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Cerebral palsy research news

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Interventions and Management

1. Effects of botulinum toxin injections in the upper limbs of children with cerebral palsy: A systematic review of the literature
Céline Klein, Richard Gouron, Vincent Barbier

Review Orthop Traumatol Surg Res. 2023 Feb 6;103578. doi: 10.1016/j.otsr.2023.103578. Online ahead of print.

Background: Spasticity is a common motor disorder in children with cerebral palsy (CP). Upper limb CP impairment has a significant negative impact on daily activities. Botulinum toxin (BTX-A) injections are widely used to reduce spasticity, but their effectiveness is not well defined. We performed a systematic review of literature to answer questions about the effectiveness of BTX-A injections in the upper limb in children with CP. Methods: A systematic review of literature was conducted according to PRISMA guidelines. Eligible studies were randomized controlled trials with a high level of evidence on BTX-A upper limb injections in children. The outcomes analyzed included the study population, spasticity, quality of movement, activity limitations, quality of life, pain, appearance and side effects. Results: A total of 24 studies were included. The number of patients included was 1358 with a mean age between 3 and 11 years. Improvement after BTX-A injection compared to the control group was observed for spasticity (n=10/19 studies), bimanual activities (Assisting Hand Assessment) (n=3/7), activity limitations (n=6/11), pain (n=2/2) and appearance (n= 2/2). No study found an improvement in quality of life. Side effects were described in 16 studies and were moderate in all cases. Conclusion: This review of literature showed that BTX-A injections can improve spasticity and particularly activity limitations when reasonable objectives are established. Level of evidence: IV, systematic review.

PMID: 36754169

2. Developmental and acquired brain injury have opposite effects on finger coordination in children
Aviva Mimouni-Bloch, Sharon Shaklai, Moran Levin, Moria Ingber, Tanya Karolitsky, Sigal Grunbaum, Jason Friedman

Front Hum Neurosci. 2023 Jan 23;17:1083304. doi: 10.3389/fnhum.2023.1083304. eCollection 2023.

The ability to coordinate finger forces to dexterously perform tasks develops in children as they grow older. Following brain injury, either developmental (as in cerebral palsy-CP) or acquired (as in traumatic brain injury-TBI), this developmental trajectory will likely be impaired. In this study, we compared finger coordination in a group of children aged 4-12 with CP and TBI to a group of typically developing children using an isometric pressing task. As expected, deficits were observed in functional tests (Jebsen Taylor test of hand function, Box and Block test) for both groups, and children in both groups performed the pressing task less well than the control group. However, differing results were observed between the CP and TBI groups when using the uncontrolled manifold hypothesis to look at the synergy index. This index measures the relative amount of "good" (does not affect the outcome measure) and "bad" (does affect the outcome measure) variability, where in

this case the outcome measure is the total force produced by the fingers. While children with CP were more variable in their performance, their synergy index was not significantly different from typically developing children, suggesting the development of compensatory strategies. In contrast, the children following TBI showed performance that got worse as a function of age (i.e., the older children with TBI performed worse than the younger children with TBI). These differences between the groups may be a result of different areas of brain injury typically observed in CP and TBI, and the different amount of time that has passed since the injury.

PMID: 36755895

3. Exploration of the relationship between functional motor and communication performance levels and amount of use of the more affected upper extremity based on the caregivers' perceptions in children with hemiplegic cerebral palsy: A cross-sectional study
H Bingol, M Kerem Gunel

Rehabilitacion (Madr). 2023 Feb 3;57(4):100784. doi: 10.1016/j.rh.2023.100784. Online ahead of print.

Objective: To investigate relationships between amount of use of the more affected upper extremity and functional motor and communication performance classification systems. Material and methods: The study comprised 95 children with congenital hemiplegic cerebral palsy (CP) aged 6-15 years (52 males, 43 females; mean age 9.53, SD 3.1) and their parents/caregivers. The amount of use of the more affected upper extremity was assessed using Pediatric Motor Activity Log-Revised-How Often subscale (PMAL-R HO). Functional levels of the enrolled children were defined by the parents/caregivers using Manual Ability Classification System (MACS), Gross Motor Function Classification System-Expanded and Revised (GMFCS-E&R), and Communication Function Classification System (CFCS). Results: A strong and negative correlation was found between PMAL-R HO subscale score and MACS (r=-0.819), suggesting that children with lower MACS levels are more likely to use their more affected upper extremity spontaneously. Additionally, negative and moderate associations between PMAL-R HO subscale score and GMFCS and CFCS were revealed (r1=-0.549 and r2=-0.567). Conclusion: The amount of use of the more affected upper extremity is more sensitive to MACS than GMFCS-E&R and CFCS. Children with a given MACS level had a wide range of PMAL-R HO subscale score. In addition to MACS, a score on the PMAL-R HO subscale related to the more affected upper extremity should be included as an inclusion criterion in clinical trials to avoid misleading effects of intervention approaches aimed at improving the amount of use of the more affected upper extremity in children with congenital hemiplegic CP.

