Early intervention programmes for infants at high risk of atypical neurodevelopmental outcome

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The purpose of this review is to present a new framework, EI SMART (early intervention: sensorimotor development, attention and regulation, relationships, and therapist support) for identifying key components that could contribute to more effective interventions for infants at high risk of atypical neurodevelopmental outcome. We present a clinical consensus of current challenges and themes in early intervention, based on multidisciplinary group discussions, including parents of high-risk infants, supported by a literature review. Components to include in early intervention programmes are: (1) promotion of self-initiated, developmentally appropriate motor activity; (2) supporting infant self-regulation and the development of positive parent-infant relationships; and (3) promotion of early communication skills, parent coaching, responsive parenting, and supporting parental mental well-being. Such multimodal programmes may need to be evaluated as a package.

Improvements in neonatal intensive care have been associated with increased survival of very low birthweight and infants born preterm; however, morbidity remains high. There is an increased risk of cognitive and/or behavioural impairments, speech and language delay, and sensorimotor deficits, including cerebral palsy (CP). This greatly increases the emotional and financial burdens on families, society, and health care systems.

Multiple factors have an impact on cognitive, behavioural, language, and sensorimotor outcomes in infants born preterm. Periventricular leukomalacia, grades 3 to 4 intraventricular haemorrhage, and bronchopulmonary dysplasia are associated with a high risk of CP. There are also justifiable concerns about the impact of environmental factors in the neonatal intensive care unit (NICU) on infants born preterm, including noise, bright lights, medical equipment, and altered social interactions. Loud noises and bright lights have short-term negative effects on weight gain and cardiorespiratory parameters; these factors influence developmental outcomes. Isolation in an incubator and skin-to-skin caregiver contact are very different sensory experiences that affect cardiorespiratory parameters and sleep organization. Non-medical factors including sensory experiences and feeling information. These interventions are underpinned by an ethos of individually tailored collaborative care,
developmentally supportive environments, and support for the parent–infant relationship. All of these interventions are supported by evidence from large-scale, randomized-controlled trials.

Early intervention strategies that extend beyond discharge from the NICU are also critical to ameliorating the negative effects of preterm birth on parenting, parent–infant interactions, and developmental outcomes. Details of parent-focused early intervention programmes extending beyond the NICU are provided in a narrative review by van Wassenaer-Leemhuis et al., which considers the multimodal nature of early intervention strategies and issues around practicalities of implementation and evaluation.

Since 2016, a multidisciplinary, UK-based group of early intervention therapists (occupational therapy, physiotherapy, speech and language therapy), a paediatric neurologist, a child psychotherapist with an interest in early intervention, and parents of high-risk infants, have convened to consider the largely fragmentary nature of the evidence around early intervention and propose how a more integrated approach could be implemented. The group was initiated by one of the authors (BHI) who, from clinical experience and awareness of increasing interest in early intervention in other centres worldwide, became strongly motivated to ensure UK-based infants and their families receive optimal early intervention services. She, therefore, invited colleagues from relevant disciplines to form a group addressing this issue.

This paper addresses the gap in current literature about which early interventions facilitate the best outcomes for high-risk infants. Identifying interventions that can reliably improve global development across domains is crucial, as the cost of minimal efficacy of early intervention is high in terms of quality of life for families. We propose a clinical reasoning framework for use in early intervention settings with high-risk infants.

WHAT IS AN EARLY INTERVENTION PROGRAMME?
An early intervention programme for high-risk infants typically begins within the first year of life. All early development (cognitive, motor, language and communication, and growth) is influenced by social and emotional development through caregiving relationships. Interventions during this period have a high potential to impact positively on neurodevelopmental outcomes. Early intervention aims ‘to promote child health and well-being, enhance emerging competencies, minimize developmental delays, remediate existing or emerging disabilities, prevent functional deterioration, and promote adaptive parenting and overall family function’. A meta-analysis showed improvement in cognitive outcomes up to preschool age and in early motor outcomes in infants born preterm with early intervention, however, the effect on motor outcomes was small. Furthermore, fewer than half of the studies provided information on motor outcomes at preschool and school age.

