The telemedicine approach of this intervention will allow to reduce the inequality of  
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14 32 access to family-centered care in neurodevelopmental disability.  
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16  
17 33 • Neurodevelopmental disabilities include a wide range of clinical conditions and this will  
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19 34 be reflected in a heterogenous sample. Nonetheless, this intervention targets parenting  
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21 35 challenges that are generally common and shared among parents of infants with  
22  
23 36 diverse disability phenotypes.  
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25  
26 37 • The telemedicine nature of the intervention is not free from potential technical issues  
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28 38 related to poor quality of internet signal; nonetheless, no specific resources are needed  
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31 39 on family side which will be able to connect with any device.  
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## INTRODUCTION

Every year, about 53 million infants worldwide receive a diagnosis of neurodevelopmental disability (ND) and they represent the 13% of all health problems in infancy and childhood [1]. Although ND includes a wide range of diverse clinical conditions (e.g., cerebral palsy, genetic or malformative syndromes, outcomes of severe prematurity, autism spectrum disorders), these infants may share common difficulties in emotional regulation, cognitive skills, and socio- interactive abilities [2,3]. Such pattern of multi-systemic dysregulation negatively impacts on parents' psychological and physical health as well as on caregiving behavior [4,5]. Parents may report critical emotional burden with heightened risk for chronic levels of distress, depression and anxiety [4–6]. This constitutes a crucial point considering that parenting represents the frontline of developmental resilience and prevention for infants' development, even in the presence of ND conditions. Indeed, infants with ND whose parents are rated high in parental sensitivity show better outcomes in emotional, cognitive and socio-interactive developmental trajectories [7–9].

From this perspective, it is not surprising that early rehabilitation interventions have been found to be the most effective when they engage parents in a family-centered approach [10] and they also are the most rewarding care strategies for healthcare systems in terms of economic return in the long-term [11]. Specifically, the Video Feedback Intervention (VFI) [12] constitutes an early family-centered care strategy that proved to be effective in promoting sensitive parenting and infants' behavioral and socio-emotional adjustment [13]. The use of VFI intervention has been also documented to be beneficial in dyads of children with ND [14–16]. Previous research reported that these interventions may result in reduced child's disruptive and emotionally negative behaviors [17], improved maternal sensitivity [18,19], increased parental self-efficacy and less parenting stress [20]. As such, early

1  
2 64 supportive interventions directed at improving the quality of parental sensitivity and parent-  
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4 65 infant interaction should be prioritized even in a population of children diagnosed with ND  
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6 66 [10,21].  
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9 67 It should be highlighted that delivering VFI in hospital- or home-based context should be  
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11 68 highly demanding for the healthcare systems due to high cost and disparities in access to  
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13 69 services for families who live in remote areas. As such, delivering VFI through telemedicine  
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15 70 (TVFI) approaches using videoconferencing is a valuable option that may reduce care  
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17 71 access inequalities, promote early family-centered care culture, and contribute to a more  
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19 72 effective and efficient healthcare approach to the rehabilitation program of infants with ND.  
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21 73 Notably, videoconference approaches to parent training interventions (e.g., cognitive  
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23 74 educational training) showed to be as effective as traditional face-to-face approaches and  
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25 75 were accepted with the same degree of satisfaction by parents [22,23]. Nonetheless, we still  
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27 76 do not know how a TVFI support may end up in being effective in terms of promoting  
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29 77 parental health and infants' development in families of little patients with ND.  
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34 78 **METHODS AND ANALYSIS**

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36 79 **Aims**

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39 80 The SPHERE project is a randomized control trial that aims to develop, deliver, and test the  
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41 81 effectiveness of an early TVFI compared to an alternative educational intervention. The first  
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43 82 specific aim is to assess the TVFI effectiveness in supporting maternal sensitive parenting  
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45 83 in dyads of infants with ND. Previous studies suggested that a traditional VFI increased  
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47 84 parental capacity to respond to child's communicative signals in parents of children with  
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49 85 disabilities [18,19]. Consistently, we hypothesize that the TVFI will be effective in increasing  
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51 86 maternal sensitivity (within-group difference) and will be more effective than the alternative  
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53 87 not individualized intervention (between-group difference).  
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The second specific aim is to test the TVFI effectiveness in reducing infants' emotional stress reactivity. In line with a previous study highlighting a significant reduction of emotionally negative behaviors after VFI [17], we hypothesize that the TVFI will be effective in reducing infants' emotional stress in the experimental group. Additional exploratory aims include the assessment of the SPHERE TVFI intervention on other maternal (i.e., parenting stress, depressive symptoms, anxiety symptoms) outcomes and on infants' behavioral regulation (i.e., temperament profile).

### Study design

The SPHERE project is a randomized control trial with two arms and three assessment phases: T0, baseline; T1, immediate post-intervention; T2, follow-up (6 months after the intervention). The experimental arm consists in the telemedicine intervention (TVFI arm), whereas the control arm consists in the delivery of a psychoeducational booklet (PEB arm). For both arms, each assessment session will include (a) an on-line questionnaire on maternal and infants' well-being (see below) and a 15-minute mother-child dyadic interaction will be videotaped in remote. This interaction will include approximately 10-minute face-to-face play interaction followed by a Face-to-Face Still-Face (FFSF) procedure [24]. The FFSF is a well-validated observational procedure to assess infants' socio-emotional regulation and parenting sensitivity that has been previously used with infants with ND [25]. During the FFSF, the mother is asked to interact with her infant for two minutes (Play episode), then to interrupt any communication and to maintain a still poker face for two minutes (Still-Face episode), and finally to resume the interaction for two final minutes. An overview of the study protocol and procedures is reported in figure 1.

---- Please, insert Figure 1 here ----

### Population

1  
2 112 The mother-infant dyads will be enrolled at the Child Neurology and Psychiatry Unit of the  
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4 113 IRCCS Mondino Foundation, Pavia (Italy). They will be enrolled consecutively according to  
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6 114 the following inclusion criteria: infants' (corrected) age between 1 and 18 months; presence  
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8 115 of developmental risk for or diagnosis of ND as defined by standardized clinical criteria;  
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10 116 parental age greater than 18 years; parental mastery of Italian language; both parents living  
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12 117 together with the infant. Exclusion criteria include presence of twins, infant's life-threatening  
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14 118 conditions, and maternal full-blown psychiatric disorders. The dyad's randomized allocation  
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16 119 to each arm will be done through a computerized 0/1 sequence generator.  
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20 120 **Intervention, TVFI arm**