PMID: 36739683

4. A 25-Year-Old Patient With Chest Wall Erythema and Tenderness Who Was Ventilated Mechanically
Benjamin Schluger, Lisle Winston, Pallavi Juneja, Keaton C Stoner, Subani Chandra

Case Reports Chest. 2023 Feb;163(2):e57-e61. doi: 10.1016/j.chest.2022.09.002.

A 25-year-old man with cerebral palsy, scoliosis, and ventilator dependence since SARS-CoV-2 infection 11 months earlier presented with a 2-week history of chest redness and swelling. The area of erythema and edema was located on the left side of the anterior chest and had grown to approximately 9 cm in diameter over the 2 weeks. It was tender to palpation. There was no history of trauma, injury, or bug bites at that site. He had not had a rash or similar lesions elsewhere on his body and had not taken any new medications. He did have increased, thick, yellow secretions from his tracheostomy, but no fevers. He was born in the Dominican Republic and moved to the United States as a child. He had not traveled anywhere outside the United States in more than a decade.

PMID: 36759118

5. The relation of energy cost of walking with gait deviation, asymmetry, and lower limb muscle co-activation in children with cerebral palsy: a retrospective cross-sectional study
Yngvild Gagnat, Siri Merete Brændvik, Inge Ringheim, Karin Roeleveld

BMC Musculoskelet Disord. 2023 Feb 9;24(1):111. doi: 10.1186/s12891-023-06223-1.

Background: Compared to typically developing children, children with cerebral palsy (CP) have increased energy expenditure during walking, limiting activity and participation. Insight into whether the also deviating and more asymmetric gait with increased muscle co-activation contributes to this increased energy expenditure is important for clinical decision making. The aim of this study was to investigate the relation between energy cost of walking with gait deviation, asymmetry, and muscle co-activation in children with CP. Methods: Forty ambulant children with CP, with Gross Motor Function Classification System (GMFCS) level I (N = 35) and II (N = 5), aged between 5-17y, were tested at one or two occasions with 24 weeks in between, resulting in 71 observations. Gross energy cost (J/kg/m) was measured during a 5-min walk test at self-selected speed. From a 3 -dimensional gait analyses, kinematic variables and electromyography were extracted to calculate the gait deviation index (GDI) and co-activation index. The relation between energy cost and GDI, GDI asymmetry, and co-activation index of the lower limb muscles was evaluated through mixed model analyses. Height was included to control for growth-related variation. Results: Gait deviation and height combined explained about 40% of the variance in gross energy cost. No significant contribution was found for gait asymmetry or co-activation index. Conclusions: This cross-sectional study indicates that increased gait deviation contributes to increased energy cost of walking in children with GMFCS level I and II.

PMID: 36759806

6. High-intensity interval training in children and adolescents with special educational needs: a systematic review and narrative synthesis

Eric Tsz-Chun Poon, Waris Wongpipit, Fenghua Sun, Andy Choi-Yeung Tse, Cindy Hui-Ping Sit

Review Int J Behav Nutr Phys Act. 2023 Feb 9;20(1):13. doi: 10.1186/s12966-023-01421-5.

Background: High-intensity interval training (HIIT) has been promoted as a time-efficient exercise strategy to improve health and fitness in children and adolescents. However, there remains little consensus in the literature regarding its efficacy in children and adolescents with special educational needs (SEN). This study aimed to examine HIIT as a means of improving key health and fitness parameters in children and adolescents with SEN. Methods: A systematic search was conducted on eight databases (MEDLINE, Embase, SPORTDiscus, Web of Science, Scopus, PsycINFO, CINAHL, and Cochrane Library). Studies were eligible if they 1) included an HIIT protocol, 2) examined parameters related to both physical and mental aspects of health and fitness, and 3) examined children and adolescents with SEN aged 5-17 years. Results: Of the 1727 studies yielded by the database search, 13 (453 participants) were included and reviewed. We found that HIIT generally improved body composition, physical fitness, and cardiometabolic risk biomarkers across a spectrum of SEN (e.g., attention deficit hyperactivity disorder, cerebral palsy, developmental coordination disorder, and mental illness). Improvements in mental health and cognitive performance following HIIT have also been observed. Conclusion: This review provides up-to-date evidence for HIIT as a viable exercise strategy for children and adolescents with SEN. Further research investigating the benefits of HIIT in a wider range of SEN populations is warranted. Trial registration: This study was registered in the International Prospective Register of Systematic Review (PROSPERO; registration number CRD42022352696).

