INPLASY PROTOCOL

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Early intervention for vision and neurodevelopment in infants and very young children with visual impairment: a systematic review

Dale, NJ[1] Khan, A[2]; Jenter, S[3].

Review question / Objective: **Research question - What is the effectiveness of Early Childhood Intervention (ECI) in the first 3 years of life? Population (P) Infants and very young children with diagnosed visual impairment. Intervention (I) ECI programmes that includes vision and developmental stimulation, play, learning and responsive parenting Comparison (C) Standard care or control Outcomes (O) Primary: Vision function or and/or neurodevelopment and/or parent-child interaction outcomes Secondary: Parental context factors eg parental wellbeing and mental health, parental satisfaction with service provision.**
Condition being studied: **Childhood congenital or very early visual impairment arising from congenital disorders of the peripheral or anterior visual system or cerebral-based vision disorders. This includes all vision disorders of the globe, retina and anterior optic nerve and all vision disorders that are considered cerebral based along visual pathways that are retro-chiasmatic and include central brain regions and networks involved in vision processing.**

INPLASY registration number: This protocol was registered with the International Platform of Registered Systematic Review and Meta-Analysis Protocols (INPLASY) on 21 August 2022 and was last updated on 21 August 2022 (registration number INPLASY202280080).

INTRODUCTION

Review question / Objective: **Research question - What is the effectiveness of Early Childhood Intervention (ECI) in the first 3 years of life? Population (P) Infants and very young children with diagnosed visual impairment. Intervention (I) ECI programmes that includes vision and developmental stimulation, play, learning and responsive parenting Comparison (C) Standard care or control Outcomes (O) Primary: Vision function or and/or neurodevelopment and/or parent-child interaction outcomes Secondary: Parental context factors eg parental wellbeing and**

mental health, parental satisfaction with service provision.

Rationale: Infants and very young children with visual disorder and visual impairment are at high risk of developmental delays, difficulties, deceleration and setback and emerging intellectual disability and social and communication difficulties/ autism spectrum disorder. The development of the visual system is also constrained by the underlying impairment but may also be affected by lack of appropriate environmental stimulation and training. The parenting context is also at risk with parents having greater difficulty in interacting with their infant with visual impairment and also at risk of parenting stress and poorer mental health. Early childhood intervention (ECI) programmes are potentially beneficial in improving the vision function, neurodevelopmental and parent-child interactional outcomes in this at risk clinical population. Professional providers and policy-makers need information in relation to which ECI programmes are more effective with this population, to provide evidence-based guidance to support their design and delivery of appropriate, feasible and more effective ECI programmes.

Condition being studied: Childhood congenital or very early visual impairment arising from congenital disorders of the peripheral or anterior visual system or cerebral-based vision disorders. This includes all vision disorders of the globe, retina and anterior optic nerve and all vision disorders that are considered cerebral based along visual pathways that are retro-chiasmatic and include central brain regions and networks involved in vision processing.

METHODS

Search strategy: 1. PubMed ((((((((((("early intervention education[MeSH Terms]) OR ("early intervention"[Title/Abstract])) OR (early[Title/Abstract])) OR (intervention[Title/Abstract])) OR (interventions[Title/Abstract])) AND ("early intervention education[MeSH Terms])))) OR ("early intervention"[Title/Abstract])) OR (rehabilitation[MeSH Terms])) OR (rehabilitation[Title/Abstract])))) AND (((((("vision impairment"[Title/Abstract]) OR ("visual impairment"[Title/Abstract])) OR ("vision disorder*"[Title/Abstract])) OR (blindness[Title/Abstract])) OR ("low vision"[Title/Abstract])) Filter: 2000-2021 (Publication date), Humans, English Language, Age filter – Newborn: birth-1 month, Infant: birth-23 months, infant:1-23 months, Preschool child:2-5years Databases included: PubMed, EMBASE, Scopus.

Participant or population: population is children with diagnosed visual impairment of any visual disorder, 2) child age is 0-3 years at baseline of study; the strategy for studies with a subset of eligible participants (for example part of the sample is above the age cut-off) was to include studies with all children under the age of 5 years and at least half under the age of 3 years at baseline,

Intervention: Early Childhood Intervention (ECI) involving specified intervention treatment or programme focuses on therapeutic, neurodevelopmental, educational or parenting approaches and not medical or surgical methods,

Comparator: Standard care and control or comparison group of other intervention or no intervention

Study designs to be included: RCTs and non-randomised intervention studies including observational cohort, case-control and comparison trials without full randomisation.

