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Cerebral palsy research news

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Interventions and Management

1. Mortality after spinal fusion in children with cerebral palsy and cerebral-palsy-like conditions: A 30-year follow-up study
Arun Hariharan, Julieanne P Sees, Carlos Pargas, Kenneth J Rogers, Tim Niiler, Michael Wade Shrader, Freeman Miller

Dev Med Child Neurol. 2023 Mar 7. doi: 10.1111/dmcn.15568. Online ahead of print.

Aim: To report survival probability of a large cohort of children with cerebral palsy (CP) after spinal fusion. Method: All children with CP who had spinal fusion between 1988 and 2018 at the reporting facility were reviewed for survival. Death records of the institutional CP database, institutional electronic medical records, publicly available obituaries, and the National Death Index through the US Centers for Disease Control were searched. Survival probabilities with different surgical eras, comorbidities, ages, and curve severities were compared using Kaplan-Meier curves. Results: A total of 787 children (402 females, 385 males) had spinal fusion at a mean age of 14 years 1 month (standard deviation 3 years 2 months). The 30-year estimated survival was approximately 30%. Survival decreased for children who had spinal fusion at younger ages, longer postoperative hospital stays, longer postoperative intensive care unit stays, gastrostomy tubes, and pulmonary comorbidities. Interpretation: Children with CP who required spinal fusions had reduced long-term survival compared with an age-matched typically developing cohort; however, a substantial number survived 20 to 30 years after the surgery. This study had no comparison group of children with CP scoliosis; therefore, we do not know whether correction of scoliosis affected their survival.

PMID: 36882978

2. Correlation between Selective Motor Control of the Lower Extremities and Balance in Spastic Hemiplegic Cerebral Palsy: a randomized controlled trial
Amira H Mohammed, Hager R El-Serougy, Amel E Abdel Karim, Mohamad Sakr, Samah M Sheha

BMC Sports Sci Med Rehabil. 2023 Mar 5;15(1):24. doi: 10.1186/s13102-023-00636-0.

Background: Children with cerebral palsy (CP) have motor deficits caused by spasticity, weakness, contractures, diminished selective motor control (SMC), and poor balance. The purpose of the current study was to evaluate the influence of mirror feedback on lower extremity selective motor control and balance in children with hemiplegic cerebral palsy. Understanding the relationship between SMC and balance will help children with hemiplegic CP receive more appropriate therapies. Methods: Forty-seven children of both sexes diagnosed with hemiplegic CP participated in the study. Group1 (Gr1 - control group) received conventional physical therapy training while group 2 (Gr2 - intervention group) received conventional physical therapy training in addition to bilateral lower extremity mirror therapy (MT). The primary outcome measure used was Selective Control Assessment of Lower Extremity scale (SCALE), while the secondary outcome measure was the

Pediatric Balance Scale (PBS). Results: There were significant differences in Selective Control Assessment of Lower Extremity Scale (SCALE) and Pediatric Balance Scale (PBS) between both groups in favor of Gr2. After treatment, both groups improved significantly, yet Gr2 outperformed Gr1 by a large margin. Conclusion: Mirror therapy may be a useful addition to home-based motor interventions for children with hemiplegic CP due to its relative simplicity, low cost, and high patient adherence. Additionally, it may help children improve their selective motor skills and balance. Trial registration: Current Controlled Trials using African Clinical Trials Registry website with ID number PACTR202105604636415 retrospectively registered on 21/01/202.

PMID: 36872326

3. The effect of whole-body vibration on lower extremity function in children with cerebral palsy: A meta-analysis
Xiaoye Cai, Guoping Qian, Siyuan Cai, Feng Wang, Yingjuan Da, Zbigniew Ossowski

PLoS One. 2023 Mar 10;18(3):e0282604. doi: 10.1371/journal.pone.0282604. eCollection 2023.