PMID: 36759853

7. Abnormal Activity of Masticatory Muscles in Patients with Diagnosis of Cerebral Palsy. A Systematic Review and Meta-Analysis of Observational Studies

Rafael Zaratin Beltramin, Ana Luiza Cabrera Martimbianco, Marcela Leticia Leal Gonçalves, Monise Mendes Rocha, Soraia Micaela Silva, Anna Carolina Ratto Tempestini Horliana, Elaine Marcílio Santos, Karina Helga Turcio, Raquel Agnelli Mesquita-Ferrari, Kristianne Porta Santos Fernandes, Lara Jansiski Motta, Sandra Kalil Bussadori

Phys Occup Ther Pediatr. 2023 Feb 9;1-16. doi: 10.1080/01942638.2023.2173038. Online ahead of print.

Aims: to map and synthesize the results from studies that assessed whether individuals diagnosed with cerebral palsy (CP) have abnormal masseter and temporal muscles activation during the masticatory cycle. Methods: Six databases were searched for comparative observational studies assessing masticatory muscles activation in individuals with CP through electromyography analysis. Methodological quality was evaluated using the Joanna Briggs Critical Appraisal Checklist. Outcome data were combined in meta-analysis using the Review Manager software. Results: We included five cross-sectional studies with an overall low risk of bias. Meta-analyses showed no difference between CP and healthy individuals regarding maximum

voluntary isometric contraction: right masseter (Standard mean difference [SMD] - 0.95; 95% CI -2.03 to 0.13); left masseter (SMD -0.92; 95% CI -1.93 to 0.09); right temporal (SMD -0.72; 95% CI -1.63 to 0.18); and left temporal (SMD -0.68; 95% CI -1.76 to 0.40). Electrical activity amplitude in the inactive period was superior in the CP group, and maximum bite pressure presented higher values in the control group (Mean difference [MD] - 17.38; CI 95% -26.62 to -10.15). Conclusions: Based on observational studies with a lower level of evidence, individuals with CP seem to present difficulties activating masticatory muscles. Future prospective cohort studies with rigorous methodology are still necessary to support these findings. PROSPERO register CRD42020208444.

PMID: 36760123

8. Marginal and Canonical Babbling in 10 Infants at Risk for Cerebral Palsy
Helen L Long, Katherine C Hustad

Am J Speech Lang Pathol. 2023 Feb 9;1-15. doi: 10.1044/2022_AJSLP-22-00165. Online ahead of print.

Purpose: This study is a preliminary quantification and characterization of the development of marginal and canonical syllable patterns in 10 infants at risk for cerebral palsy (CP). Method: We calculated marginal and canonical babbling ratios from parent -infant laboratory recordings of 10 infants at two time points, approximately 13 and 16 months of age. The frequency and diversity of labial, coronal, and velar types of marginal and canonical syllables were also examined. Differences across three outcome groups were compared: infants later diagnosed with CP (n = 3, CP group), risk of CP due to ongoing gross motor delays (n = 4, risk group), and current typically developing status with resolved gross motor delays (n = 3, TDx group). Performance on the Mullen Scales was included for perspective on cognitive development. Results: Higher marginal syllable ratios were observed in the CP and risk groups than the TDx group. An increasing canonical syllable ratio across the two ages was consistently observed in the TDx group. The TDx group produced a greater frequency and diversity of canonical syllable types than the risk and CP groups, and of marginal syllable types than the CP group. Conclusions: This study offers preliminary support for the possibility that speech motor impairment in infants with CP have the potential to be observed and quantified early in vocal development prior to the expected onset of first words. Prolonged rates of marginal syllable forms may be suggestive of speech motor impairment; however, additional longitudinal outcome data over a longer time course and a larger sample of infants are needed to provide further support for this possibility.

PMID: 36758205

9. Antibiotics for chronic pulmonary infection in children with a neurodisability (neurodevelopmental disorder)
Juliane Rf Sanner, Kamini Jain, Jane Williams, Matthew N Hurley

Review Cochrane Database Syst Rev. 2023 Feb 9;2(2):CD013813. doi: 10.1002/14651858.CD013813.pub2.

