Cerebral palsy research news

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Interventions and Management

1. A Scoping Review on Mobile Health Technology for Assessment and Intervention of Upper Limb Motor Function in Children with Motor Impairments

Md Raihan Mia, Sheikh Iqbal Ahamed, Alissa Fial, Samuel Nemanich

Review Games Health J. 2024 Mar 25. doi: 10.1089/g4h.2023.0224. Online ahead of print.

Upper limb (UL) motor dysfunctions impact residual movement in hands/shoulders and limit participation in play, sports, and leisure activities. Clinical and laboratory assessments of UL movement can be time-intensive, subjective, and/or require specialized equipment and may not optimally capture a child's motor abilities. The restrictions to in-person research experienced during the COVID-19 pandemic have inspired investigators to design inclusive at-home studies with child participants and their families. Relying on the ubiquity of mobile devices, mobile health (mHealth) applications offer solutions for various clinical and research problems. This scoping review article aimed to aggregate and synthesize existing research that used health technology and mHealth approaches to evaluate and assess the hand function and UL movement in children with UL motor impairment. A scoping review based on the Preferred Reporting Items for Systematic Reviews and Meta-Analyses extension for Scoping Reviews (PRISMA-ScR) model was conducted in March 2023 yielding 25 articles (0.32% of 7891 studies). Assessment characteristics included game or task-based tests (13/25, 52%), primarily for neurological disorders (e.g., autism spectrum disorder [ASD], dystonia, dysgraphia) or children with cerebral palsy (CP). Although several mHealth studies were conducted in the clinical environment (10/25, 40%), studies conducted at home or in nonclinical settings (15/25, 60%) reported acceptable and highly satisfactory to the patients as minimizing the potential risks in participation. Moreover, the remaining barriers to clinical translation included object manipulation on a touch screen, offline data analysis, real-world usability, and age-appropriate application design for the wider population. However, the results emphasize the exploration of mHealth over traditional approaches, enabling user-centered study design, family-oriented methods, and large-scale sampling in future research.

PMID: 38700552

2. Can postoperative Cobb and pelvic obliquity corrections be predicted using supine traction X-rays in non-ambulatory patients with cerebral palsy fused to L5? A case series study

Ruben Alejandro Morales Ciancio, Jonathan Lucas, Stewart Tucker, Thomas Ember, Mark Harris, Edel Broomfield

Spine Deform. 2024 Apr 29. doi: 10.1007/s43390-024-00880-5. Online ahead of print.

Purpose: This study aimed to determine whether Cobb and pelvic obliquity corrections can be predicted using supine traction radiographs in patients with cerebral palsy (CP) who underwent posterior spinal fusion (PSF) from T2/3 to L5. Methods: From January 2010 to January 2020, 167 non-ambulatory patients with CP scoliosis underwent PSF using pedicle screws in two quaternary centers with a minimum of 2 years follow-up (FU). Radiological measurements and chart reviews were performed. Results: A total of 106 patients aged 15.6 ± 0.4 years were included. All patients had significant correction of the Cobb angle (MC), pelvic obliquity (PO), thoracic kyphosis (TK), and lumbar lordosis (LL) without loss of correction at the last FU (LFU). Curve flexibility was significantly correlated with Cobb correction (δMC) immediately postoperatively (p <

0.0001, r = 0.8950), followed by the amount of correction in pelvic obliquity under traction (δPOT) (p = 0.0252, r = 0.2174). For correction in PO (δPO), the most significant variable was δPOT (p < 0.0001, r = 0.7553), followed by curve flexibility (p = 0.0059, r = 0.26) and the amount of correction in Cobb under traction (p = 0.0252, r = 0.2174). Conclusions: Cobb and PO corrections can be predicted using supine traction films for non-ambulatory CP patients treated with PSF from T2/3 to L5. The variables evaluated were interconnected, reinforcing preoperative planning for these patients. Comparative large-scale studies on patient-related clinical outcomes are required to determine whether this predicted correction is associated with improved surgical outcomes and reduced complication rates. Level of evidence: IV.

PMID: 38683285

3. Long-term reoperation rates following spinal fusion for neuromuscular scoliosis in nonambulatory patients with cerebral palsy

Christopher D Seaver, Sara J Morgan, Candice S Legister, Casey L Palmer, Eduardo C Beauchamp, Tenner J Guillaume, Walter H Truong, Steven E Koop, Joseph H Perra, John E Lonstein, Daniel J Miller

Spine Deform. 2024 Apr 29. doi: 10.1007/s43390-024-00878-z. Online ahead of print.