Objective: The aim of this meta-analysis was to evaluate the effect of whole-body vibration training on lower limb motor function in children with cerebral palsy in randomized-controlled trials (RCTs). Methods: Two independent reviewers systematically searched the records of nine databases (PubMed, Cochrane, Web of Science, EMBASE, CNKI, etc.) from inception to December 2022. Tools from the Cochrane Collaboration were used to assess risk of bias. Standard meta-analyses were performed using Stata 16.0 and Revman 5.3. For continuous variables, the arms difference was calculated as the weighted mean difference (WMD) between the values before and after the intervention and its 95% confidence interval (95% CI). Results: Of the 472 studies identified, 13 (total sample size 451 participants) met the inclusion criteria. Meta-analysis showed that WBV training could effectively improve GMFM88-D [WMD = 2.46, 95% CI (1.26, 3.67), P<0.01] and GMFM88-E [WMD = 3.44, 95% CI (1.21, 5.68), P = 0.003], TUG [WMD = -3.17, 95% CI (-5.11, -1.24), P = 0.001], BBS [WMD = 4.00,95% CI (3.29, 4.71), P<0. 01] and the range of motion of ankle joint and the angle of ankle joint during muscle reaction in children with cerebral palsy. The effect of WBV training on 6MWT walking speed [WMD = 47.64, 95% CI (-25.57, 120.85), p = 0.20] in children with cerebral palsy was not significantly improved. Conclusion: WBV training is more effective than other types of conventional physical therapy in improving the lower limb motor function of children with cerebral palsy. The results of this meta-analysis strengthen the evidence of previous individual studies, which can be applied to the clinical practice and decision-making of WBV training and rehabilitation in children with cerebral palsy.

PMID: 36897858

4. Medial gastrocnemius muscle properties of children with cerebral palsy after different tone treatments - a pilot study
Christiana J Raymond-Pope, Daniel B Hoffman, Rachael M Bloxsom, Sarah M Greising, Tom F Novacheck, Elizabeth R Boyer

Am J Phys Med Rehabil. 2023 Mar 8. doi: 10.1097/PHM.0000000000002235. Online ahead of print.

Objective: Spasticity in children with cerebral palsy (CP) can be managed by a spectrum of approaches, from conservative therapy to temporary botulinum toxin A (BoNT-A) injections, to permanent transection of sensory nerves with a selective dorsal rhizotomy (SDR). This pilot study investigated whether these three tone management approaches are associated with histological and biochemical properties of the medial gastrocnemius. Design: A convenience sample of children with CP undergoing gastrocnemius lengthening surgery were enrolled. Intraoperative biopsies were obtained from three individuals (1 each: minimal tone treatment; frequent gastrocnemius BoNT-A injections; prior SDR). All individuals had plantarflexor contractures, weakness, and impaired motor control prior to the biopsy. Results: Differences between participants were observed for muscle fiber cross-sectional area, fiber type, lipid content, satellite cell density, and centrally located nuclei. The most pronounced difference was the abundance of centrally located nuclei in the BoNT-A participant (52%) compared to the others (3-5%). Capillary density, collagen area and content, and muscle protein content were similar across participants. Conclusions: Several muscle properties appeared to deviate from reported norms, though age- and muscle-specific references are sparse. Prospective studies are necessary to distinguish cause and effect and to refine the risks and benefits of these treatment options.

PMID: 36897794

5. Efficacy of aerobic exercise on the functioning and quality of life of children and adolescents with cerebral palsy: A systematic review and meta-analysis
Esther G Soares, Cláudio H V Gusmão, Deisiane O Souto

Review Dev Med Child Neurol. 2023 Mar 9. doi: 10.1111/dmcn.15570. Online ahead of print.