Background: 'Neurodisability' refers to a group of conditions that result primarily from a neurological problem (e.g. cerebral palsy), neuromuscular problem (e.g. a muscular dystrophy) or developmental problems (e.g. developmental impairment, Down syndrome). Children and young people with these conditions may have similar problems with mobility, feeding and airway clearance. Chest and breathing problems (including pulmonary infections) are commonly experienced by children and young people with neurodisabilities and are often a cause for them requiring hospital care. For those who are unable to completely clear their airway of secretions, or have frequent infections, pulmonary infections may not be able to be completely eradicated and therefore become chronic. It is unclear what treatment is best for children and young people in this position. Objectives: To assess the effectiveness and adverse effects of antibiotic treatment for chronic pulmonary infection in children and young people living with a neurodisability, including quality-of-life measures, effects on hospitalisation and healthcare contacts. Search methods: We searched the Cochrane Airways Trials Register, Cochrane Acute Respiratory Infections Group Register of Trials (CARIGRT), Cochrane Central Register of Controlled Trials (CENTRAL), MEDLINE (Ovid), Embase (Ovid), Cumulative Index to Nursing and Allied Health Literature (CINAHL), OpenGrey (www.opengrey.eu) and three trials registries up to 8 February 2022. Additionally, we identified related systematic reviews through Epistemonikos.org (8 February 2022) and searched reference lists of these. Selection criteria: All randomised controlled trials of antibiotic therapy for chronic pulmonary infection in children and young people up to the age of 18 living with a neurodisability were eligible. Data collection and analysis: Two independent review authors screened results of the searches against predetermined inclusion criteria, resolving any discrepancies by discussion. Main results: We identified a total of 1968 independent records through our searches, of which we assessed six full-text articles for eligibility. We identified one ongoing study as well as one related substudy but did not identify any completed studies eligible for inclusion in this systematic review. Authors' conclusions: The

findings of this systematic review highlight a lack of evidence in the antibiotic treatment of chronic pulmonary infection in children and young people up to the age of 18 living with a neurodisability. Further research examining this topic is therefore required.

PMID: 36757320

10. Participation patterns of children with cerebral palsy: A caregiver's perspective
Lethabo E Africa, Anri Human, Muziwakhe D Tshabalala

Afr J Disabil. 2023 Jan 31;12:1058. doi: 10.4102/ajod.v12i0.1058. eCollection 2023.

Background: Participation in activities of daily living (ADL), education, leisure and play in children living with cerebral palsy (CP) may be affected by various factors, as outlined in the International Classification of Functioning, Disability and Health Framework (ICF). The aim of this study was to describe the participation patterns of a group of these children. Objectives: This study aimed to describe participation patterns in ADL, education, leisure and play activities of children living with CP in Modimolle. Method: An exploratory-descriptive qualitative (EDQ) study design was used. A researcher-constructed bio-demographic data sheet and a semi-structured interview schedule were used to collect data from the primary caregivers of children (5-17 years) living with CP in Modimolle. Interviews were transcribed verbatim, translated from Sepedi to English and analysed using the content analysis approach and NVivo software. Results: The findings of this study indicated that children living with CP in Modimolle require set-up and assistance to participate in various ADL such as self-care, family and community activities. They also participate in formal and informal educational programmes as well as active and passive leisure and play activities. However, at the moment, they have limited opportunities to participate because of resource constraints and inaccessible infrastructure. Conclusion: Although children with CP in Modimolle perform some ADL, and participate in educational, leisure and play activities, they are not fully integrated into their community. Legislative support and policy implementation are required to improve participation and integration of children living with CP. Further studies on community-specific integrative strategies to enhance participation among children living with disabilities are recommended. Contribution: This paper provides valuable information on the participation patterns of children with CP living in a rural area of South Africa. The findings can assist with development and implementation of community-specific, integrative health and social care strategies to enhance participation among children living with disabilities.

PMID: 36756463

11. Translation and validation of Cerebral Palsy Quality of Life Questionnaire-Teen in Hong Kong Chinese population [CP QoL-Teen (HK)]
Shirley P C Ngai, L Y Wong, Vitti W K Poon, Candice Y C Poon, Beverley P H Yiu, Teresa P S Wong, C P Chow

Eur J Pediatr. 2023 Feb 9. doi: 10.1007/s00431-023-04845-0. Online ahead of print.