Purpose: To describe the incidence of reoperation and factors contributing to surgical revision within a minimum of 10 years after spinal fusion for scoliosis in patients with nonambulatory cerebral palsy (CP). Methods: We conducted a retrospective review of consecutive nonambulatory patients with CP who underwent primary spinal fusion at a single specialty care center with a minimum of 10 years from their index surgery (surgery dates 2001-2011). Causes of reoperation were classified as implant failure/pseudoarthrosis, surgical site infection (SSI), proximal junctional kyphosis, prominent/symptomatic implants, and implant removal. Reoperation rates with 95% confidence intervals were calculated for each time interval, and an actuarial survival curve was generated. Results: 144 patients met inclusion criteria (mean age = 14.3 ± 2.6 years, 62.5% male); 85.4% had 5 years follow-up data; and 66.0% had 10 years follow-up data. Estimates from the actuarial analysis suggest that 14.9% (95% CI: 10.0-22.0) underwent reoperation by 5 years postsurgery, and 21.7% (95% CI: 15.4-30.1) underwent reoperation by 10 years postsurgery. The most common causes for reoperation were implant failure/pseudoarthrosis, SSI, and prominent/symptomatic implants. Conclusions: To our knowledge, this study is the largest long-term follow-up of nonambulatory patients with CP and neuromuscular scoliosis who underwent spinal fusion. Approximately 22% of these patients required reoperation 10 years after their index surgery, primarily due to implant failure/pseudoarthrosis, SSI, and prominent/symptomatic implants. Complications and reoperations continued throughout the 10 years period after index surgery, reinforcing the need for long-term follow-up as these patients transition into adulthood. Level of evidence: III.

PMID: 38683283

4. Jump Performance and its Relationship with Lower Body Joint Kinetics and Kinematics in Children with Cerebral Palsy

Shelby E Thompson, Sydni V W Whitten, Katelyn S Campbell, Tarkeshwar Singh, Harshvardhan Singh, Li Li, Joshua Vova, Christopher M Modlesky

Med Sci Sports Exerc. 2024 Apr 30. doi: 10.1249/MSS.0000000000003421. Online ahead of print.

Purpose: The aim was to quantify jump performance in children with cerebral palsy (CP) and determine if the expected deficit is related to their lower-body-joint kinetics and kinematics. Methods: Twenty-four ambulatory (n = 17 level I and 7 level II in the Gross Motor Function Classification System) children with spastic CP (n = 13 unilateral and 11 bilateral) and 24 age-, sex-, and race-matched typically developing controls were studied. Jump height and peak power and range of motion at the hip, knee, and ankle of the more affected limb in children with CP and the nondominant limb in controls were assessed during a countermovement jump using three-dimensional motion capture and a force platform. Results: Compared to controls, children with CP had lower jump height (33%, Cohen's d (d) = 1.217), peak power at the knee (39%, d = 1.013) and ankle (46%, d = 1.687), and range of motion at the hip (32%, d = 1.180), knee (39%, d = 2.067), and ankle (46%, d = 3.195; all p < 0.001). Jump height was positively related to hip, knee, and ankle power and range of motion in children with CP (rs range = 0.474-0.613, p < 0.05), and hip and ankle power and knee and ankle range of motion in controls (rs range = 0.458-0.630, p < 0.05). The group difference in jump height was no longer detected when ankle joint power, ankle range of motion, or knee range of motion was statistically controlled (p > 0.15). Conclusions: Jump performance is compromised in children with CP and is associated with low power generation and range of motion in the lower limb, especially at the ankle.

PMID: 38686962

5. FNIRS based study of brain network characteristics in children with cerebral palsy during bilateral lower limb movement

Ping Xie, Zichao Nie, Tengyu Zhang, Gongcheng Xu, Aiping Sun, Tiandi Chen, Yan Lv

Med Phys. 2024 Apr 29. doi: 10.1002/mp.17106. Online ahead of print.