Aim: To investigate the efficacy of aerobic exercise on the functioning (participation, activities, and body functions and structures) and quality of life (QoL) of children and adolescents with cerebral palsy (CP). Method: A systematic review with meta-analysis was performed. A comprehensive search of articles was performed using the electronic databases Embase, PubMed, PEDro, and CINAHL. Methodological quality and certainty of evidence were evaluated with the PEDro and Grading of Recommendations Assessment, Development, and Evaluation (GRADE) scales respectively. The effects of aerobic exercise on functioning were assessed using meta-analytic techniques. However, given the broad nature of outcomes related to functioning and QoL, different instruments can be used to measure them; this made it impossible to synthesize results in a meta -analysis for some of the outcomes. Results: Fifteen randomized controlled trials with 414 participants with CP were included. Analysis of the methodological quality of the studies revealed a low risk of bias. The effect of aerobic exercise compared to usual care or other interventions was significant for aerobic capacity (standardized mean difference [SMD] = 0.81; 95% confidence interval [CI] = 0.16-1.47; $p < 0.002$; $I2 = 68\%$), gross motor function (SMD = 0.70; 95% = CI 0.21-1.19; $p = 0.005$; $I2 = 49\%$), mobility (SMD = 0.53; 95% CI = 0.05-1.05; $p = 0.03$; $I2 = 27\%$), balance, and participation (SMD = 0.74; 95% CI = 0.10-1.39; $p = 0.02$; $I2 = 0\%$). Aerobic exercise was not effective for muscle strength, spasticity, gait parameters, and QoL ($p > 0.05$). The certainty of evidence for most comparisons was moderate to low. Interpretation: This review provides the most up -to-date evidence on the efficacy of aerobic exercise on the functioning and QoL of children and adolescents with CP.

PMID: 36895173

6. Determinants of Frame Running capacity in athletes with cerebral palsy to improve training routines and classification strategies: A cross-sectional observational study
Emma Hjalmarsson, Cecilia Lidbeck, Laura Barrero Santiago, Jessica Pingel, Jessica Norrbom, Gema Sanz, Alexandra Palmcrantz, Eva Pontén, Ferdinand von Walden, Rodrigo Fernandez-Gonzalo

Am J Phys Med Rehabil. 2023 Mar 7. doi: 10.1097/PHM.0000000000002233. Online ahead of print.

Objective: Investigate 1) what physical and physiological parameters are most important for Frame Running (FR) capacity, a parasport for individuals with ambulatory difficulties, and 2) determine if FR capacity can be predicted in athletes with cerebral palsy (CP). Design: Athletes with CP (n = 62, GMFCS I-V; 2/26/11/21/2) completed a 6-minute FR test (6-MFRT). Before the 6-MFRT, muscle thickness, passive range of motion (hip, knee, ankle), selective motor control, and spasticity (hip, knee, ankle) were measured in both legs. In total, 54 variables per individual were included. Data were analyzed using correlations, Principal Component Analysis (PCA), Orthogonal Partial Least Square (OPLS) regression, and Variable Importance in Projection (VIP) analysis. Results: Mean 6-MFRT distance was 789 ± 335 m and decreased with motor function severity. The OPLS analysis revealed a modest degree of covariance in the variables analyzed, and that the variance in the 6-MFRT distance could be predicted with 75% accuracy based on all the variables measured. VIP analysis indicated hip and knee extensor spasticity (negative effect), and muscle thickness (positive effect) arose as the most important factors contributing to FR capacity. Conclusions: These results are an important resource to enable optimization of training regimes to improve FR capacity and contribute to evidence-based and fair classification for this parasport.

PMID: 36897812

7. Developmental Cut-Points for Atypical Speech Intelligibility in Children With Cerebral Palsy
Katherine C Hustad, Tristan J Mahr, Jennifer U Soriano, Paul J Rathouz

J Speech Lang Hear Res. 2023 Mar 9;1-11. doi: 10.1044/2022_JSLHR-22-00310. Online ahead of print.

Purpose: Early identification of speech motor involvement (SMI) in children with cerebral palsy (CP) is difficult because of overlapping features with many aspects of typical speech development. Quantitative measures of speech intelligibility have the

potential to differentiate between children with SMI and those with no SMI (NSMI). We examined thresholds for speech intelligibility development in children with CP relative to the low end of age-specific typical developmental expectations. We sought to determine whether there were intelligibility differences between children with CP and NSMI versus typically developing (TD) age-mates across the range of development and whether there were differences between children with CP who have NSMI and those with CP who have SMI across the range of development based on speech intelligibility. Method: We used two large existing data sets that included speech samples from children between the ages of 2.5 and 8 years. One data set included 511 longitudinal speech samples from children with CP; the other included 505 cross-sectional speech samples from TD children. We examined receiver operating characteristic curves and sensitivity/specificity results by age for differentiating among groups of children. Results: TD children versus those with CP and NSMI showed differentiation in their speech intelligibility across all ages, but the strength of differentiation was only marginally above chance. Children with CP and NSMI showed clear differentiation in their speech intelligibility from those with CP and SMI beginning at the earliest age point. Children with CP who have intelligibility below 40% at the age of 3 years have a very high probability of having SMI. Conclusions: Early intelligibility screening should be performed in children diagnosed with CP. Those with intelligibility below 40% at 3 years of age should be referred immediately for speech assessment and treatment.