Cerebral palsy (CP) is an early onset, non-progressive, neuromotor disorder. Adolescence is the transition from childhood to adulthood when changes in physical and emotional aspects and self-perception occur further imposing an impact to quality of life (QoL) in individuals with CP. Cerebral Palsy Quality of Life (CP QoL) Teen is a questionnaire examining different domains of QoL for adolescents with CP. This study is aimed at translating and validating self-report and proxy-report CP QoL -Teen (HK). Prior approval of translation has been obtained. Forward and backward translations were performed following standardized translation procedures. Participants and their caregivers were asked to complete self-report and proxy-report CP QoL-Teen (HK), and Child Health Questionnaire (CHQ). Internal consistency and test-retest reliability were assessed by Cronbach's alpha and intraclass correlation coefficient (ICC), respectively. Concurrent validity was evaluated by Spearman's rank correlation between subscales of CP QoL-Teen (HK) and CHQ as well as expanded and revised version of Gross Motor Function Classification System (GMFCS-E&R). Ninety-six participants completed the study. Of these, twenty participants completed CP QoL-Teen (HK) twice. Cronbach's α of CP QoL-Teen (HK) ranged from 0.84 to 0.95 suggesting excellent internal consistency. Moderate to excellent test-retest reliability were demonstrated in all subscales of CP QoL-Teen (HK) (self -report: ICC = 0.46-0.8; proxy-report: ICC = 0.40-0.72, $p < 0.05$). Weak to moderate association between subscales of CP QoL -Teen (HK) and CHQ (self-report: rs = 0.24-0.61; proxy-report: rs = - 0.41-0.60) was reported. Conclusion: This study showed that CP QoL-Teen (HK) has good psychometric properties. It is a valid and reliable tool to assess quality of life of adolescents with CP. What is known: • Cerebral Palsy Quality of life-Teen (CP QoL-Teen) is a validated tool with strong psychometric properties and clinical utility in gauging the QoL in adolescents with CP during their transition from childhood to adulthood when changes in physical and emotional aspects and self-perception occur. Yet, a locally validated tool is lacking in measuring

the QoL for adolescents with CP in Hong Kong. What is new: • The Chinese translated version CP QoL-Teen (HK) is a valid and reliable tool to assess quality of life of adolescents with CP tailoring to the local cultural and social background with good psychometric properties being demonstrated.

PMID: 36757493

12. Correction to Validation of eligibility criteria for the Japan Obstetric Compensation System for Cerebral Palsy in preterm infants: A case-control cohort study
No authors listed

Published Erratum J Obstet Gynaecol Res. 2023 Feb;49(2):775. doi: 10.1111/jog.15581.

No abstract available

PMID: 36754629

13. Preimplantation genetic testing and child health: a national register-based study
Erica Ginström Ernstad, Charles Hanson, Kjell Wånggren, Ann Thurin-Kjellberg, Cecilia Hulthe Söderberg, Elisabeth Syk Lundberg, Max Petzold, Ulla-Britt Wennerholm, Christina Bergh

Hum Reprod. 2023 Feb 7;dead021. doi: 10.1093/humrep/dead021. Online ahead of print.