Background: Motor dysfunctions in children with cerebral palsy (CP) are caused by nonprogressive brain damage. Understanding the functional characteristics of the brain is important for rehabilitation. Purpose: This paper aimed to study the brain networks of children with CP during bilateral lower limb movement using functional near-infrared spectroscopy (fNIRS) and to explore effective fNIRS indices for reflecting functional brain activity. Methods: Using fNIRS, cerebral oxygenation signals in the bilateral prefrontal cortex (LPFC/RPFC) and motor cortex (LMC/RMC) were recorded from fifteen children with spastic CP and seventeen children with typical development (CTDs) in the resting state and during bilateral lower limb movement. Functional connectivity matrices based on phase-locking values (PLVs) were calculated using Hilbert transformation, and binary networks were constructed at different sparsity levels. Network metrics such as the clustering coefficient, global efficiency, local efficiency, and transitivity were calculated. Furthermore, the time-varying curves of network metrics during movement were obtained by dividing the time window and using sparse inverse covariance matrices. Finally, conditional Granger causality (GC) was used to explore the causal relationships between different brain regions. Results: Compared to CTDs, the connectivity between RMC-RPFC ($p = 0.017$) and RMC-LMC ($p = 0.002$) in the brain network was decreased in children with CP, and the clustering coefficient ($p = 0.003$), global efficiency ($p = 0.034$), local efficiency ($p = 0.015$), and transitivity ($p = 0.009$) were significantly lower. The standard deviation of the changes in global efficiency of children with CP during motion was also greater than that of CTDs. Using GC, it was found that there was a significant increase in causal strength from the RMC to the RPFC ($p = 0.04$) and from the RMC to the LMC ($p = 0.042$) in children with CP during motion. Additionally, there were significant negative correlations between the PLV of LMC-RMC ($p = 0.002$) and the Gross Motor Function Classification System (GMFCS) and between the GMFCS and the clustering coefficient ($p = 0.01$). Conclusions: During rehabilitation training of the lower limbs, there were significant differences in brain network indices between children with CP and CTDs. The indicators proposed in this paper are effective at evaluating motor function and the real-time impact of rehabilitation training on the brain network and have great potential for application in guiding clinical motor function assessment and planning rehabilitation strategies.

PMID: 38683184

6. Corrective Efficacy of Calcaneal Lengthening Osteotomy for Planovalgus Deformity in Cerebral Palsy Patients

Vinod Dubey, Sohilkhan R Pathan, Dhruv Sharma

Cureus. 2024 Mar 27;16(3):e57092. doi: 10.7759/cureus.57092. eCollection 2024 Mar.

Introduction Planovalgus deformity is common in children with spastic cerebral palsy (CP), particularly spastic diplegia and spastic quadriplegia. It results from muscle imbalance over the immature foot skeleton, leading to hindfoot valgus, forefoot abduction, and joint subluxation. Surgical interventions, like calcaneal lengthening osteotomy (CLO), are frequently employed to correct this deformity, but objective guidelines for its use in CP patients are lacking. Material and methods This retrospective cohort study examined the efficacy of CLO in correcting plano valgus deformity in pediatric CP patients at the Pediatric Orthopedic Unit of Christian Medical College (CMC) in Vellore, India. Data from patient records and radiographs were collected, including demographics, pre- and postoperative angles, and surgical details. Statistical analysis was performed to assess changes in angles and associations with various factors. Results After the surgery, there was a notable enhancement in the calcaneal pitch, lateral talo-first metatarsal angle, and naviculocuboid overlap, as shown by the CLO results. However, tibiocalcaneal angles did not show significant changes. Associations were observed between age, Gross Motor Function Classification System (GMFCS) level, additional surgeries, and postoperative angle corrections. Conclusion CLO shows promise in correcting plano valgus deformity, with age, GMFCS level, and comorbidities influencing outcomes. Long-term follow-up is crucial to monitor correction durability. Specific radiographic angles provide insights into CLO's biomechanical effects, but study limitations warrant caution in interpretation. CLO effectively corrects plano valgus deformity in pediatric CP patients, with age, GMFCS level, and comorbidities influencing outcomes. Long-term follow-up and further research are needed to optimize management strategies and enhance understanding of surgical outcomes.

PMID: 38681348

7. Intrathecal baclofen efficacy for managing motor function and spasticity severity in patients with cerebral palsy: a systematic review and meta-analysis

Mahdi Masrour, Amir Zare, Ana Presedo, Mohammad Hossein Nabian

Meta-Analysis BMC Neurol. 2024 Apr 27;24(1):143. doi: 10.1186/s12883-024-03647-7.

Background: Spasticity can significantly affect a patient's quality of life, caregiver satisfaction, and the financial burden on the healthcare system. Baclofen is one of only a few options for treating spasticity. The purpose of this study is to investigate the impact of intrathecal baclofen (ITB) therapy on severe40.23 spasticity and motor function in patients with cerebral palsy. Methods: We conducted a systematic review in PubMed, Scopus, Ovid, and the Cochrane Library in accordance with the PRISMA guidelines. We included studies based on eligibility criteria that included desired participants (cerebral palsy patients with spasticity), interventions (intrathecal baclofen), and outcomes (the Ashworth scales and the Gross Motor Function