<table>
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<th>What this paper adds</th>
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<td>• Early intervention programmes should address sensorimotor development, attention, self-regulation, and early communication skills.</td>
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<td>• Therapist input to the programme should include parent coaching and support for parental mental well-being.</td>
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ATTRIBUTES OF EARLY INTERVENTION PROGRAMMES
Environmental enrichment
Increasingly, early intervention programmes specify ‘environmental enrichment’. The term derives from animal studies describing an environment that facilitates enhanced cognitive, motor, and sensory stimulation in comparison to standard housing conditions. In their review of the effectiveness of early intervention programmes for infants with CP, Morgan et al. operationally defined environmental enrichment as ‘interventions that aim to enrich at least one of the motor, cognitive, sensory, or social aspects of the infant’s environment for the purposes of promoting learning’. Environmental enrichment components of interventions are often implicit in their approaches but have rarely been overtly described.

Responsive parenting: attention, regulation, and relationships
It is key that health professionals create a central and ongoing role for parents in their infant’s care, starting in the NICU, to foster parental feelings of inclusion, helpfulness, efficacy, and control. Post-discharge early intervention programmes must also focus on nurturing the parent–infant relationship by promoting responsive parenting. Increased parent–infant attunement and responsive parenting are associated with improved developmental outcomes and increased resilience in the child. Given the high prevalence of poor parental mental health outcomes, which have an impact on the ability to provide responsive parenting, it is important for effective parental psychosocial support to be available where needed.

Within the setting of a strong parent–infant relationship, the infant learns self-regulatory skills (e.g. crying but being consolable; self-soothing; state changes between sleeping and waking; feeding). Various structured assessments appraise these adaptive behavioural capabilities, such as the Neonatal Behavioural Assessment Scale, Newborn Behavioural Observations, and the NICU Network Neurobehavioral Scale. The NICU Network Neurobehavioral Scale was developed from the Neonatal Behavioural Assessment Scale; its advantages are its strong psychometric properties and the availability of standardized norms. Administration of the Neonatal Behavioural Assessment Scale and NICU Network Neurobehavioral Scale sensitizes the therapist to the complex dynamics of infant behaviour, physiological stability, and attempts at self-regulation. The Newborn Behavioural Observations, which primarily addresses relationship building, is a shorter, clinical tool, allowing the clinician to engage with the family in observing and interpreting the infant’s behaviour and
communication cues. These approaches provide a framework that inherently includes parents by providing information about infant behaviours and ways to modify interactions to match infant needs. They provide structured feedback about the infant’s adaptive behavioural development and regulation that can support sensitive, contingent, and responsive parent caregiving, promote a parental sense of competence, and help parents make informed choices about caring for their infant. A Cochrane review found a significant improvement in parent-infant interactions in the intervention group (Neonatal Behavioural Assessment Scale/Newborn Behavioural Observations) compared with controls, though individual studies were of poor quality.

Self-regulation in early development is embedded in the child’s relationships with others. Beginning with attachment to primary caregivers, early relationships are the scaffold on which cognitive, linguistic, emotional, and social development unfolds. A critical element of good caregiving is to provide a supportive environment, enabling the child to take over and self-regulate in one area of functioning after another. The Infant Behavioral Assessment and Intervention Program is an effective intervention focusing on supporting infant self-regulatory competence and responsive parenting that uses similar approaches to the Newborn Individualized Developmental Care and Assessment Program but beyond the neonatal period. In a randomized-controlled trial, Infant Behavioral Assessment and Intervention Program for very low birthweight infants until 6 months’ corrected age led to improved Performance IQ, visuomotor skills, and certain gross motor skills at age 5 years 6 months compared with standard care.

Emotional regulation is strongly associated with the development of attentional control. Attention is defined as achieving and maintaining an alert state. Parenting that is sensitive to the infant’s signals of interest and distress, that participates in collaborative play, helping the infant to engage with challenging situations and persist in achieving their goals, will improve attention and emotional regulation. Attention promotes learning by allowing the infant to engage in activities for longer periods of time and to shift attention flexibly. Self-regulatory behaviours are critical early mediators of successful development of emotional reactivity, attention and activity level, school readiness, and cognition. A large cohort study compared sustained attention, temperament, and maternal sensitivity at 10 months with self-regulation outcomes at 18 months. Early ability to engage in sustained attention was positively correlated with regulation outcomes. Moreover, responsive maternal behaviour predicted better infant regulatory behaviour and a longer latency to infant distress. The findings highlight the importance of the caregiving environment in infant development.

Sustained attention develops from the earlier emergence of alertness, spatial attention, and feature identification. Deficits in sustained attention are likely to emerge from a complex interaction between biological factors and the individual’s prenatal and postnatal environment, including cascading effects resulting from atypical sensory and motor interactions. Effective early intervention could help limit such escalation of difficulties.