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22 121 The TVFI will be standardized according to previously published RCTs [26,27]. The TVFI is  
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24 122 inspired to concepts from the Collaborative Consultation approach from Zack Boukydis [28].  
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26 123 Specifically, six weekly TVFI sessions will be delivered in two subsequent phases, as in  
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28 124 previous VFI research with families of children with ND [29]. Four *sharing the focus* 1-hour  
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30 125 sessions will be dedicated to the discussion between the psychologist and the mother of  
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32 126 specific themes related to parenting and parent-child interaction: physical stimulation,  
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34 127 responsiveness, teaching, and parenting experience (see Table 1). During these sessions,  
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36 128 the psychologist will conduct a dialogic interactive session connecting with the mother in  
37  
38 129 videoconference. The videotaped interaction obtained at T0 will be discussed by the  
39  
40 130 principal investigator (SG) and a senior author (LP) who is experienced in mother-infant  
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42 131 interaction coding and early VFI interventions and it will be segmented into specific videoclip  
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44 132 lasting up to 10 seconds. Each videoclip will be labeled with one of the thematic contents  
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46 133 reported in Table 1. During the *sharing the focus* sessions, the psychologist will propose to  
47  
48 134 the mother to jointly review some of these segments, usually starting from potential  
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50 135 curiosity, comments or requests from the mother herself. The specific order in which the  
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52 136 themes will be discussed will be tailored on each specific case. The number of videoclips  
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used for each thematic label in the TVFI of each family will be noted by the psychologist on a diary. The goal of the *sharing the focus* sessions is to develop insights about the infants' behavioral signals, the best ways to provide stimulations and get in contact, strategies to promote adaptive emotion regulation and to sustain cognitive and behavioral achievements. In the subsequent two 1-hour *integration* sessions, the mother will play with the infant while the psychologist will provide a dyadic-tailored guidance based on topics previously discussed during the first four sessions. The goal of the *integration* sessions is to promote a pragmatical translation of the insights developed during the *sharing the focus* sessions into the interactive exchanges between the mother and the infant. By doing so, the mother can introduce variations in her caregiving behavior in a safe environment under the supervision of a trained specialist.

---- Please, insert Table 1 here ----

### **Intervention, PEB arm**

Mothers assigned to condition B will receive an informative booklet addressing the same themes discussed in the experimental intervention (i.e., responsiveness, physical stimulation, teaching, and parenting experience), but not tailored on their own infant or specific parenting challenges.

### **Core variables and measures for the specific aims**

Maternal sensitivity will be assessed using the Global Rating Scales (GRS) [30] from the first 5-minute segment of videotaped mother-infant interactions at T0, T1, and T2. The GRS provides indexes of maternal sensitivity and intrusiveness that ranges from 1 (low scores) to 5 (high scores) and require a holistic, macro-analytic coding of the interaction. Infants' emotional stress reactivity will be coded in terms of negative emotionality display (by voice and/or facial expression) across the FFSF procedure videotaped at T0, T1, and T2. Negative emotionality display will be coded according to previous system validated in FFSF

1  
2 162 research [31]. For both coding, students will be trained using available reference videoclips  
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4 163 according to a gold standard 85% inter-rater agreement. Trained coders will be blind to the  
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6 164 arm allocation and to the specific goals of the intervention.  
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9 165 **Other measures (additional exploratory aims)**

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11 166 Information on the clinical characteristics and the diagnosis of the infant will be obtained  
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13 167 from medical records. By filling in on-line questionnaires at T0, mothers will provide  
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15 168 information on infants' neonatal characteristics (i.e., sex, gestational age in weeks, birth  
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17 169 weight in grams), socio-demographic variables (i.e., maternal age, educational level, and  
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19 170 occupational status). In the same questionnaire as well as in the following questionnaires  
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21 171 administered at T1 and T2, mothers will fill in the 36-item Parenting Stress Index – Short  
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23 172 Form (PSI-SF) [32], the 21-item Beck Depression Inventory (BDI-II) [33], the 20-item state  
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25 173 subscale of the State-Trait Anxiety Inventory (STAI-Y) [34], and the 91-item Infant Behavior  
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27 174 Questionnaire-Revised (IBQ-R) short form [35]. All these questionnaires have been  
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29 175 validated in Italian and they are largely used in parent-infant research in experimental and  
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31 176 clinical settings.  
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36 177 **Statistical power and sample size estimation**

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38 178 The sample size was estimated for what pertains the first specific aim, i.e., the effect of an  
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40 179 early VFI intervention on maternal sensitivity. A minimum sample size of 59 subjects per  
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42 180 group [ $\alpha = .05$ ,  $\beta = .05$ ,  $\text{power} = .95$ , effect size,  $d = .67$ ] was estimated using  
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44 181 G\*Power software on the basis of meta-analytical evidence of online interventions focused  
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46 182 on parenting [36]. Moreover, due to the longitudinal nature of the study, attrition rate of 20%  
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48 183 for each phase was considered, therefore a starting sample size of 84 infants for each  
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50 184 group (total sample size = 168) has been estimated.  
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55 185 **Plan of statistical analyses**

R Studio packages [37] and IBM SPSS 27 [38] will be used for the statistical analyses. Participants with more than 20% missing data in the questionnaires as well as those who did not complete at least T0 and T1 assessments will be excluded from the analyses. TVFI and PEB dyads will be preliminarily compared for socio-demographic and neonatal characteristics. In order to respond to the specific aims of the SPHERE study, standard mixed analyses of variance will be used to test within- and between-group differences in maternal sensitivity and infant's emotional stress reactivity. The difference between T0 and T1 measures will be considered as the primary output of these analyses and long-term maintenance of the effect will be tested by including a third time point (T2) in the model. Advanced modelling will be used to assess the effects of potential mediators/moderators (e.g., infants' sex and age) on maternal and infants' outcome variables. Standard mixed analyses of variance will be used to test within- and between-group differences in maternal parenting stress, depression, anxiety, and infant's behavioral regulation across the three assessments time points.

### **Patient and public involvement**

The intervention was not developed in concert with families or parents. Nonetheless, webinars and educational events dedicated to parents and rehabilitation care professionals will be organized to disseminate the findings of the SPHERE project and to promote a culture of family-centered care in the context of ND healthcare. Moreover, after the end of the RCT, the booklet of the PEB will be distributed within the clinical unit of the IRCCS Mondino Foundation and will become a freely available resource for parents and staff. The booklet will also circulate to families thanks to the engagement of parents' associations that collaborate with authors for educational and clinical purposes.

## **DISCUSSION**

### **Limitations**

1  
2 211 First, the telemedicine nature of this study may imply technical problems due to internet  
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4 212 instability. Therefore, the quality of internet signal may be in some cases low or moderate  
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6 213 and the quality of video output may be partially suitable for micro-analytic coding. However,  
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8 214 the GRS coding system used for the assessment of the primary outcome of the intervention  
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10 215 relies majorly on global scorings that can be performed with medium quality video output.  
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12 216 Moreover, attrition rate has been included in the sample size estimation also to take care of  
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14 217 potential loss of dyads due to technical problems. Second, as dealing with the ND clinical  
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16 218 condition of their son or daughter may be highly stressful for mothers, it should not be  
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18 219 excluded that mothers may express the need of more specific emotional and psychological  
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20 220 needs during the SPHERE project. In these cases, proper referral to specialists will be  
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22 221 discussed with the mother. Third, ND include a wide ensemble of clinical conditions and the  
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24 222 different levels of psychomotor delay as well as the different domains that may be impaired  
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26 223 certainly play a role in affecting maternal sensitivity and well-being and also impact on the  
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28 224 quality of parent-infant interaction. For example, the presence of a mild psychomotor delay  
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30 225 or a severe sensory impairment may pose different challenges in the daily interactive  
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32 226 exchanges. Nonetheless, the heterogeneity of the clinical conditions is a inherent  
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34 227 characteristics of ND and it is often a limit to studies that are mainly focused on promoting  
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36 228 directly specific infants' developmental outcomes. In the present study, the main focus is on  
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38 229 supporting maternal sensitivity as a proxy to further promote infants' development and well-  
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40 230 being. Moreover, due to the early age at which infants are enrolled in the study, it is highly  
41  
42 231 probably that they will partially share a risk condition for ND and they will not have already  
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44 232 received a specific diagnosis. Notwithstanding, when available, a quantitative psychomotor  
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46 233 developmental quotient with standardized scales will be obtained from medical records and  
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48 234 will concur to define the enrolled sample.