Measure [GMFM]). The within-group Cohen's d standardized mean differences (SMD) were analyzed using the random effect model. Results: We screened 768 papers and included 19 in the severity of spasticity section and 6 in the motor function section. The pre-intervention average spasticity score (SD) was 3.2 (0.78), and the post-intervention average score (SD) was 1.9 (0.72), showing a 40.25% reduction. The SMD for spasticity reduction was - 1.7000 (95% CI [-2.1546; -1.2454], p-value < 0.0001), involving 343 patients with a weighted average age of 15.78 years and a weighted average baclofen dose of 289 µg/day. The SMD for the MAS and Ashworth Scale subgroups were - 1.7845 (95% CI [-2.8704; -0.6986]) and - 1.4837 (95% CI [-1.8585; -1.1088]), respectively. We found no relationship between the participants' mean age, baclofen dose, measurement time, and the results. The pre-intervention average GMFM (SD) was 40.03 (26.01), and the post-intervention average score (SD) was 43.88 (26.18), showing a 9.62% increase. The SMD for motor function using GMFM was 0.1503 (95% CI [0.0784; 0.2223], p-value = 0.0030), involving 117 patients with a weighted average age of 13.63 and a weighted average baclofen dose of 203 µg/day. In 501 ITB implantations, 203 medical complications were reported, including six new-onset seizures (2.96% of medical complications), seven increased seizure frequency (3.45%), 33 infections (16.26%), eight meningitis (3.94%), and 16 cerebrospinal fluid leaks (7.88%). Delivery system complications, including 75 catheter and pump complications, were also reported. Conclusion: Despite the risk of complications, ITB has a significant impact on the reduction of spasticity. A small but statistically significant improvement in motor function was also noted in a group of patients.

PMID: 38678195

8. Early detection of cerebral palsy with the Standardized Infant NeuroDevelopmental Assessment versus the General Movements Assessment: Is another assessment the way forward?

Mary Lauren Neel

Dev Med Child Neurol. 2024 May 1. doi: 10.1111/dmcn.15944. Online ahead of print.

No abstract available

PMID: 38693661

9. Training effect of repeated rolling motions on boccia players with severe cerebral palsy: comparison with the effect of conventional upper-limb training using a crossover test

Kosuke Yahagi, Masataka Kataoka, Tomomi Ichiba, Shuji Imura

J Phys Ther Sci. 2024 May;36(5):245-251. doi: 10.1589/jpts.36.245. Epub 2024 May 1.

The present study aimed to establish evidence regarding comparisons between training methods for boccia players with severe cerebral palsy. [Participants and Methods] Ten boccia players with severe cerebral palsy were enrolled in this study. This intervention had a cross-over design. Participants were divided into two groups based on the training method applied. The study period was divided into an upper extremity training period and an interval rolling training period, which lasted 4 weeks each. Each was separated by a 4-week training detraining period. Joint range of motion and throwing distance were measured before and after the interventions and the effects were compared and verified. [Results] Compared to upper extremity training, interval rolling training significantly expanded the range of motion and improved throwing distance in bocce players with severe cerebral palsy. [Conclusion] Collectively, interval rolling training is more effective than upper limb training in improving the physical function and throwing distance of boccia players with severe cerebral palsy.

PMID: 38694004

10. Motor Effects of Intervention With Transcranial Direct Current Stimulation for Physiotherapy Treatment in Children With Cerebral Palsy: Protocol for a Randomized Clinical Trial

Anna Izabel Cangussu, Beatriz Lucarini, Igor de Freitas Melo, Paula Araújo Diniz, Marisa Mancini, Bernardo de Mattos Viana, Marco Aurélio Romano-Silva, Débora Marques de Miranda

Randomized Controlled Trial JMIR Res Protoc. 2024 Apr 30:13:e52922. doi: 10.2196/52922.

Background: Children diagnosed with cerebral palsy (CP) often experience various limitations, particularly in gross motor function and activities of daily living. Transcranial direct current stimulation (tDCS) is a noninvasive brain stimulation technique that has been used to improve movement, gross motor function, and activities of daily living. Objective: This study aims to evaluate the potential additional effects of physiotherapy combined with tDCS in children with CP in comparison with physiotherapy only. Methods: This is a 2-arm randomized controlled trial that will compare the effects of tDCS as an adjunctive treatment during rehabilitation sessions to rehabilitation without tDCS. Children with CP classified by the Gross Motor Function Classification System as levels I and II will be randomly assigned to either the sham + rehabilitation group or the tDCS + rehabilitation group. The primary outcome will be the motor skills assessed using the Gross Motor Function