Eligibility criteria: Quantitative analysis was conducted, with participant numbers of at least n=20 in each study group, 7) scientific peer-reviewed publications of completed trials only.Exclusion criteria included failure to meet any of the above inclusion criteria. Grey matter including chapters, conference proceedings, editorial comments/ opinion articles, review papers and discussion papers were excluded. Previous systematic

reviews of relevance were selected at abstract and title stage but not at full-text analysis stage: they were read for information and were cited in the discussion of the paper where relevant. Study trials that are registered but not completed and published at the time of this study were omitted.

Information sources: Information sources are electronic databases (PubMed, SCOPUS and EMBASE), trial register (CENTRAL) and Google Scholar search for any recent papers which have not reached electronic databases. Reference list of relevant papers will be scrutinised to check for any further papers. The time period of interest is 2000 to present day.

Main outcome(s): Outcomes of the review are effect measures in relation to neurodevelopment: cognition, language, behaviour, adaptation and vision function: fixation, detection, saccades, acuity, perception. These will be measures undertaken at final outcome during follow up. The timing of these measures are likely to be variable according to design of study, but expected to be under 4 years of age.

Additional outcome(s): Outcomes of the review are effect measures in relation to neurodevelopment: cognition, language, behaviour, adaptation and vision function: fixation, detection, saccades, acuity, perception. These will be measures undertaken at final outcome during follow up. The timing of these measures are likely to be variable according to design of study, but expected to be under 4 years of age.

Data management: Records and data will be managed on EndNote reference manager. Spreadsheet Excel database will be used to store decisions in relation to Inclusion and Exclusion following full-text analysis of the final papers.

Quality assessment / Risk of bias analysis: Quality assessment will be undertaken using the Risk of Bias (ROB-2) tool for RCT trials and Risk of Bias in Non-randomised Studies for Non-randomised studies including observational, cohort, case-controlled and trials lacking full randomisation (ROBINS-I). The ROBINS-I template including signalling questions will be used.

Strategy of data synthesis: The extracted data (findings of individual studies) will be presented on a data extraction matrix table with appropriate headings covering all domains of relevance and combined and evaluated for data synthesis. This will be done through synthesis according to SWiM guidance (synthesis without meta-analysis) or meta-analysis if feasible.

Subgroup analysis: Of interest is to consider separately and compare ECI programme outcomes in children with congenital disorders of the peripheral or anterior visual system and children with congenital/ early cerebral visual disorders. Also of interest is comparison according to whether the infant has no vision or light perception at best (profound visual impairment) and some basic 'form' vision (severe visual impairment or better). Further vulnerable subgroups will be considered if data available including those with greater brain complexity (with and without known brain disorder as part of paediatric diagnosis) and those with 'developmental setback' (showing plateau'ing and deceleration or regression in the second to third year of life).

Sensitivity analysis: A sensitivity analysis will be undertaken by examining whether any change on a factor of the search or selection criteria and running the analysis again leads to any change in final decisions.

Language restriction: Restricted to English language due to lack of translation resources.

Country(ies) involved: United Kingdom.

Keywords: early intervention; education, early intervention; vision training; vision; neurodevelopment; infant; child; toddler; cognition; behaviour; parenting; parental sensitivity; parenting stress.

Dissemination plans: Publication in scientific peer-reviewed journal. Currently submitted and under review by Developmental Medicine Child Neurology.

Contributions of each author:

Author 1 - Naomi Dale - Author 1 trained and supervised the reviewer team, assisted in consensus decisions when disagreement between the other Author reviewers and drafted the manuscript.
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Author 2 - Aneesa Khan - Author 2 undertook the initial search and also undertook independent selection of papers and critical appraisal using risk of bias tool. Author 2 undertook inter-rater reliability testing and prepared final documents for publication. Author 2 maintained the spreadsheet database of the papers, including inclusion and exclusion decisions.
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Author 3 - Sophie Jenter - Author 3 undertook second independent search and also undertook independent selection of papers and critical appraisal using risk of bias.
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Conflicts of interest: The Principal Investigator (ND) receives royalties for license usage of the Developmental Journal for babies with visual impairment (DJVI), which is an Early Childhood Intervention programme for babies and young children with visual impairment.