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57 235 **Expected results and implications**  
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The SPHERE project holds promises to test a new model for the telemedicine care and rehabilitation of infants with neurodevelopmental disabilities. By investing in an early and family-centered intervention we aim to provide families with timely and efficient support to the rehabilitation journey, contributing to grow a sense of self-efficacy in the caregivers of infant with ND. Moreover, by promoting a telemedicine VFI we can contribute to reduce the healthcare inequalities for families who face challenges in accessing to traditional rehabilitation programs because they live in rural areas or because of long waiting lists. The SPHERE project may provide evidence-base support to the development and application of TVFI approaches that may maximize the benefit of early family-centered interventions for both parents' well-being and infant's development.

### **Ethics and dissemination**

The study has received the approval of the Ethics Committee Pavia on November 2<sup>th</sup>, 2020, Protocol number 20200096046. All the procedures are consistent with the Declaration of Helsinki ethical principles for research involving human subjects. The procedures do not imply any harm to the participating subjects. Moreover, the study interventions represent additional opportunities for families that do not imply changes in usual mother-infant care programs. All infants will take part to all the diagnostic and therapeutic interventions that are planned in the child neurology and psychiatric unit IRCCS Mondino Foundation, Pavia, Italy. The study protocol has been also registered on NIH Clinical Trials (protocol code NCT04656483). The dissemination plan includes the presentation of findings at national and international scientific meetings as well as the publication in scientific journals in the field of developmental psychology. The findings will also be disseminated to the public through reach-out activities involving families and healthcare specialists, in order to promote early family-centered intervention.

1  
2 260 **Authors' contributions:** All authors conceived the study protocol and the rationale for the  
3  
4 261 project. SG and LP wrote the funding proposal for this study. SG and LP coordinates  
5  
6 262 recruitment of participants and data collection. LP and RB supervise the progress of the  
7  
8 263 study. SG and LP wrote the first version of this manuscript draft. RB provided suggestions  
9  
10 264 for the improvement of the manuscript. All authors read and approved the final manuscript.  
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15  
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26  
27 270 participants' enrolment.  
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## ABBREVIATIONS

- VFI, Video Feedback Intervention
- ND, Neurodevelopmental Disabilities
- TVFI, Telemedicine Video Feedback Intervention
- PEB, psychoeducational booklet
- GRS, Global Rating Scales
- PSI-SF, Parenting Stress Index, Short Form
- BDI-II, Beck Depression Inventory
- STAI-Y, State-Trait Anxiety Inventory
- IBQ-R, Infant Behavior Questionnaire – Revised

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**Table 1.** Description of the TVFI *sharing the focus* sessions' themes

Theme	Sub-themes	Description
1 Stimulation	Type of stimulations	Type of stimulations (e.g., auditory, tactile) preferred by the infant
	Intensity of stimulations	Infant's sensitivity to the intensity of stimulations
	Social touch	Maternal touch in promoting infant's body awareness and attentional orientation
2 Responsiveness	Sensory integration	Infant's sensory integration and body awareness
	Sense of agency	Supporting the infant's initiative to promote the development of his sense of agency
	Sensitivity	Perceiving and interpreting child's signals and responding in a prompt and appropriate way
	Exploration	Supporting the child's exploration and his use of the parent as a secure base
3 Teaching	Reparation	Repairing communicative ruptures
	Attention skills	Supporting the infant's attention orientation
	Modelling	Providing a model to the infant in order to foster the observational learning
	Scaffolding	Parental guidance to allow the infant to solve a task that he cannot yet carry out on his own
4 Parenting experience	Proximal development zone	Encouraging learning in the infant's proximal development zone
	Representations of the baby	Maternal representations of the infant and curiosity about his mind
	Self-regulation	Taking care of herself
	Self-efficacy	Mother's sense of efficacy and trust in her own experience

## FIGURE LEGENDS

**Figure 1.** Schematic overview of the study design.

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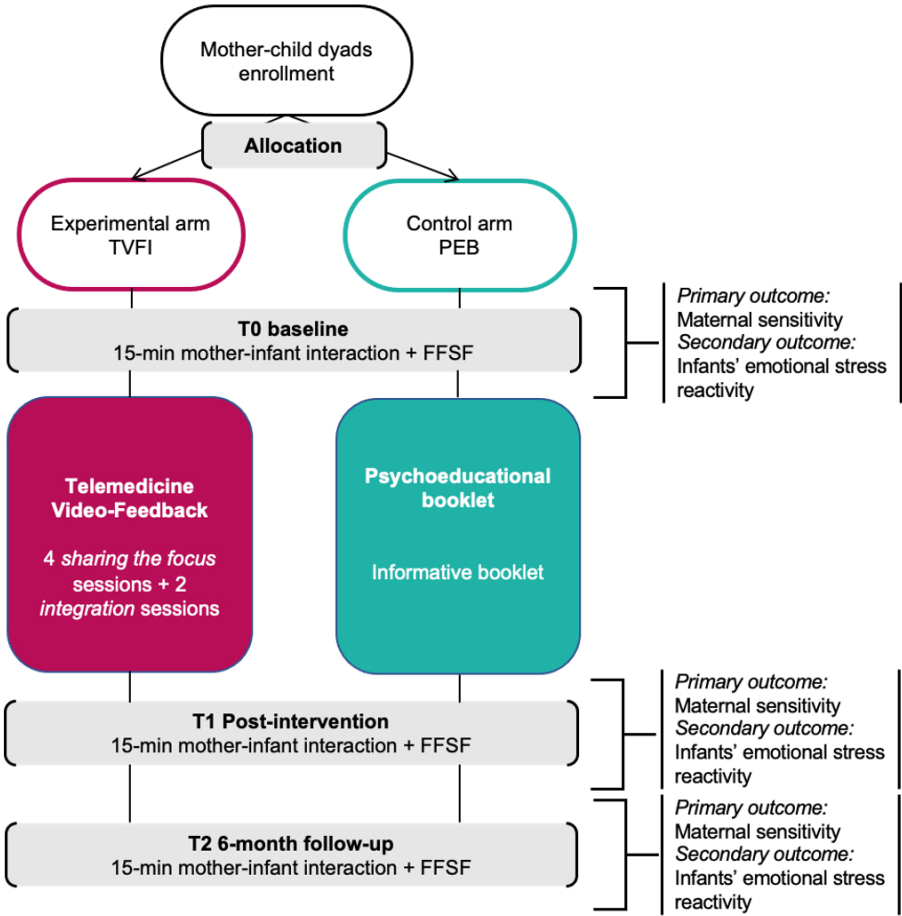


Figure 1. Schematic overview of the study design.