PMID: 36892950

8. Effects of repetitive transcranial magnetic stimulation on motor function and language ability in cerebral palsy: A systematic review and meta-analysis
Ying-Ying Sun, Lei Wang, Jin-Lin Peng, Yi-Jie Huang, Fu-Qiang Qiao, Pu Wang

Review Front Pediatr. 2023 Feb 16;11:835472. doi: 10.3389/fped.2023.835472. eCollection 2023.

Objective: This review was conducted to assess the quality of the evidence of effectiveness of repetitive transcranial magnetic stimulation (rTMS) in treating motor and language ability of cerebral palsy (CP). Method: Medline, Cochrane library, Web of Science, Embase, PubMed, and CNKI databases were searched up to July 2021 by two independent reviewers. Randomized controlled trials (RCTs) that were published in English and Chinese and met the following criteria were included. The population comprised patients who met the diagnostic criteria for CP. Intervention included the following: comparison about rTMS and sham rTMS or comparison about rTMS combine with other physical therapy and other physical therapy. Outcomes included motor function, as follows: gross motor function measure (GMFM), Gesell Development Diagnosis Scale, fine motor function measure (FMFM), Peabody developmental motor scale, and Modified Ashworth scale. For language ability, sign-significant relation (S-S) was included. Methodological quality was assessed using the Physiotherapy Evidence Database (PEDro) scale. Results: Finally, 29 studies were included in the meta-analysis. Results of evaluation using the Cochrane Collaborative Network Bias Risk Assessment Scale showed that 19 studies specifically explained randomization, among which two studies described allocation concealment, four studies blinded participants and persons and had low risk of bias, and six studies explained that the assessment of outcome measures was blinded. Significant improvements in motor function were observed. The GMFM of total score was determined by using the random-effect model [I2 = 88%; MD = -1.03; 95% CI (-1.35, -0.71); P < 0.0001] and FMFM was determined by using the fixed-effect model [P = 0.40 and I2 = 3%; SMDs = -0.48, 95% CI (-0.65, -0.30); P < 0.01]. For language ability, the language improvement rate was determined using a fixed-effect model [P = 0.88 and I2 = 0%; MD = 0.37, 95% CI (0.23, 0.57); P < 0.01]. According to the PEDro scale, 10 studies had low-quality, four studies had excellent quality, and the other studies had good quality. Using the GRADEpro GDT online tool, we included a total of 31 outcome indicators, as follows: 22 for low quality, seven for moderate quality, and two for very low quality. Conclusion: The rTMS could improve the motor function and language ability of patients with CP. However, rTMS prescriptions varied, and the studies had low sample sizes. Studies using rigorous and standard research designs about prescriptions and large samples are needed to collect sufficient evidence about the effectiveness of using rTMS to treat patients with CP.

PMID: 36873646

9. Organization of rehabilitation services for youth with physical disabilities and mental health problems: A scoping review
Stephanie Tremblay, Shalini Lal, Lucille Xiang, Mark A Ferro, Dana Anaby

Review Front Rehabil Sci. 2023 Feb 20;4:1085827. doi: 10.3389/fresc.2023.1085827. eCollection 2023.