Measure domain E scores, and the secondary outcome will be the measurement scores of the children's quality of life. The intervention will consist of a 10-day stimulation protocol with tDCS spread over 2 weeks, with stimulation or sham tDCS administered for 20 minutes at a frequency of 1 Hz, in combination with physiotherapy. Physical therapy exercises will be conducted in a circuit based on each child's baseline Gross Motor Function Measure results. The participants' changes will be evaluated and compared in both groups. Intervenient features will be tested. Results: Data collection is ongoing and is expected to be completed by January 2025. A homogeneous sample and clear outcomes may be a highlight of this protocol, which may allow us to understand the potential use of tDCS and for whom it should or should not be used. Conclusions: A study with good evidence and clear outcomes in children with CP might open an avenue for the potential best use of neurostimulation. Trial registration: Brazilian Registry of Clinical Trials RBR-104h4s4y; https://tinyurl.com/47r3x2e4.

PMID: 38687586

11. Health-related quality of life and associated factors among primary caregivers of children with cerebral palsy, in Bahir Dar and Gondar cities, Ethiopia, 2022

Tesfa Kassa, Hiruy Tadese, Getachew Azeze Eriku, Yohannes Abich, Molla Fentanew

PLoS One. 2024 Apr 30;19(4):e0301050. doi: 10.1371/journal.pone.0301050. eCollection 2024.

Background: Caring for a child with cerebral palsy (CP) could negatively affect the Health-Related Quality of Life (HRQOL) of the mothers who are usually the primary caregivers. To the best of our 'knowledge, there is a dearth of information on the HRQOL of primary caregivers of children with CP in Ethiopia. Therefore, this study aimed to investigate caregivers' HRQOL and factors associated with it in Gondar and Bahir Dar Cities, Northwest Ethiopia, 2022. Methods: A community-based cross-sectional study was conducted among primary caregivers of Children with CP from April 20 to June 20, 2022, in Gondar and Bahir Dar cities. Convenience sampling was used to get study participants. Data were collected by trained health extension and community-based rehabilitation workers. The collected data were coded, cleaned, entered into EPI data, and exported to Stata-16 for analysis. A generalized linear model was employed to show the relationship between dependent and independent variables. A P-value≤0.05 was considered statistically significant at a 95% confidence interval. Result: In this study, HRQOL among primary caregivers of children with CP was 28.72(±13.38) and 23.26(±12.37) in the physical summary score (PSC) and mental summary score (MSC) respectively. Age 17-30yeas (p-value = 0.03), unable to read and write (p = 0.01), privately employed (p = 0.01) and government employed (p = 0.02), monthly income<1000 Ethiopian Birr (ETB) (p = 0.01), insufficient sleeping (p = 0.001), others relationship(p = 0.001), have three and above children (p = 0.001), others house composition (p = 0.003), have no helpers (p = 0.001) and third birth order of child (p = 0.03) were all factors associated with HRQOL in PSC. On the other hand, income<1000ETB (p = 0.05), insufficient sleeping (p = 0.001), others in relation to the child (p = 0.001), others in house composition (p = 0.03), dyskinetic CP (p = 0.01) and ataxic CP (p = 0.001) were all factors associated with HRQOL of caregivers in MSC components. Conclusion: The HRQOL among primary caregivers of children with CP in Bahir Dar and Gondar cities was low. Age, monthly income, educational status, sleeping status, relationship and house composition, number of living children, birth order of child, helpers, and type of CP were all significantly associated with HRQOL of primary caregivers of children with CP.

PMID: 38687770

12. Diagnosis and therapies for patients with cerebral palsy over the past 30 years: a bibliometric analysis

Lili Jiang, Weifang Yang, Huai Chen, Huangcheng Song, Song Zhang

Front Neurol. 2024 Apr 17:15:1354311. doi: 10.3389/fneur.2024.1354311. eCollection 2024.