190x199mm (150 x 150 DPI)



# CONSORT 2010 checklist of information to include when reporting a randomised trial\*

Section/Topic	Item No	Checklist item	Reported on page No
<b>Title and abstract</b>			
	1a	Identification as a randomised trial in the title	1
	1b	Structured summary of trial design, methods, results, and conclusions (for specific guidance see CONSORT for abstracts)	1 (results are NA)
<b>Introduction</b>			
Background and objectives	2a	Scientific background and explanation of rationale	4-5
	2b	Specific objectives or hypotheses	5-6
<b>Methods</b>			
Trial design	3a	Description of trial design (such as parallel, factorial) including allocation ratio	6
	3b	Important changes to methods after trial commencement (such as eligibility criteria), with reasons	6-7
Participants	4a	Eligibility criteria for participants	7
	4b	Settings and locations where the data were collected	7
Interventions	5	The interventions for each group with sufficient details to allow replication, including how and when they were actually administered	7-8
Outcomes	6a	Completely defined pre-specified primary and secondary outcome measures, including how and when they were assessed	8-9
	6b	Any changes to trial outcomes after the trial commenced, with reasons	NA
Sample size	7a	How sample size was determined	9-10
	7b	When applicable, explanation of any interim analyses and stopping guidelines	NA
<b>Randomisation:</b>			
Sequence generation	8a	Method used to generate the random allocation sequence	7
	8b	Type of randomisation; details of any restriction (such as blocking and block size)	7
Allocation concealment mechanism	9	Mechanism used to implement the random allocation sequence (such as sequentially numbered containers), describing any steps taken to conceal the sequence until interventions were assigned	7
Implementation	10	Who generated the random allocation sequence, who enrolled participants, and who assigned participants to interventions	7

1	Blinding	11a	If done, who was blinded after assignment to interventions (for example, participants, care providers, those assessing outcomes) and how	9
2		11b	If relevant, description of the similarity of interventions	7-8
3	Statistical methods	12a	Statistical methods used to compare groups for primary and secondary outcomes	9-10
4		12b	Methods for additional analyses, such as subgroup analyses and adjusted analyses	9-10
5				
6	<b>Results</b>			
7	Participant flow (a diagram is strongly recommended)	13a	For each group, the numbers of participants who were randomly assigned, received intended treatment, and were analysed for the primary outcome	NA
8		13b	For each group, losses and exclusions after randomisation, together with reasons	NA
9	Recruitment	14a	Dates defining the periods of recruitment and follow-up	NA
10		14b	Why the trial ended or was stopped	NA
11	Baseline data	15	A table showing baseline demographic and clinical characteristics for each group	NA
12	Numbers analysed	16	For each group, number of participants (denominator) included in each analysis and whether the analysis was by original assigned groups	NA
13	Outcomes and estimation	17a	For each primary and secondary outcome, results for each group, and the estimated effect size and its precision (such as 95% confidence interval)	NA
14		17b	For binary outcomes, presentation of both absolute and relative effect sizes is recommended	NA
15	Ancillary analyses	18	Results of any other analyses performed, including subgroup analyses and adjusted analyses, distinguishing pre-specified from exploratory	NA
16	Harms	19	All important harms or unintended effects in each group (for specific guidance see CONSORT for harms)	NA
17	<b>Discussion</b>			
18	Limitations	20	Trial limitations, addressing sources of potential bias, imprecision, and, if relevant, multiplicity of analyses	10
19	Generalisability	21	Generalisability (external validity, applicability) of the trial findings	NA
20	Interpretation	22	Interpretation consistent with results, balancing benefits and harms, and considering other relevant evidence	NA
21	<b>Other information</b>			
22	Registration	23	Registration number and name of trial registry	12
23	Protocol	24	Where the full trial protocol can be accessed, if available	NA
24	Funding	25	Sources of funding and other support (such as supply of drugs), role of funders	13

\*We strongly recommend reading this statement in conjunction with the CONSORT 2010 Explanation and Elaboration for important clarifications on all the items. If relevant, we also recommend reading CONSORT extensions for cluster randomised trials, non-inferiority and equivalence trials, non-pharmacological treatments, herbal interventions, and pragmatic trials. Additional extensions are forthcoming: for those and for up to date references relevant to this checklist, see [www.consort-statement.org](http://www.consort-statement.org).

# BMJ Open

## Supporting Parenting at Home – Empowering Rehabilitation through Engagement (SPHERE): Study protocol for a randomized control trial

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Secondary Subject Heading:	Evidence based practice, Patient-centred medicine
Keywords:	Telemedicine < BIOTECHNOLOGY & BIOINFORMATICS, Paediatric pathology < PAEDIATRICS, Developmental neurology & neurodisability < PAEDIATRICS

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Manuscripts

**Supporting Parenting at Home – Empowering Rehabilitation through Engagement  
(SPHERE): Study protocol for a randomized control trial**

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## ABSTRACT

**Introduction.** Infants with neurodevelopmental disabilities (NDs) show emotional, cognitive, and socio-interactive dysregulation dramatically impacting on caregiving behavior. Early video-feedback interventions (VFIs) are effective in promoting sensitive parenting, which in turn supports infants' development, even in case of ND. In the light of limited resources of the healthcare systems, technological advances in telemedicine may facilitate the delivery of VFI to a greater number of families of infants with ND. To date, no study has implemented a telemedicine VFI (TVFI) for families of infants diagnosed with ND. **Methods and analysis.** The Supporting Parenting at Home – Empowering Rehabilitation through Engagement (SPHERE) project is a randomized controlled trial aimed at assessing the effectiveness of an early family-centered TVFI parenting support on dyads with infants diagnosed with ND. It includes two arms (TVFI vs. Booklet Psychoeducational Intervention) and three assessment phases: T0, baseline; T1, immediate post-intervention; T2, 6-month follow-up. **Ethics and dissemination.** This study is funded by the Italian Ministry of Health and was approved by the Ethics Committee (Pavia). Results will be published in peer-reviewed journals and presented at national and international scientific conferences.

**Keywords:** family-centered intervention; maternal sensitivity; neurodevelopmental disability; parenting; telemedicine; video-feedback.

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2 27 **Strengths and limitations of this study**  
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- 5 28 • This RCT will develop, deliver, and test a scalable video-feedback intervention to  
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7 29 improve parental skills and developmental outcomes of infants with neurodevelopmental  
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10 30 disabilities.  
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12 31 • The telemedicine approach of this intervention will allow to reduce the inequality of  
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14 32 access to family-centered care in neurodevelopmental disability.  
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17 33 • Although neurodevelopmental disabilities include a wide range of clinical conditions, this  
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19 34 intervention targets parenting challenges that are generally common and shared among  
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21 35 parents of infants with diverse disability phenotypes.  
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24 36 • The telemedicine nature of the intervention is not free from potential technical issues  
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26 37 related to poor quality of internet signal; nonetheless, no specific resources are needed  
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28 38 on family side which will be able to connect with any device.  
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## INTRODUCTION

Every year, about 53 million infants worldwide receive a diagnosis of neurodevelopmental disability (ND) and they represent the 13% of all health problems in infancy and childhood [1]. Although ND includes a wide range of diverse clinical conditions (e.g., cerebral palsy, genetic or malformative syndromes, outcomes of severe prematurity, autism spectrum disorders), these infants may share common difficulties in emotional regulation, cognitive skills, and socio- interactive abilities [2,3]. Such pattern of multi-systemic dysregulation negatively impacts on parents' psychological and physical health as well as on caregiving behavior [4,5]. Parents may report critical emotional burden with heightened risk for chronic levels of distress, depression and anxiety [4–6]. This constitutes a crucial point considering that parenting represents the frontline of developmental resilience for infants' development, even in the presence of ND conditions. Indeed, infants with ND whose parents are rated high in parental sensitivity show better outcomes in emotional, cognitive and socio- interactive developmental trajectories [7–9].