Study question: Is preimplantation genetic testing (PGT) associated with adverse perinatal outcome and early childhood health? Summary answer: Children born after PGT had comparable perinatal outcomes to children born after IVF/ICSI and comparable findings regarding early childhood health. What is known already: PGT is offered to couples affected by monogenic disorders (PGT-M) or inherited chromosomal aberrations (PGT-SR), limiting the risk of transferring the disorder to the offspring. PGT, an invasive technique, requires genetic analysis of one or up to ten cells from the embryo and is combined with IVF or ICSI. Several studies, most of them small, have shown comparable results after PGT and IVF/ICSI concerning perinatal outcome. Only a few studies with limited samples have been published on PGT and childhood health. Study design, size, duration: We performed a register-based study including all singletons born after PGT (n = 390) in Sweden during 1 January 1996-30 September 2019. Singletons born after PGT were compared with all singletons born after IVF/ICSI (n = 61 060) born during the same period of time and with a matched sample of singletons (n = 42 034) born after spontaneous conception selected from the Medical Birth Register. Perinatal outcomes, early childhood health, and maternal outcomes were compared between pregnancies after PGT and IVF/ICSI as well as between pregnancies after PGT and spontaneous conception. Primary outcomes were preterm birth (PTB) and low birthweight (LBW) whereas childhood morbidity was the secondary outcome. Participants/ materials, setting, methods: Data on women who went through PGT and gave birth were obtained from the local databases at the two PGT centres in Sweden, whereas data on IVF treatment for the IVF/ICSI group were obtained from the national IVF registers. These data were then cross-linked to national health registers; the Medical Birth Register, the Patient Register, and the Cause of Death Register. Logistic multivariable regression analysis and Cox proportional hazards models were performed with adjustment for relevant confounders. Main results and the role of chance: The mean follow-up time was 4.6 years for children born after PGT and 5.1 years for children born after spontaneous conception, whereas the mean follow-up time was 9.0 years for children born after IVF/ICSI. For perinatal outcomes, PTB occurred in 7.7% of children after PGT and in 7.3% of children after IVF/ICSI, whereas the rates were 4.9% and 5.2% for LBW (adjusted odds ratio (AOR) 1.22, 95% CI 0.82-1.81 and AOR 1.17, 95% CI 0.71-1.91, respectively). No differences were observed for birth defects. In comparison to spontaneous conception, children born after PGT had a higher risk for PTB (AOR 1.73, 95% CI 1.17-2.58). Regarding early childhood health, the absolute risk of asthma was 38/390 (9.7%) in children born after PGT and 6980/61 060 (11.4%) in children born after in IVF/ICSI, whereas the corresponding numbers were 34/390 (8.7%) and 7505/61 060 (12.3%) for allergic disorders. Following Cox proportional hazards models, no significant differences were found for these outcomes. Sepsis, hypothyroidism, attention deficit hyperactivity disorder, autism spectrum disorders, mental retardation, cerebral palsy, and epilepsy were diagnosed in a maximum of three PGT children. No PGT children died during the follow-up period. Regarding maternal outcomes, the rates of placenta praevia and caesarean delivery were significantly higher after PGT in comparison to spontaneous conception (AOR 6.46, 95% CI 3.38-12.37 and AOR 1.52, 95% CI 1.20-1.92, respectively), whereas no differences were seen comparing pregnancies after PGT and IVF/ICSI. Limitations, reasons for caution: The rather small sample size of children born after PGT made it impossible to adjust for all relevant confounders including fertilization method and culture duration. Moreover, the follow-up time was short for most of the children especially in the PGT group, probably lowering the absolute number of diagnoses in early childhood. Wider implications of the findings: The results are reassuring

and indicate that the embryo biopsy itself has no adverse effect on the perinatal, early childhood, or maternal outcomes. Although the results are comparable to IVF/ICSI also regarding early childhood outcome, they should be taken with caution due to the low number of children with diagnoses and short follow-up time. Long-term follow-up studies on children born after PGT are scarce and should be conducted considering the invasiveness of the technique. Study funding/competing interest(s): The study was financed by grants from the Swedish state under the agreement between the Swedish government and the county councils, the ALF-agreement (LUA/ALF 70940), the Board of National Specialised Medical Care at Sahlgrenska University Hospital and Hjalmar Svensson Research Foundation. There are no conflicts of interest to declare. Trial registration number: N/A.

PMID: 36749096

14. One-person versus two-person mask ventilation in preterm infants at birth: a pilot randomised controlled trial
Dharmesh Shah, Mark B Tracy, Murray Kenneth Hinder, Nadia Badawi

Randomized Controlled Trial BMJ Paediatr Open. 2023 Feb;7(1):e001768. doi: 10.1136/bmjpo-2022-001768.

Background: Mask leak and airway obstruction are common with mask ventilation in newborn infants, leading to suboptimal ventilation. We aimed to perform a pilot study measuring respiratory mechanics during one-person and two-person mask ventilation in preterm infants at birth. Methods: Infants less than 30 weeks' gestation were eligible for the study. In the two-person method, one person holds the mask in place and the other provides positive pressure ventilation compared with the standard one-person mask hold. A respiratory function monitor was used in line with a T-piece resuscitator to measure mask leak and airway obstruction. Deferred consent was obtained. Results: Twenty-five infants were recruited. The mean (SD) birth weight was 920.4 g (188.3), and mean (SD) gestational age was 27.3 weeks (3.0). Percentage mask leak was higher in the one-person mask method (26.4±18.5) compared with the two-person mask method (17.6±9.3) (p=0.018). The mean (SD) expired tidal volume (VTe, mL) in breaths with leak was 3.9 (1.57) in the one-person method compared with 3.05 (1.0) the two-person method (p=0.31). A significantly lower mean (SD) end-tidal carbon dioxide (EtCO2, mm Hg) was measured at 25.3 (9.9) in breaths with mask leak, compared with 30.8 (12.1) in breaths without leak. The breaths with airway obstruction had lower mean EtCO2 (25.9 vs 30.8, p=0.003) and lower mean VTe (1.71 vs 6.95, p<0.001). Conclusion: Mask leak and airway obstruction are common in resuscitation of preterm infants at birth. The use of the two-person mask technique is effective and it could be a useful option if mask ventilation with the one-person method is not effective. Trial registration number: ACTRN12614000245695.