Background: Currently, the incidence of cerebral palsy is high in newborns. However, the current methods for diagnosing and treating patients with cerebral palsy are complex and poorly targeted. Moreover, these studies lack the support of bibliometric analysis results. Objective: Our study focused on a bibliometric analysis of published papers on the diagnosis and treatment of patients with cerebral palsy. This study identified the primary authors, institutions, and countries involved in analyzing the status and trends of research on the diagnosis and treatment of patients with cerebral palsy. Additionally, the study also involved screening pathways related to cerebral palsy. Methods: The PubMed database was searched for publications on the diagnosis and treatment of patients with cerebral palsy between 1990 and 2023. R v4.2.2 and VOSviewer v1.6.18 software tools were utilized to perform bibliometric analysis and visualization. Results: There were 1,965 publications on cerebral palsy diagnosis and 5,418 articles on the qualified treatment strategies, and the annual number of publications also increased. The United States dominated in this field of research. Gregory Y.H. Lip and Patrizio Lancellotti published the most number of papers. The Cleveland Clinic published the most number of papers in the field. According to the analysis of the co-occurrence of keywords, we found that the main research directions were age, sex, disease diagnosis, and treatment. Newly emerging research has focused mainly on heart failure, which is related to valvular heart disease. Conclusion: The findings presented in this study offer valuable insights into ongoing research and potential future directions pertaining to cerebral palsy. These insights can assist researchers in identifying suitable collaborators and enhancing their investigations aimed at identifying the underlying molecular mechanisms associated with cerebral palsy, encompassing its etiology, preventive measures, and therapeutic interventions.

PMID: 38694779

13. Comparing Self-Report Fatigue Assessment Tools for Adults with Cerebral Palsy in a Danish Context

Ro Julia Robotham, Michelle Barner Bærentzen, Frederik Lehman Dornonville de la Cour

Dev Neurorehabil. 2024 May 2:1-10. doi: 10.1080/17518423.2024.2347989. Online ahead of print.

Purpose: Many youth and adults with Cerebral Palsy (CP) experience high levels of fatigue. This study aimed to compare three fatigue self-report questionnaires to guide clinicians. Method: Thirty youth and adults (age range 17-64) with CP were assessed with Danish versions of the Fatigue Impact and Severity Self-Assessment questionnaire, the Modified Mental Fatigue Scale, and the Multidimensional Fatigue Inventory. Psychometric properties were investigated. Rank order and classification models were compared across questionnaires. Results: The Reduced Motivation and Physical Fatigue subscales of the Multidimensional Fatigue Inventory showed inadequate internal consistency. Participants were frequently ranked differently with the questionnaires. There were issues related to the conceptualization of physical fatigue. Conclusion: The choice of assessment tool should be based on assessment purpose as the questionnaires assess different aspects of fatigue severity, impact, and management. Also, test selection can have important implications on the conclusions that are made about fatigue type and severity.

PMID: 38695209

14. Social capital of families of children with neurodevelopmental disabilities in South India

Diana Banaru, Dana Boyd, Melanie Halevy, Angel Oliver, Kayla Orsat-Parker, Marie Brien, Dinesh Krishna, Franzina Coutinho, Tatiana Ogourtsova

Dev Med Child Neurol. 2024 May 2. doi: 10.1111/dmcn.15949. Online ahead of print.

Aim: To investigate the social capital of families with children with neurodevelopmental disabilities in South India receiving a community-based early intervention (Enabling Inclusion®) program and to explore determinants and associations between social capital and program duration, socio-demographic factors, family empowerment, and caregiver burden. Method: Using purposive sampling in a cross-sectional study design, 217 families (n = 71 received short Enabling Inclusion [<5 months]; n = 146 received long Enabling Inclusion [>9 months]) were recruited and completed the Short Adapted Social Capital Tool (SASCAT: cognitive, structural), measures of family empowerment, and caregiver strain. Descriptive statistics, regression, and correlations were used for analyses. Results: In 52.1% of participants, low cognitive and structural social capital was observed. Higher odds of low structural social capital were observed for mothers with primary versus secondary education (adjusted odds ratio [OR] = 0.35; 95% confidence interval [CI] 0.13-0.90; p = 0.029); and caregivers of children with cerebral palsy versus autism (OR = 4.66; 95% CI 1.02-21.21; p = 0.046). Significant associations were found between structural social capital, the child's age, and support group membership ($\chi 2$ = 6.29; 4.70; degrees of freedom [df] = 2; 1; p = 0.04; p = 0.02 respectively), as well as between cognitive social capital and other disability in the family ($\chi 2$ = 4.62, df = 1, p = 0.03). Interpretation: While program duration was not found to mediate social capital, mother's education and child's diagnosis emerged as key influential factors, warranting their consideration in interventions supporting families of children with neurodevelopmental disabilities in low- and-middle-income countries and elsewhere.

PMID: 38698550

15. Cognitive Functioning and Assessment in Adults with Cerebral Palsy: A Scoping Review

Katrine Sand, Randi Starrfelt, Ro J Robotham

Review Dev Neurorehabil. 2024 May 4:1-11. doi: 10.1080/17518423.2024.2347991. Online ahead of print.