Introduction: Youth with childhood-onset physical disabilities receiving rehabilitation services often present with many

complex needs. Emerging evidence confirms co-occurrence of mental health problems in this population is common, and mental health is often overlooked during rehabilitation for chronic physical conditions. For example, symptoms of depression and anxiety are frequently present in adolescents with physical disability such as spina bifida or Duchenne muscular dystrophy, and access to mental health services is often limited. Addressing mental health concerns for this age group is particularly critical as it encompasses a challenging transition to adulthood. Objectives: Building upon findings from a recent scoping review on the co-occurrence of physical disabilities and mental health problems, this paper synthesizes scientific literature related to the organization and delivery of services for youth with co-occurring childhood-onset physical disabilities (e.g., cerebral palsy, spina bifida) and mental health problems (e.g., depression, anxiety). Methods: A scoping review protocol stemming from Arksey & O'Malley's framework and updated guidelines from the Joanna Briggs Institute was developed. Four databases (Medline, PsycINFO, CINAHL, Embase) were searched. The search was limited to French or English peer-reviewed articles published between 2000 and 2021. Articles included were primary papers addressing: 1) youth aged 15 to 24 with a childhood-onset physical disability, 2) mental health problems, and 3) healthcare service organization or delivery. They were screened by two reviewers and discussed with a third to establish consensus on the inclusion criteria and resolve disagreements. Results: Sixteen articles were retained from the 1,010 screened. Many (9/16) were from the United States. Two models were found: the Biopsychosocial, Collaborative, Agency-Based Service Integration Approach (including psychiatry in a pediatric rehabilitation hospital) and the Client Network Consultation (an interagency collaboration in mental health care for children with complex healthcare needs). Twelve key principles for service organization and delivery were identified and categorized into: collaboration and coordination, training and support, and delivery of care. Conclusion: Identified principles can guide improved service delivery for this population. Highlighted research gaps include the need for developing models of collaborative healthcare delivery and subsequently evaluating their effectiveness.

PMID: 36891032

10. Electromyographic biofeedback-driven gaming to alter calf muscle activation during gait in children with spastic cerebral palsy
Eline Flux, Lynn Bar-On, Annemieke I Buizer, Jaap Harlaar, Marjolein M van der Krogt

Gait Posture. 2023 Feb 19;102:10-17. doi: 10.1016/j.gaitpost.2023.02.012. Online ahead of print.

Background: Children with cerebral palsy often show deviating calf muscle activation patterns during gait, with excess activation during early stance and insufficient activation during push-off. Research question: Can children with cerebral palsy improve their calf muscle activation patterns during gait using one session of biofeedback-driven gaming? Methods: Eighteen children (6-17 y) with spastic cerebral palsy received implicit game-based biofeedback on electromyographic activity of the calf muscle (soleus or gastrocnemius medialis) while walking on a treadmill during one session. Biofeedback alternately aimed to reduce early stance activity, increase push-off activity, and both combined. Early stance and push-off activity and the double -bump-index (early stance divided by push-off activity) were determined during baseline and walking with feedback. Changes were assessed at group level using repeated measures ANOVA with simple contrast or Friedman test with post-hoc Wilcoxon signed rank test, as well as individually using independent t-tests or Wilcoxon rank sum tests. Perceived competence and interest-enjoyment were assessed through a questionnaire. Results: Children successfully decreased their electromyographic activity during early stance feedback trials (relative decrease of 6.8 ± 12.2 %, P = 0.025), with a trend during the combined feedback trials (6.5 ± 13.9 %, P = 0.055), and increased their electromyographic activity during push-off feedback trials (8.1 ± 15.8 %, P = 0.038). Individual improvements were seen in twelve of eighteen participants. All children experienced high levels of interest-enjoyment (8.4/10) and perceived competence (8.1/10). Significance: This exploratory study suggests that children with cerebral palsy can achieve small within-session improvements of their calf muscle activation pattern when provided with implicit biofeedback-driven gaming in an enjoyable manner. Follow-up gait training studies can incorporate this method to assess retention and long-term functional benefits of electromyographic biofeedback-driven gaming.

PMID: 36870265

11. From patient to maker - a workflow including people with cerebral palsy in co-creating assistive devices using 3D printing technologies
Rune Thorsen, Denise Cugnod, Marina Ramella, Rosa Maria Converti, Maurizio Ferrarin

Disabil Rehabil Assist Technol. 2023 Mar 7;1-11. doi: 10.1080/17483107.2023.2177754. Online ahead of print.