**Communication**

Programmes such as PremieStart (an adaptation of the Mother Infant Transaction Programme), which focus on increasing maternal sensitivity to infant behavioural cues, show a promising positive impact on maternal–infant synchrony at term age and on developing communication skills up to 6 months corrected age (Table S1).

Communication development begins in utero and continues throughout infancy, childhood, and into early adulthood. Fetuses are surrounded by the maternal voice; in comparison, infants born preterm in the NICU are relatively deprived of maternal voice and opportunities for early language learning. Language exposure at 32 and 36 weeks’ gestation affects infant vocalizations and conversational turn-taking, with effects on communication outcomes at 7 and 18 months. This highlights the importance of parental presence and input during the neonatal period to facilitate early language acquisition. However, communicating through incubator walls over the sounds of monitors can feel embarrassing and pointless, and lack intimacy and spontaneity. Parents may require education, encouragement, and permission to engage in behaviours such as talking and singing to their infants in the NICU, which would feel acceptable and natural in a private setting.

Between 6 months and 2 years there is development of gesture, early vocalizations and linguistic communication, and symbolic and functional play, all of which require a responsive communication environment. Despite this, most early language intervention programmes begin at 2 years of age, which may not be optimal.

**Sensory development**

Sensory information has an important role in all areas of development throughout the first year of life. Infants use sensory information to modify movement and postural control from the first months of life. However, the NICU environment presents sensory challenges, with prolonged and excessive exposure to auditory, visual, and tactile stimulation during a period of critical brain development, potentially leading to difficulties with sensory modulation. Longitudinal studies have identified a relationship between the sensory profiles of children born very preterm and neurodevelopmental outcomes at 2 years of age. Abnormalities of basic sensory processing and adaptive responses are likely to lead to later problems with more complex developmental and cognitive processes. It is important to adapt the NICU environment, where possible, to avoid exposing the infant to potentially harmful sensory stimuli and to offer more helpful stimuli. For example, infant massage was shown to promote electroencephalogram-based surrogate markers of brain maturation.
Evidence for the efficacy of interventions aiming to improve sensory processing remains weak, partly due to study methodology issues. Robust research findings in this field would be welcome. In the absence of such evidence, parents and practitioners should be aware of how the individual infant may respond to sensory input and of any limitations in sensory processing (including visual and hearing impairments), how these may impact on behaviour, and how to ameliorate the situation, including modification of the environment and interactions with the infant as necessary.

**Motor learning: active ‘scaffolding’**

Motor skill development is dictated by a mixture of genetic and environmental influences and can be profoundly altered by insults to the developing brain. The challenge is to find effective approaches to optimize motor outcomes after such insults. Interventions are being shaped by the increased ability to identify infants at high risk of developmental delay and those with emerging CP.

Postural control strategies and movements of typically developing infants when learning new skills are highly variable. This variability allows for selection and adaptation of the most effective motor patterns through afferent feedback and active learning. This process has been described using Neuronal Group Selection Theory and helps elucidate why active exploration on the part of the infant facilitates motor learning. Infants with brain lesions have a more limited movement repertoire. These infants also need more task-specific practice than typically developing infants to learn a motor task. Intervention strategies that provide ample opportunities for trial and error, active exploration, strategic use of postural support, and considerable repetition of developmentally appropriate motor skills, are key to motor learning. For some infants with severe CP, possibly with associated cognitive deficits, finding the most appropriate mix of sensory input and environmental enrichment that results in active, self-initiated movements is challenging but made possible by breaking down the task and setting realistic intervention goals.

The Goals Activity Motor Enrichment programme is an example of a multifaceted approach for infants born preterm and at term at high risk of CP, with early encouraging results. In a randomized-controlled trial with 30 participants receiving standard care or Goals Activity Motor Enrichment for 16 weeks in the first year of life, motor skills, cognitive skills, and parental satisfaction scores were higher in the intervention group. The approach encourages active infant participation, is goal-oriented, home-based, and emphasizes parent education. Scaffolding strategies are explored with parents, i.e. tasks and goals are tailored to the infant’s developmental stage. As progress is made, physical help is gradually reduced or the environment adapted to ensure the infant remains challenged but able to participate with self-initiated movement.