From this perspective, it is not surprising that early rehabilitation interventions have been found to be the most effective when they engage parents in a family-centered approach [10], and they also are the most rewarding care strategies for healthcare systems in terms of economic return in the long-term [11]. Specifically, the Video Feedback Intervention (VFI) [12] constitutes an early family-centered care strategy that proved to be effective in promoting sensitive parenting and infants' behavioral and socio-emotional adjustment [13]. The use of VFI intervention has been also documented to be beneficial in dyads of children with ND [14–16]. Previous research reported that these interventions may result in reduced child's disruptive and emotionally negative behaviors [17], improved maternal sensitivity [18,19], increased parental self-efficacy and less parenting stress [20]. As such, early

1  
2 63 supportive interventions directed at improving the quality of parental sensitivity and parent-  
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4 64 infant interaction should be prioritized especially in a population of children diagnosed with  
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6 65 ND [10,21].  
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9 66 It should be highlighted that delivering VFI in hospital- or home-based context are highly  
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11 67 demanding for the healthcare systems due to high cost and disparities in access to services  
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13 68 for families who live in remote areas. As such, delivering VFI through telemedicine (TVFI)  
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15 69 approaches using videoconferencing is a valuable option that may reduce care access  
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17 70 inequalities, promote early family-centered care culture, and contribute to a more effective  
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19 71 and efficient healthcare approach to the rehabilitation program of infants with ND [22].  
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21 72 Notably, videoconference approaches to parent training interventions (e.g., cognitive  
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23 73 educational training) showed to be as effective as traditional face-to-face approaches and  
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25 74 were accepted with the same degree of satisfaction by parents [23,24]. Nonetheless, we still  
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27 75 do not know how a TVFI support may end up in being effective in terms of promoting  
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29 76 parental health and infants' development in families of little patients with ND.  
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34 77 **METHODS AND ANALYSIS**

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36 78 **Aims**

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39 79 Given the paucity of telemedicine interventions for parents of children with ND and  
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41 80 limitations of the delivery of home-based or hospitalized Video-feedback interventions, the  
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43 81 SPHERE project was launched. The SPHERE project is a randomized control trial that aims  
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45 82 to develop, deliver, and test the effectiveness of an early TVFI compared to an alternative  
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47 83 educational intervention. The first specific aim is to assess the TVFI effectiveness in  
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49 84 supporting maternal sensitive parenting in dyads of infants with ND. Previous studies  
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51 85 suggested that a traditional VFI increased parental capacity to respond to child's  
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53 86 communicative signals in parents of children with disabilities [18,19]. Consistently, we  
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55 87 hypothesize that the TVFI will be effective in increasing maternal sensitivity (within-group  
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difference) and will be more effective than the alternative not individualized intervention (between-group difference).

The second specific aim is to test the TVFI effectiveness in reducing infants' emotional stress reactivity. In line with a previous study highlighting a significant reduction of emotionally negative behaviors after VFI [17], we hypothesize that the TVFI will be effective in reducing infants' emotional stress in the experimental group. Additional exploratory aims include the assessment of the SPHERE TVFI intervention on other maternal (i.e., parenting stress, depressive symptoms, anxiety symptoms) outcomes and on infants' behavioral regulation (i.e., temperament profile).

### Study design

The SPHERE project is a randomized control trial with two arms and three assessment phases: T0, baseline; T1, immediate post-intervention; T2, follow-up (6 months after the intervention). The experimental arm consists in the telemedicine intervention (TVFI arm), whereas the control arm consists in the delivery of a psychoeducational booklet (PEB arm). For both arms, each assessment session will include (a) an on-line questionnaire on maternal and infants' well-being (see below) and a 15-minute mother-child dyadic interaction will be videotaped in remote. This interaction will include approximately 10-minute face-to-face play interaction followed by a Face-to-Face Still-Face (FFSF) procedure [25]. The FFSF is a well-validated observational procedure to assess infants' socio-emotional regulation and parenting sensitivity that has been previously used with infants with ND [26]. During the FFSF, the mother is asked to interact with her infant for two minutes (Play episode), then to interrupt any communication and to maintain a still poker face for two minutes (Still-Face episode), and finally to resume the interaction for two final minutes. An overview of the study protocol and procedures is reported in figure 1.

---- Please, insert Figure 1 here ----

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2 113 **Population**  
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4 114 The mother-infant dyads will be enrolled at the Child Neurology and Psychiatry Unit of the  
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6 115 IRCCS Mondino Foundation, Pavia (Italy). They will be enrolled consecutively according to  
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9 116 the following inclusion criteria: infants' (corrected) age between 1 and 18 months; presence  
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11 117 of developmental risk for or diagnosis of ND as defined by standardized clinical criteria;  
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13 118 parental age greater than 18 years; parental mastery of Italian language; both parents living  
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15 119 together with the infant. Exclusion criteria include presence of twins, infant's life-threatening  
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18 120 conditions, and maternal psychiatric disorders. The dyad's randomized allocation to each  
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20 121 arm will be done through a computerized 0/1 sequence generator.  
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23 122 **Intervention, TVFI arm**  
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25 123 The TVFI will be standardized according to previously published RCTs [27,28]. The TVFI is  
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27 124 inspired to concepts from the Collaborative Consultation approach from Zack Boukydis [29].  
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29 125 Specifically, six weekly TVFI sessions will be delivered in two subsequent phases, as in  
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31 126 previous VFI research with families of children with ND [30]. Four *sharing the focus* 1-hour  
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33 127 sessions will be dedicated to the discussion between the psychologist and the mother of  
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35 128 specific themes related to parenting and parent-child interaction: physical stimulation,  
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37 129 responsiveness, teaching, and parenting experience (see Table 1). During these sessions,  
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39 130 the psychologist will conduct a dialogic interactive session connecting with the mother in  
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41 131 videoconference. The videotaped interaction obtained at T0 will be discussed by the  
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43 132 principal investigator (SG) and a senior author (LP) who is experienced in mother-infant  
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45 133 interaction coding and early VFI interventions and it will be segmented into specific videoclip  
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48 134 lasting up to 10 seconds. Each videoclip will be labeled with one of the thematic contents  
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51 135 reported in Table 1. During the *sharing the focus* sessions, the psychologist will propose to  
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53 136 the mother to jointly review some of these segments, usually starting from potential  
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55 137 curiosity, comments or requests from the mother herself. The specific order in which the  
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themes will be discussed will be tailored on each specific case. The number of videoclips used for each thematic label in the TVFI of each family will be noted by the psychologist in a diary. The goal of the *sharing the focus* sessions is to develop insights about the infants' behavioral signals, the best ways to provide stimulations and get in touch, strategies to promote emotion regulation and to sustain cognitive and behavioral achievements. In the subsequent two 1-hour *integration* sessions, the mother will play with the infant while the psychologist will provide a dyadic-tailored guidance based on topics previously discussed during the first four sessions. The goal of the *integration* sessions is to promote a pragmatical translation of the insights developed during the *sharing the focus* sessions into the interactive exchanges between the mother and the infant. By doing so, the mother can introduce variations in her caregiving behavior in a safe environment under the supervision of a trained specialist.

---- Please, insert Table 1 here ----

### **Intervention, PEB arm**

Mothers assigned to condition B will receive an informative booklet addressing the same themes discussed in the experimental intervention (i.e., responsiveness, physical stimulation, teaching, and parenting experience), but not tailored on their own infant or specific parenting challenges.