Purpose: Digital fabrication, like 3D printing, is a new opportunity for rehabilitation professionals to produce customized

assistive devices. It allows for empowerment and collaboration in device procurement, but practical implementations are scarcely described. We describe the workflow, discuss feasibility and propose directions for future work. Methods: We showcase a process of co-manufacturing a custom spoon handle together with two people with cerebral palsy. Our digital manufacturing process was centered around videoconferencing to remotely control the processes from design to final 3D printing. Device functionality and satisfaction were assessed using standard clinical questionnaires: the Individual Priority Problem Assessment Questionnaire (IPPA) and the Quebec User Satisfaction Assessment with Assistive Technology (QUEST 2.0).Results: IPPA was instrumental in assessing user needs and device effectiveness. QUEST revealed where to focus future design efforts. Conclusion: Involving people with disabilities in co-creation of assistive devices opens for new opportunities for healthcare providers that should be explored in depth using the described methodology. There may also be therapeutic benefits and we envisage specific actions to take in order to make it clinically viable. IMPLICATIONS FOR REHABILITATION: Best practices for co-creation of assistive devices, cost and benefits should be investigated and documented further. Standard questionnaires are useful for measuring effectiveness and satisfaction of co-created devices as well as for guiding design efforts. Co-creation may be a valuable element in therapeutic interventions as an opportunity to unfold creativity.

PMID: 36880457

12. Research Status and Emerging Trends in Virtual Reality Rehabilitation: Bibliometric and Knowledge Graph Study
Ting Fan, Xiaobei Wang, Xiaoxi Song, Gang Zhao, Zhichang Zhang

JMIR Serious Games. 2023 Mar 6;11:e41091. doi: 10.2196/41091.

Background: Virtual reality (VR) technology has been widely used in rehabilitation training because of its immersive, interactive, and imaginative features. A comprehensive bibliometric review is required to help researchers focus on future directions based on the new definitions of VR technologies in rehabilitation, which reveal new situations and requirements. Objective: Herein, we aimed to summarize effective research methods for and potential innovative approaches to VR rehabilitation by evaluating publications from various countries to encourage research on efficient strategies to improve VR rehabilitation. Methods: The SCIE (Science Citation Index Expanded) database was searched on January 20, 2022, for publications related to the application of VR technology in rehabilitation research. We found 1617 papers, and we created a clustered network, using the 46,116 references cited in the papers. CiteSpace V (Drexel University) and VOSviewer (Leiden University) were used to identify countries, institutions, journals, keywords, cocited references, and research hot spots. Results: A total of 63 countries and 1921 institutes have contributed publications. The United States of America has taken the leading position in this field; it has the highest number of publications; the highest h-index; and the largest collaborative network, which includes other countries. The reference clusters of SCIE papers were divided into the following nine categories: kinematics, neurorehabilitation, brain injury, exergames, aging, motor rehabilitation, mobility, cerebral palsy, and exercise intensity. The research frontiers were represented by the following keywords: video games (2017-2021), and young adults (2018-2021). Conclusions: Our study comprehensively assesses the current research state of VR rehabilitation and analyzes the current research hot spots and future trends in the field, with the aims of providing resources for more intensive investigation and encouraging more researchers to further develop VR rehabilitation.

PMID: 36877556

13. Establishing a cerebral palsy registry in Kuwait: An exploratory study
Anwar B Almutairi, Arwa E AlAbdullkarim, Afnan A Al-Shatti

J Taibah Univ Med Sci. 2023 Feb 13;18(5):947-953. doi: 10.1016/j.jtumed.2023.02.001. eCollection 2023 Oct.

Background: Cerebral palsy (CP), the most common motor disability in childhood, comprises a group of permanent non-progressive disorders affecting the antenatal, neonatal, or early postnatal development of areas in the brain responsible for posture and movement. Registries for children with CP, or surveillance programs, have been a source of consistently increasing research productivity; 38 related articles were published in 2013. In Kuwait, a CP registry would provide baseline information on children with CP and their parents. The registry could include demographic information obtained through parental interviews, or review of the mothers' and the children's medical charts. Objective: This study was aimed at exploring the establishment of a pediatric CP registry in Kuwait. Methods: In this exploratory study, caregivers of children with CP were recruited from rehabilitation clinics around Kuwait. The inclusion criteria were 1) boys or girls with a documented diagnosis of CP made between 6 months and 18 years of age, 2) caregivers with permanent residency in Kuwait, and 4) caregivers speaking Arabic and/or English fluently. The variables collected comprised registry and feasibility variables. Registry-associated