In this scoping review, we summarize the current knowledge of cognitive functioning in adults with cerebral palsy (CP), and identify the neuropsychological tests typically used in this population. 39 studies from the period January 1990 - August 2023 were included in the review, and they differ widely in their aims and approach to studying cognition. Very few studies have cognitive assessment as their core aim and use a neuropsychological test battery. The included studies show great variability in reported intelligence and cognitive functioning in adults with CP, and cognitive deficits have been reported in all cognitive domains. Most of the studies suffer from methodological limitations, and there is ample room for improvement within the field. We conclude by suggesting a number of recommendations that may contribute to increasing our understanding of cognitive impairments in adults with CP.

PMID: 38702978

16. Emerging ferroptosis inhibitors as a novel therapeutic strategy for the treatment of neonatal hypoxic-ischemic encephalopathy

Liang Huo, Jianhua Fu, Shimeng Wang, Hua Wang, Xueyan Liu

Review Eur J Med Chem. 2024 Apr 26:271:116453. doi: 10.1016/j.ejmech.2024.116453. Online ahead of print.

Neonatal hypoxia-ischemia encephalopathy (NHIE), an oxygen deprivation-mediated brain injury due to birth asphyxia or reduced cerebral blood perfusion, often leads to lifelong sequelae, including seizures, cerebral palsy, and mental retardation. NHIE poses a significant health challenge, as one of the leading causes of neonatal morbidity and mortality globally. Despite this, available therapies are limited. Numerous studies have recently demonstrated that ferroptosis, an iron-dependent non-apoptotic regulated form of cell death characterized by lipid peroxidation (LPO) and iron dyshomeostasis, plays a role in the genesis of NHIE. Moreover, recently discovered compounds have been shown to exert potential therapeutic effects on NHIE by inhibiting ferroptosis. This comprehensive review summarizes the fundamental mechanisms of ferroptosis contributing to NHIE. We focus on various emerging therapeutic compounds exhibiting characteristics of ferroptosis inhibition and delineate their pharmacological benefits for the treatment of NHIE. This review suggests that pharmacological inhibition of ferroptosis may be a potential therapeutic strategy for NHIE.

PMID: 38701713

17. Development of algorithms for estimating the Child Health Utility 9D from Caregiver Priorities and Child Health Index of Life with Disability

Utsana Tonmukayakul, Kate Willoughby, Cathrine Mihalopoulos, Dinah Reddihough, Brendan Mulhern, Rob Carter, Suzanne Robinson, Gang Chen

Qual Life Res. 2024 May 3. doi: 10.1007/s11136-024-03661-9. Online ahead of print.

Purpose: The primary aim was to determine Child Health Utility 9D (CHU9D) utilities from the Caregiver Priorities and Child Health Index of Life with Disabilities (CPCHILD) for non-ambulatory children with cerebral palsy (CP). Methods: One hundred and eight surveys completed by Australian parents/caregivers of children with CP were analysed. Spearman's coefficients were used to investigate the correlations between the two instruments. Ordinary least square, robust MM-estimator, and generalised linear models (GLM) with four combinations of families and links were developed to estimate CHU9D utilities from either the CPCHILD total score or CPCHILD domains scores. Internal validation was performed using 5-fold cross-validation and random sampling validation. The best performing algorithms were identified based on mean absolute error (MAE), concordance correlation coefficient (CCC), and the difference between predicted and observed means of CHU9D. Results: Moderate correlations (ρ 0.4-0.6) were observed between domains of the CHU9D and CPCHILD instruments. The best performing algorithm when considering the CPCHILD total score was a generalised linear regression (GLM) Gamma family and logit link (MAE = 0.156, CCC = 0.508). Additionally, the GLM Gamma family logit link using CPCHILD comfort and emotion, quality of life, and health domain scores also performed well (MAE = 0.152, CCC = 0.552). Conclusion: This study established algorithms for estimating CHU9D utilities from CPCHILD scores for non-ambulatory children with CP. The determined algorithms can be valuable for estimating quality-adjusted life years for cost-utility analysis when only the CPCHILD instrument is available. However, further studies with larger sample sizes and external validation are recommended to validate these findings.

PMID: 38700756

18. Reflections on Participation at Home, As Self-Reported by Young People with Cerebral Palsy

Jacinta R Quartermaine, Tanya A Rose, Megan L Auld, Leanne M Johnston

Dev Neurorehabil. 2024 May 2:1-14. doi: 10.1080/17518423.2024.2347993. Online ahead of print.