### **Core variables and measures for the specific aims**

Maternal sensitivity will be assessed using the Global Rating Scales (GRS) [31] from the first 5-minute segment of videotaped mother-infant interactions at T0, T1, and T2. The GRS provides indexes of maternal sensitivity and intrusiveness that ranges from 1 (low scores) to 5 (high scores) and require a holistic, macro-analytic coding of the interaction. Infants' emotional stress reactivity will be coded in terms of negative emotionality display (by voice and/or facial expression) across the FFSF procedure videotaped at T0, T1, and T2.

1  
2 163 Negative emotionality display will be coded according to previous system validated in FFSF  
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4 164 research [32]. For both coding, students will be trained using available reference videoclips  
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6 165 according to a gold standard 85% inter-rater agreement. Trained coders will be blind to the  
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9 166 arm allocation and to the specific goals of the intervention.

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11 167 **Other measures (additional exploratory aims)**  
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13 168 Information on the clinical characteristics and the diagnosis of the infant will be obtained  
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15 169 from medical records. By filling in on-line questionnaires at T0, mothers will provide  
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17 170 information on infants' neonatal characteristics (i.e., sex, gestational age in weeks, birth  
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19 171 weight in grams), socio-demographic variables (i.e., maternal age, educational level, and  
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21 172 occupational status). In the same questionnaire as well as in the following questionnaires  
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23 173 administered at T1 and T2, mothers will fill in the 36-item Parenting Stress Index – Short  
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25 174 Form (PSI-SF) [33], the 21-item Beck Depression Inventory (BDI-II) [34], the 20-item state  
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27 175 subscale of the State-Trait Anxiety Inventory (STAI-Y) [35], and the 91-item Infant Behavior  
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29 176 Questionnaire-Revised (IBQ-R) short form [36]. All these questionnaires have been  
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31 177 validated in Italian and they are largely used in parent-infant research in experimental and  
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33 178 clinical settings.  
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39 179 **Statistical power and sample size estimation**  
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41 180 The sample size was estimated for what pertains the first specific aim, i.e., the effect of an  
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43 181 early VFI intervention on maternal sensitivity. A minimum sample size of 59 subjects per  
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45 182 group [ $\alpha = .05$ ,  $\beta = .05$ , power = .95, effect size,  $d = .67$ ] was estimated using  
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47 183 G\*Power software on the basis of meta-analytical evidence of online interventions focused  
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49 184 on parenting [37]. Moreover, due to the longitudinal nature of the study, attrition rate of 20%  
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51 185 for each phase was considered, therefore a starting sample size of 84 infants for each  
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53 186 group (total sample size = 168) has been estimated.  
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57 187 **Plan of statistical analyses**  
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R Studio packages [38] and IBM SPSS 27 [39] will be used for the statistical analyses. Participants with more than 20% missing data in the questionnaires as well as those who did not complete at least T0 and T1 assessments will be excluded from the analyses. TVFI and PEB dyads will be preliminarily compared for socio-demographic and neonatal characteristics. In order to respond to the specific aims of the SPHERE study, standard mixed analyses of variance will be used to test within- and between-group differences in maternal sensitivity and infant's emotional stress reactivity. The difference between T0 and T1 measures will be considered as the primary output of these analyses and long-term maintenance of the effect will be tested by including a third time point (T2) in the model. Advanced modelling will be used to assess the effects of potential mediators/moderators (e.g., infants' sex and age) on maternal and infants' outcome variables. Standard mixed analyses of variance will be used to test within- and between-group differences in maternal parenting stress, depression, anxiety, and infant's behavioral regulation across the three assessments time points.

### **Patient and public involvement**

The intervention was not developed in concert with families or parents. Nonetheless, webinars and educational events dedicated to parents and rehabilitation care professionals will be organized to disseminate the findings of the SPHERE project and to promote a culture of family-centered care in the context of ND healthcare. Moreover, after the end of the RCT, the booklet of the PEB will be distributed within the clinical unit of the IRCCS Mondino Foundation and will become a freely available resource for parents and staff. The booklet will also circulate to families thanks to the engagement of parents' associations that collaborate with authors for educational and clinical purposes.

## **DISCUSSION**

### **Limitations**

1  
2 213 First, the telemedicine nature of this study may imply technical problems during video  
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4 214 recording due to internet instability or to the loss of a good frame of the infant. For example,  
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6 215 the quality of the internet signal may be in some cases low or moderate influencing the  
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8 216 quality of the video output. Moreover, specific characteristics of the child (e.g. high levels of  
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10 217 physical activity) may pose challenges that can make it difficult to keep the interactive  
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12 218 partners perfectly at the center of the recording scene. Mothers will be asked to position the  
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14 219 webcam or smartphone to have the widest possible view of the play area and to see the  
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16 220 entire body of both the mother and the infant. However, it is possible that during the session  
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18 221 infants may come out of the frame or that in some moments the face of participants is not  
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20 222 visible. Therefore, portions of the videotapes might be only partially suitable for micro-  
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22 223 analytic coding. However, the GRS coding system used for the assessment of the primary  
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24 224 outcome of the intervention relies majorly on global scorings that can be performed with  
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26 225 medium quality video output. Moreover, attrition rate has been included in the sample size  
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28 226 estimation also to take care of potential loss of dyads due to technical problems.  
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32 227 Second, as dealing with the ND clinical condition of their son or daughter may be highly  
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34 228 stressful for mothers, it should not be excluded that mothers may express the need of more  
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36 229 specific emotional and psychological needs during the SPHERE project. In these cases,  
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38 230 proper referral to specialists will be discussed with the mother. Third, ND include a wide  
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40 231 ensemble of clinical conditions and the different levels of psychomotor delay as well as the  
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42 232 different domains that may be impaired certainly play a role in affecting maternal sensitivity  
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44 233 and well-being and also impact on the quality of parent-infant interaction. For example, the  
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46 234 presence of a mild psychomotor delay or a severe sensory impairment may pose different  
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48 235 challenges in the daily interactive exchanges. Nonetheless, the heterogeneity of the clinical  
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50 236 conditions is an inherent characteristics of ND and it is often a limit to studies that are  
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52 237 mainly focused on directly promoting specific infants' developmental outcomes. In the  
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present study, the main focus is on supporting maternal sensitivity as a proxy to further promote infants' development and well-being. Moreover, due to the early age at which infants are enrolled in the study, it is highly probably that they will partially share a risk condition for ND and they will not have already received a specific diagnosis. Notwithstanding, when available, a quantitative psychomotor developmental quotient with standardized scales will be obtained from medical records and will concur to define the enrolled sample. Finally, despite the substantial variation in the paternal involvement in caregiving and the need for studies on fathers, the SPHERE project will enroll only mothers as participants, due to practical difficulties (e.g. limited parental leave for fathers) in engaging fathers in the weekly videoconferences foreseen for the experimental arm.

### **Expected results and implications**

The SPHERE project holds promises to test a new model for the telemedicine care and rehabilitation of infants with neurodevelopmental disabilities. By investing in an early and family-centered intervention we aim to provide families with timely and efficient support to the rehabilitation journey, contributing to grow a sense of self-efficacy in the caregivers of infant with ND. Moreover, by promoting a telemedicine VFI we can contribute to reduce the healthcare inequalities for families who face challenges in accessing traditional rehabilitation programs because they live in rural areas or because of long waiting lists. The SPHERE project may provide evidence-base support to the development and application of TVFI approaches that may maximize the benefit of early family-centered interventions for both parents' well-being and infant's development.