PMID: 36746525

15. The Long-Term Outcome and Rehabilitative Approach of Intraventricular Hemorrhage at Preterm Birth
Juntaek Hong, Dong-Wook Rha

J Korean Neurosurg Soc. 2023 Feb 8. doi: 10.3340/jkns.2022.0262. Online ahead of print.

Technological advances in neonatology led to the improvement of the survival rate in preterm babies with very low birth weights. However, intraventricular hemorrhage (IVH) has been one of the major complications of prematurity. IVH is relevant to neurodevelopmental disorders, such as cerebral palsy, language and cognitive impairments, and neurosensory and psychiatric problems, especially when combined with brain parenchymal injuries. Additionally, severe IVH requiring shunt insertion is associated with a higher risk of adverse neurodevelopmental outcomes. Multidisciplinary and longitudinal rehabilitation should be provided for these children based on the patients' life cycles. During the infantile period, it is essential to detect high-risk infants based on neuromotor examinations and provide early intervention as soon as possible. As babies grow up, close monitoring of language and cognitive development is needed. Moreover, providing continuous rehabilitation with task-specific and intensive repetitive training could improve functional outcomes in children with mild-to-moderate disabilities. After school age, maintaining the level of physical activity and managing complications are also needed.

PMID: 36751692

16. A big data approach to evaluate receipt of optimal care in childhood cerebral palsy
Alexis Mitelpunkt, Megan A Stodola, Jilda Vargus-Adams, Brad G Kurowski, Kelly Greve, Surbhi Bhatnagar, Bruce Aronow, Janet Zahner, Amy F Bailes

Disabil Rehabil. 2023 Feb 8;1-8. doi: 10.1080/09638288.2023.2175919. Online ahead of print.

Purpose: Through automated electronic health record (EHR) data extraction and analysis, this project systematically quantified actual care delivery for children with cerebral palsy (CP) and evaluated alignment with current evidence-based recommendations. Methods: Utilizing EHR data for over 8000 children with CP, we developed an approach to define and quantify receipt of optimal care, and pursued proof-of-concept with two children with unilateral CP, Gross Motor Function Classification System (GMFCS) Level II. Optimal care was codified as a cluster of four components including physical medicine and rehabilitation (PMR) care, spasticity management, physical therapy (PT), and occupational therapy (OT). A Receipt of Care Score (ROCS) quantified the degree of adherence to recommendations and was compared with the Pediatric Outcomes Data Collection Instrument (PODCI) and Pediatric Quality of Life Inventory (PEDS QL). Results: The two children (12 year old female, 13 year old male) had nearly identical PMR and spasticity component scores while PT and OT scores were more divergent. Functional outcomes were higher for the child who had higher adjusted ROCS. Conclusions: ROCSs demonstrate variation in real-world care delivered over time and differentiate between components of care. ROCSs reflect overall function and quality of life. The ROCS methods developed are novel, robust, and scalable and will be tested in a larger sample. IMPLICATIONS FOR REHABILITATION: Optimal practice, with an emphasis on integrated multidisciplinary care, can be defined and quantified utilizing evidence-based recommendations. Receipt of optimal care for childhood cerebral palsy can be scored using existing electronic health record data. Big Data approaches can contribute to the understanding of current care and inform approaches for improved care.

PMID: 36755522

17. Brain structural and functional connectivity and network organization in cerebral palsy: A scoping review
Nina P T Jacobs, Petra J W Pouwels, Marjolein M van der Krogt, Pieter Meyns, Kangdi Zhu, Loïs Nelissen, Linda J Schoonmade, Annemieke I Buizer, Laura A van de Pol

Review Dev Med Child Neurol. 2023 Feb 7. doi: 10.1111/dmcn.15516. Online ahead of print.