This study explored the home-based participation of young people with cerebral palsy (CP) and described factors that make participation easier or harder. Fifteen young people with CP aged 15 to 26 years provided written reflections, photographs, or videos about their home-based participation experiences. Data were analyzed using reflexive thematic analysis. Self-reported reflections were grouped inductively into 129 codes, then 20 subthemes and 5 themes which emphasized CP characteristics, thoughts, emotions, equipment, environment, supports, and inclusion as important factors influencing home-based participation. Young people with CP largely described the home environment as an inclusive place to participate.

PMID: 38695307

19. Exome sequencing reveals genetic heterogeneity and clinically actionable findings in children with cerebral palsy

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Cerebral palsy (CP) is the most common motor disability in children. To ascertain the role of major genetic variants in the etiology of CP, we conducted exome sequencing on a large-scale cohort with clinical manifestations of CP. The study cohort comprised 505 girls and 1,073 boys. Utilizing the current gold standard in genetic diagnostics, 387 of these 1,578 children (24.5%) received genetic diagnoses. We identified 412 pathogenic and likely pathogenic (P/LP) variants across 219 genes associated with neurodevelopmental disorders, and 59 P/LP copy number variants. The genetic diagnostic rate of children with CP labeled at birth with perinatal asphyxia was higher than the rate in children without asphyxia (P = 0.0033). Also, 33 children with CP manifestations (8.5%, 33 of 387) had findings that were clinically actionable. These results highlight the need for early genetic testing in children with CP, especially those with risk factors like perinatal asphyxia, to enable evidence-based medical decision-making.

PMID: 38693247

20. Novel Therapeutic Strategies of Non-Invasive Brain Stimulation and Nanomedicine in Pediatric Cerebral Palsy Patients

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Infantile central palsy (CP) is caused due to damage to the immature developing brain usually before birth, leading to altered topography and biochemical milieu. CP is a life-limiting disorder, which causes changes in sensory, motor, cognitive, and behavioral functioning. Understanding its pathophysiology is complex, and current therapeutic modalities, oral medication, surgical treatment, physical therapy, and rehabilitation provide minimal relief. As the brain is plastic, it has an inherent capacity to adapt to altered activity; thus, non-invasive brain stimulation (NIBS) strategies, like repetitive transcranial magnetic stimulation, which can modulate the neuronal activity and its function, may lead to recovery in CP patients. Further, in recent years, nanomedicine has shown a promising approach in pre-clinical studies for the treatment of central nervous system disorder because it can cross the blood-brain barrier, improve penetration, and provide sustained release of the drug. The review focuses on the principles and mechanisms of various NIBS techniques used in CP. We have also contemplated the effect of rehabilitation and nanomedicine in CP children, which will definitely lead to advancing our diagnostic as well as therapeutic abilities, in a vulnerable group of little ones.

PMID: 38691468

21. Protocol for combined N-of-1 trials to assess cerebellar neurostimulation for movement disorders in children and young adults with dyskinetic cerebral palsy

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Background: Movement and tone disorders in children and young adults with cerebral palsy are a great source of disability. Deep brain stimulation (DBS) of basal ganglia targets has a major role in the treatment of isolated dystonias, but its efficacy in dyskinetic cerebral palsy (DCP) is lower, due to structural basal ganglia and thalamic damage and lack of improvement of comorbid choreoathetosis and spasticity. The cerebellum is an attractive target for DBS in DCP since it is frequently spared from hypoxic ischemic damage, it has a significant role in dystonia network models, and small studies have shown promise of dentate stimulation in improving CP-related movement and tone disorders. Methods: Ten children and young adults with DCP and disabling movement disorders with or without spasticity will undergo bilateral DBS in the dorsal dentate nucleus, with the most distal contact ending in the superior cerebellar peduncle. We will implant Medtronic Percept, a bidirectional neurostimulator that can sense and store brain activity and deliver DBS therapy. The efficacy of cerebellar DBS in improving quality of life and motor outcomes will be tested by a series of N-of-1 clinical trials. Each N-of-1 trial will consist of three blocks, each consisting of one month of effective stimulation and one month of sham stimulation in a random order with weekly motor and quality of life scales as primary and secondary outcomes. In addition, we will characterize abnormal patterns of cerebellar oscillatory activity measured by local field potentials from the intracranial electrodes related to clinical assessments and wearable monitors. Pre- and 12-month postoperative volumetric structural and functional MRI and diffusion tensor imaging will be used to identify candidate imaging markers of baseline disease severity and response to DBS. Discussion: Our goal is to test a cerebellar neuromodulation therapy that produces meaningful changes in function and well-

being for people with CP, obtain a mechanistic understanding of the underlying brain network disorder, and identify physiological and imaging-based predictors of outcomes useful in planning further studies. Trial registration: ClinicalTrials.gov NCT06122675, first registered November 7, 2023.

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