### **Ethics and dissemination**

The study has received the approval of the Ethics Committee Pavia on November 2<sup>th</sup>, 2020, Protocol number 20200096046. All the procedures are consistent with the Declaration of

1  
2 263 Helsinki ethical principles for research involving human subjects. The procedures do not  
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4 264 imply any harm to the participating subjects. Moreover, the study interventions represent  
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6 265 additional opportunities for families that do not imply changes in usual mother-infant care  
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9 266 programs. All infants will take part to all the diagnostic and therapeutic interventions that are  
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11 267 planned in the child neurology and psychiatric unit IRCCS Mondino Foundation, Pavia, Italy.  
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13 268 The study protocol has been also registered on NIH Clinical Trials (protocol code  
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15 269 NCT04656483). The dissemination plan includes the presentation of findings at national and  
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18 270 international scientific meetings as well as the publication in scientific journals in the field of  
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20 271 developmental psychology. The findings will also be disseminated to the public through  
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22 272 reach-out activities involving families and healthcare specialists, in order to promote early  
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25 273 family-centered intervention.  
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**Authors' contributions:** All authors conceived the study protocol and the rationale for the project. SG and LP wrote the funding proposal for this study. SG and LP coordinates recruitment of participants and data collection. LP and RB supervise the progress of the study. SG and LP wrote the first version of this manuscript draft. RB provided suggestions for the improvement of the manuscript. All authors read and approved the final manuscript.

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**Competing interests:** The authors declare that they have no competing interests.

**Data availability statement:** The raw data will be made available through an online repository by the authors, upon reasonable request.

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1

2 287 **ABBREVIATIONS**

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- 4 288       • VFI, Video Feedback Intervention
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- 6 289       • ND, Neurodevelopmental Disabilities
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- 9 290       • TVFI, Telemedicine Video Feedback Intervention
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- 11 291       • PEB, Psychoeducational booklet
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- 14 292       • FFSF, Face-to-face Still Face
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- 16 293       • GRS, Global Rating Scales
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- 18 294       • PSI-SF, Parenting Stress Index, Short Form
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- 21 295       • BDI-II, Beck Depression Inventory
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- 23 296       • STAI-Y, State-Trait Anxiety Inventory
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- 25 297       • IBQ-R, Infant Behavior Questionnaire – Revised
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**Table 1.** Description of the TVFI *sharing the focus* sessions' themes

Theme	Sub-themes	Description
1 Stimulation	Type of stimulations	Type of stimulations (e.g., auditory, tactile) preferred by the infant
	Intensity of stimulations	Infant's sensitivity to the intensity of stimulations
	Social touch	Maternal touch in promoting infant's body awareness and attentional orientation
	Sensory integration	Infant's sensory integration and body awareness
2 Responsiveness	Sense of agency	Supporting the infant's initiative to promote the development of his sense of agency
	Sensitivity	Perceiving and interpreting child's signals and responding in a prompt and appropriate way
	Exploration	Supporting the child's exploration and his use of the parent as a secure base
	Reparation	Repairing communicative ruptures
3 Teaching	Attention skills	Supporting the infant's attention orientation
	Modelling	Providing a model to the infant in order to foster the observational learning
	Scaffolding	Parental guidance to allow the infant to solve a task that he cannot yet carry out on his own
	Proximal development zone	Encouraging learning in the infant's proximal development zone
4 Parenting experience	Representations of the baby	Maternal representations of the infant and curiosity about his mind
	Self-regulation	Taking care of herself
	Self-efficacy	Mother's sense of efficacy and trust in her own experience

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**FIGURE LEGENDS**

**Figure 1.** Schematic overview of the study design.

For peer review only

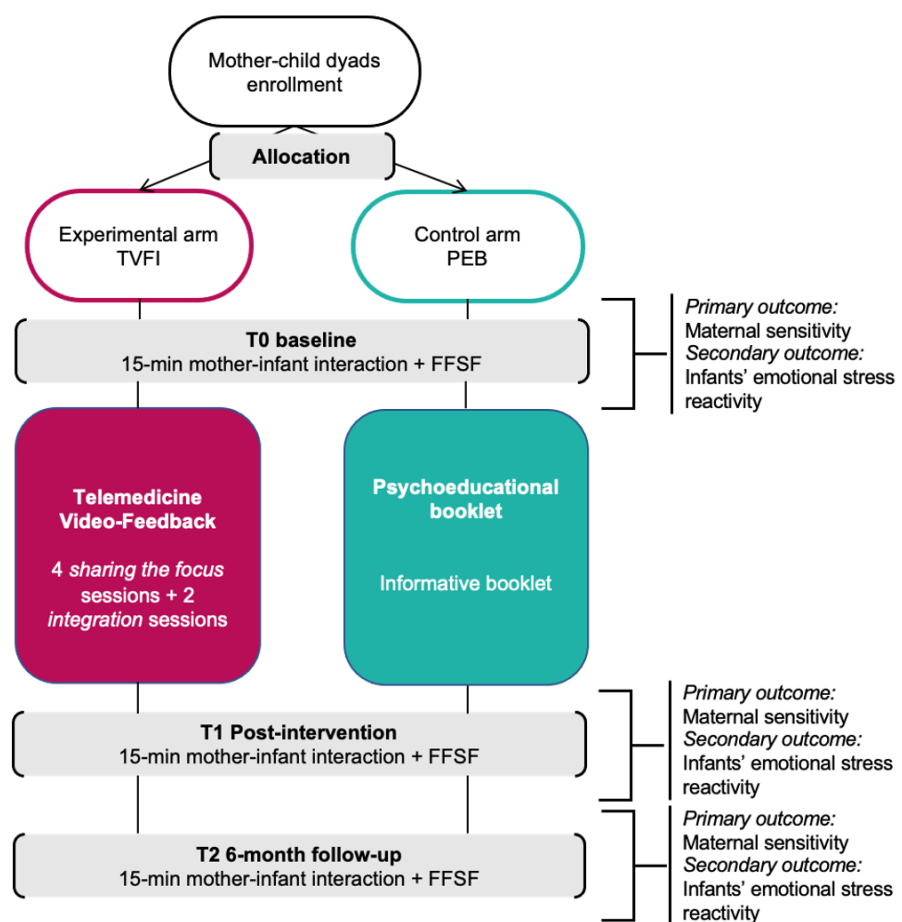


Figure 1. Schematic overview of the study design.