Aim: To explore altered structural and functional connectivity and network organization in cerebral palsy (CP), by clinical CP subtype (unilateral spastic, bilateral spastic, dyskinetic, and ataxic CP). Method: PubMed and Embase databases were systematically searched. Extracted data included clinical characteristics, analyses, outcome measures, and results. Results: Sixty-five studies were included, of which 50 investigated structural connectivity, and 20 investigated functional connectivity using functional magnetic resonance imaging (14 studies) or electroencephalography (six studies). Five of the 50 studies of structural connectivity and one of 14 of functional connectivity investigated whole-brain network organization. Most studies included patients with unilateral spastic CP; none included ataxic CP. Interpretation: Differences in structural and functional connectivity were observed between investigated clinical CP subtypes and typically developing individuals on a wide variety of measures, including efferent, afferent, interhemispheric, and intrahemispheric connections. Directions for future research include extending knowledge in underrepresented CP subtypes and methodologies, evaluating the prognostic potential of specific connectivity and network measures in neonates, and understanding therapeutic effects on brain connectivity.

PMID: 36750309

Prevention and Cure

18. Safety and efficacy outcomes after intranasal administration of neural stem cells in cerebral palsy: a randomized phase 1/2 controlled trial
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Clinical Trial Stem Cell Res Ther. 2023 Feb 9;14(1):23. doi: 10.1186/s13287-022-03234-y.

Background: Neural stem cells (NSCs) are believed to have the most therapeutic potential for neurological disorders because they can differentiate into various neurons and glial cells. This research evaluated the safety and efficacy of intranasal administration of NSCs in children with cerebral palsy (CP). The functional brain network (FBN) analysis based on electroencephalogram (EEG) and voxel-based morphometry (VBM) analysis based on T1-weighted images were performed to evaluate functional and structural changes in the brain. Methods: A total of 25 CP patients aged 3-12 years were randomly assigned to the treatment group (n = 15), which received an intranasal infusion of NSCs loaded with nasal patches and rehabilitation therapy, or the control group (n = 10) received rehabilitation therapy only. The primary endpoints were the safety (assessed by the incidence of adverse events (AEs), laboratory and imaging examinations) and the changes in the Gross Motor Function Measure-88 (GMFM-88), the Activities of Daily Living (ADL) scale, the Sleep Disturbance Scale for Children (SDSC), and some adapted scales. The secondary endpoints were the FBN and VBM analysis. Results: There were only four AEs happened during the 24-month follow-up period. There was no significant difference in the laboratory examinations before and after treatment, and the magnetic resonance imaging showed no abnormal nasal and intracranial masses. Compared to the control group, patients in the treatment group showed apparent improvements in GMFM-88 and ADL 24 months after treatment. Compared with the baseline, the scale scores of the Fine Motor Function, Sociability, Life Adaptability, Expressive Ability, GMFM-88, and ADL increased significantly in the treatment group 24 months after treatment, while the SDSC score decreased considerably. Compared with baseline, the FBN analysis showed a substantial decrease in brain network energy, and the VBM analysis showed a significant increase in gray matter volume in the treatment group after NSCs treatment. Conclusions: Our results showed that intranasal administration of NSCs was well-tolerated and potentially beneficial in children with CP. Trial registration: The study was registered in ClinicalTrials.gov (NCT03005249, registered 29 December 2016, https://www.clinicaltrials.gov/ct2/show/NCT03005249?term=NCT03005249&draw=2&rank=1) and the Medical Research Registration Information System (CMR-20161129-1003).

PMID: 36759901

19. Acceptability of neural stem cell therapy for cerebral palsy: survey of the Australian cerebral palsy community

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Background: Neural stem cells (NSCs) have the potential to engraft and replace damaged brain tissue, repairing the damaged neonatal brain that causes cerebral palsy (CP). There are procedures that could increase engraftment of NSCs and may be critical for efficacy, but hold notable risks. Before clinical trials progress, it is important to engage with the CP community to understand their opinions. The aim of this study was to determine the acceptability of NSC therapy for CP in the CP community. Methods: Australian residents with CP and parents/carers of those with CP completed a questionnaire to determine their willingness to use NSCs from three sources (fetal, embryonic and induced pluripotent stem cells) and their willingness to undergo accompanying procedures (neurosurgery, immunosuppression) that carry potential risks. To further explore their views, participants also answered free text questions about their ethical concerns regarding the source of NSCs and their perceptions of meaningful outcomes following NSC treatment. Results: In total, 232 responses were analyzed. Participants were willing to use NSCs from all three cell sources and were willing to undergo NSC therapy despite the need for neurosurgery and immunosuppression. Participants identified a range of outcome domains considered important following NSC treatment including gross motor function, quality of life, independence and cognitive function. Conclusions: Hypothetical NSC therapy was acceptable to the Australian CP community. This study has identified important findings from the CP community which can be used to inform future NSC research, including the design of clinical trials which may help to increase recruitment, compliance and participant satisfaction.

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