The coping with and caring for infants with special needs (COPCA) approach emphasizes the importance of parent coaching in solution-focused approaches to facilitate active infant participation. A randomized-controlled trial comparing COPCA with traditional infant physiotherapy for high-risk infants born preterm and at term found no difference in motor outcomes up to age 18 months. Process evaluation revealed, however, that for infants who developed CP, motor outcomes correlated with the time spent by physiotherapists in coaching caregivers and with time spent challenging the infant to produce active motor behaviours. At follow-up, COPCA-trained parents retained a trial-and-error approach when their school-age children were learning new skills. Providing rigid instructions to parents regarding intervention approaches was associated with worse motor outcomes at school age. However, application of these findings to infants with CP may be limited because of the relatively low proportion of infants in the study who ultimately developed CP.

To date there has been no single early intervention approach designed specifically for use by physiotherapists, occupational therapists, and speech and language therapists that encompasses all of the above evidence-based ‘ingredients’. Similarly, there is no single approach built around a central strategy of parental inclusion, education, and empowerment beyond the NICU, to maximize the effectiveness of early intervention for the infant. Typically, programmes are delivered by individual therapists who focus on their professional area of expertise (e.g. physiotherapists in COPCA). Parents may not know what to expect from service providers and may (potentially correctly) perceive different therapists as having competing intervention agendas. The focus needs to shift towards the holistic needs of the infant and family. A new approach is needed to produce collaborative, evidence-based services with a common purpose, supporting professional development, placing parental empowerment and education at the centre of early intervention strategies, and bringing about a change in practice.

**THE EI SMART APPROACH**

We propose ‘early intervention: sensorimotor development, attention and regulation, relationships, and therapist support’ (EI SMART) as a clinical reasoning framework, derived from consensus clinical expertise in partnership with parents and supported by current evidence. The framework was developed to encourage practitioners working with infants born preterm and other high-risk infants to work collaboratively with parents with the aim of optimizing and making manageable, early interventional support for infants and their families, as well as identifying their own professional development requirements. Articulating the components enables therapists to support their consideration of the interplay of these elements and how they may impact an infant’s development and the parent-infant relationship, which will underpin the planning and delivery of their specific therapeutic intervention. As such, EI SMART can
promote a multidisciplinary, team-working ethos with the knowledge, skills, and attitudes to undertake appropriate assessments and work with parents to identify and address the needs of the child and family in a tailored programme.

We consider the following as core components: (1) Actively involving, educating, and empowering parents in biopsychosocial aspects of their infant’s care; (2) Supporting a consistent and responsive parent–infant relationship; (3) Recognizing, supporting, and promoting the infant’s self-regulatory behaviours; (4) Scaffolding the infant’s next developmental cognitive, motor, sensory, and communication steps to stimulate and elicit active participation; (5) Modifying the infant’s environment to ensure the infant remains challenged and able to participate in a wide variety of self-initiated, self-produced motor activities in a variety of conditions; and (6) Promoting parental well-being.

Spittle and Treyvaud also provide recommendations regarding key components of early intervention (e.g. when to commence, where to deliver, who should receive it, content, and duration). In addition to their suggestions, it is important to consider who should deliver early intervention. We propose that early intervention should not be delivered in a time-limited way by one specialist therapist. Infants and parents will benefit from an integrated approach within the therapy team. Physiotherapists, occupational therapists, and speech and language therapists, along with other health care professionals as appropriate, bring their unique skills, knowledge, and expertise to the family (who bring their own vast situational knowledge, motivation, and relationship with the infant) but in a coordinated approach with a lead contact, making intervention coherent and preventing overburdening. A ‘one-size-fits-all’ approach does not work for high-risk infants and their families: interventions must be tailored to individual needs and circumstances. This is achievable with a strong, multidisciplinary team, good communication, appropriate assessments, and sensitivity to individual situations.

Tailoring of interventions creates challenges for formal evaluation of therapy. In clinical practice, however, a pragmatic approach may be required, based on a combination of elements that are effective in isolation. Evaluation of the contribution of individual components to the overall outcome may not be possible, but this does not preclude evaluation of the intervention package (e.g. the effect of having an early intervention team on child and family outcomes). Plans for development, evaluation, and implementation must be made in partnership with parents. We are currently exploring parent views of their involvement in early intervention, barriers, and facilitators to parent involvement, and the influence of the parent–therapist partnership, before formal evaluation of the EI SMART approach.

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SUPPORTING INFORMATION

The following additional material may be found online:

Table S1: Examples of developmentally supportive care interventions in the neonatal intensive care unit

REFERENCES