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SPIRIT 2013 Checklist: Recommended items to address in a clinical trial protocol and related documents\*

Section/item	Item No	Description	Reported on page number
<b>Administrative information</b>			
Title	1	Descriptive title identifying the study design, population, interventions, and, if applicable, trial acronym	1
Trial registration	2a	Trial identifier and registry name. If not yet registered, name of intended registry	Registered on NIH CT p. 12
	2b	All items from the World Health Organization Trial Registration Data Set	NA
Protocol version	3	Date and version identifier	NA
Funding	4	Sources and types of financial, material, and other support	13
Roles and responsibilities	5a	Names, affiliations, and roles of protocol contributors	1; 13
	5b	Name and contact information for the trial sponsor	NA
	5c	Role of study sponsor and funders, if any, in study design; collection, management, analysis, and interpretation of data; writing of the report; and the decision to submit the report for publication, including whether they will have ultimate authority over any of these activities	NA
	5d	Composition, roles, and responsibilities of the coordinating centre, steering committee, endpoint adjudication committee, data management team, and other individuals or groups overseeing the trial, if applicable (see Item 21a for data monitoring committee)	NA

**Introduction**

Background and rationale	6a	Description of research question and justification for undertaking the trial, including summary of relevant studies (published and unpublished) examining benefits and harms for each intervention	4-6
	6b	Explanation for choice of comparators	8
Objectives	7	Specific objectives or hypotheses	5-6
Trial design	8	Description of trial design including type of trial (eg, parallel group, crossover, factorial, single group), allocation ratio, and framework (eg, superiority, equivalence, noninferiority, exploratory)	6
<b>Methods: Participants, interventions, and outcomes</b>			
Study setting	9	Description of study settings (eg, community clinic, academic hospital) and list of countries where data will be collected. Reference to where list of study sites can be obtained	7
Eligibility criteria	10	Inclusion and exclusion criteria for participants. If applicable, eligibility criteria for study centres and individuals who will perform the interventions (eg, surgeons, psychotherapists)	7
Interventions	11a	Interventions for each group with sufficient detail to allow replication, including how and when they will be administered	7-8
	11b	Criteria for discontinuing or modifying allocated interventions for a given trial participant (eg, drug dose change in response to harms, participant request, or improving/worsening disease)	11
	11c	Strategies to improve adherence to intervention protocols, and any procedures for monitoring adherence (eg, drug tablet return, laboratory tests)	NA
	11d	Relevant concomitant care and interventions that are permitted or prohibited during the trial	12
Outcomes	12	Primary, secondary, and other outcomes, including the specific measurement variable (eg, systolic blood pressure), analysis metric (eg, change from baseline, final value, time to event), method of aggregation (eg, median, proportion), and time point for each outcome. Explanation of the clinical relevance of chosen efficacy and harm outcomes is strongly recommended	8-9

Participant timeline	13	Time schedule of enrolment, interventions (including any run-ins and washouts), assessments, and visits for participants. A schematic diagram is highly recommended (see Figure)	6, figure 1
Sample size	14	Estimated number of participants needed to achieve study objectives and how it was determined, including clinical and statistical assumptions supporting any sample size calculations	9
Recruitment	15	Strategies for achieving adequate participant enrolment to reach target sample size	7

Methods: Assignment of interventions (for controlled trials)

Allocation:

Sequence generation	16a	Method of generating the allocation sequence (eg, computer-generated random numbers), and list of any factors for stratification. To reduce predictability of a random sequence, details of any planned restriction (eg, blocking) should be provided in a separate document that is unavailable to those who enrol participants or assign interventions	7
Allocation concealment mechanism	16b	Mechanism of implementing the allocation sequence (eg, central telephone; sequentially numbered, opaque, sealed envelopes), describing any steps to conceal the sequence until interventions are assigned	NA
Implementation	16c	Who will generate the allocation sequence, who will enrol participants, and who will assign participants to interventions	NA
Blinding (masking)	17a	Who will be blinded after assignment to interventions (eg, trial participants, care providers, outcome assessors, data analysts), and how	9
	17b	If blinded, circumstances under which unblinding is permissible, and procedure for revealing a participant's allocated intervention during the trial	NA

Methods: Data collection, management, and analysis

Data collection methods	18a	Plans for assessment and collection of outcome, baseline, and other trial data, including any related processes to promote data quality (eg, duplicate measurements, training of assessors) and a description of study instruments (eg, questionnaires, laboratory tests) along with their reliability and validity, if known. Reference to where data collection forms can be found, if not in the protocol	8-9
	18b	Plans to promote participant retention and complete follow-up, including list of any outcome data to be collected for participants who discontinue or deviate from intervention protocols	NA
Data management	19	Plans for data entry, coding, security, and storage, including any related processes to promote data quality (eg, double data entry; range checks for data values). Reference to where details of data management procedures can be found, if not in the protocol	12
Statistical methods	20a	Statistical methods for analysing primary and secondary outcomes. Reference to where other details of the statistical analysis plan can be found, if not in the protocol	9-10
	20b	Methods for any additional analyses (eg, subgroup and adjusted analyses)	9-10
	20c	Definition of analysis population relating to protocol non-adherence (eg, as randomised analysis), and any statistical methods to handle missing data (eg, multiple imputation)	10
<b>Methods: Monitoring</b>			
Data monitoring	21a	Composition of data monitoring committee (DMC); summary of its role and reporting structure; statement of whether it is independent from the sponsor and competing interests; and reference to where further details about its charter can be found, if not in the protocol. Alternatively, an explanation of why a DMC is not needed	NA
	21b	Description of any interim analyses and stopping guidelines, including who will have access to these interim results and make the final decision to terminate the trial	NA

Harms	22	Plans for collecting, assessing, reporting, and managing solicited and spontaneously reported adverse events and other unintended effects of trial interventions or trial conduct	12
Auditing	23	Frequency and procedures for auditing trial conduct, if any, and whether the process will be independent from investigators and the sponsor	NA
<b>Ethics and dissemination</b>			
Research ethics approval	24	Plans for seeking research ethics committee/institutional review board (REC/IRB) approval	12
Protocol amendments	25	Plans for communicating important protocol modifications (eg, changes to eligibility criteria, outcomes, analyses) to relevant parties (eg, investigators, REC/IRBs, trial participants, trial registries, journals, regulators)	12
Consent or assent	26a	Who will obtain informed consent or assent from potential trial participants or authorised surrogates, and how (see Item 32)	12
	26b	Additional consent provisions for collection and use of participant data and biological specimens in ancillary studies, if applicable	NA
Confidentiality	27	How personal information about potential and enrolled participants will be collected, shared, and maintained in order to protect confidentiality before, during, and after the trial	12
Declaration of interests	28	Financial and other competing interests for principal investigators for the overall trial and each study site	13
Access to data	29	Statement of who will have access to the final trial dataset, and disclosure of contractual agreements that limit such access for investigators	13
Ancillary and post-trial care	30	Provisions, if any, for ancillary and post-trial care, and for compensation to those who suffer harm from trial participation	11; 12

Dissemination policy	31a	Plans for investigators and sponsor to communicate trial results to participants, healthcare professionals, the public, and other relevant groups (eg, via publication, reporting in results databases, or other data sharing arrangements), including any publication restrictions	12
	31b	Authorship eligibility guidelines and any intended use of professional writers	NA
	31c	Plans, if any, for granting public access to the full protocol, participant-level dataset, and statistical code	13
<b>Appendices</b>			
Informed consent materials	32	Model consent form and other related documentation given to participants and authorised surrogates	Supplementary
Biological specimens	33	Plans for collection, laboratory evaluation, and storage of biological specimens for genetic or molecular analysis in the current trial and for future use in ancillary studies, if applicable	NA

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\*It is strongly recommended that this checklist be read in conjunction with the SPIRIT 2013 Explanation & Elaboration for important clarification on the items. Amendments to the protocol should be tracked and dated. The SPIRIT checklist is copyrighted by the SPIRIT Group under the Creative Commons "[Attribution-NonCommercial-NoDerivs 3.0 Unported](https://creativecommons.org/licenses/by-nc-nd/3.0/)" license.