variables comprised demographic and medical information about the children, and caregivers' willingness to be contacted for a follow-up or participation in other research projects. Feasibility variables were the percentage of information gathered, and the willingness of caregivers to participate in, and of therapists to recruit for, the registry. Results: Fifty-three caregivers of children with CP participated in this study. The mean age of the recruited children with CP was 5 years and 5 months (SD = 3 y 4 m, range = 11 m to 16 y 8 m/female n = 25). GMFCS level V was reported by half of the sample (n = 29/55.77%). Of the 112 caregivers screened, fewer than half (n = 53 of 112/47.32%) participated in the study. Most caregivers (n = 48/90.56%) used the Arabic version of the form.

PMID: 36875341

14. Population- and individual-level trajectories of opioid prescription patterns among adults with cerebral palsy: a retrospective cohort study
Daniel G Whitney, Mark D Peterson, Edward A Hurvitz

Int J Clin Pharm. 2023 Mar 10;1-12. doi: 10.1007/s11096-023-01553-5. Online ahead of print.

Background: There is little epidemiologic evidence on opioid prescription among adults with cerebral palsy (CP). Aim: To describe the population- and individual-level opioid prescription patterns for adults with versus without CP. Method: This retrospective cohort study used commercial claims (Optum's de-identified Clinformatics® Data Mart Database) from the USA from 01/01/2011-12/31/2017 from adults ≥ 18 years old with CP and matched adults without CP. For the population-level analysis, monthly estimates of opioid exposure were described for adults ≥ 18 years old with CP and matched adults without CP. For the individual-level analysis, group based trajectory modelling (GBTM) was used to identify groups of similar individual-level monthly opioid exposure patterns for adults with CP and matched adults without CP for 1-year starting from their first opioid exposure month. Results: For the population-level, adults with (n = 13,929) versus without (n = 278,538) CP had a higher prevalence of opioid exposure (~ 12%, ~ 8%) and days supplied (median, ~ 23, ~17) monthly over 7 years. For the individual-level, there were 6 trajectory groups for CP (n = 2099) and 5 for non-CP (n = 10,361). Notably, 14% of CP (comprising 4 distinct trajectory groups) and 8% (comprising 3 distinct groups) of non-CP had variably high monthly opioid volume for extended periods; exposure was higher for CP. The remaining had low/absent opioid exposure trajectories; for CP (non-CP), 55.7% (63.3%) had nearly absent exposure and 30.4% (28.9%) had consistently low exposure to opioids. Conclusion: Adults with versus without CP were more likely to be exposed to opioids and for a longer duration, which may alter the risk-benefit balance of opioids.

PMID: 36897434

15. Global prevalence of developmental disabilities in children and adolescents: A systematic umbrella review
Bolajoko O Olusanya, Tracey Smythe, Felix A Ogbo, M K C Nair, Mark Scher, Adrian C Davis

Review Front Public Health. 2023 Feb 16;11:1122009. doi: 10.3389/fpubh.2023.1122009. eCollection 2023.

Aim: The provisions of the United Nation's Sustainable Development Goals (SDGs) for disability-inclusive education have stimulated a growing interest in ascertaining the prevalence of children with developmental disabilities globally. We aimed to systematically summarize the prevalence estimates of developmental disabilities in children and adolescents reported in systematic reviews and meta-analyses. Methods: For this umbrella review we searched PubMed, Scopus, Embase, PsycINFO, and Cochrane Library for systematic reviews published in English between September 2015 and August 2022. Two reviewers independently assessed study eligibility, extracted the data, and assessed risk of bias. We reported the proportion of the global prevalence estimates attributed to country income levels for specific developmental disabilities. Prevalence estimates for the selected disabilities were compared with those reported in the Global Burden of Disease (GBD) Study 2019. Results: Based on our inclusion criteria, 10 systematic reviews reporting prevalence estimates for attention-deficit/hyperactivity disorder, autism spectrum disorder, cerebral palsy, developmental intellectual disability, epilepsy, hearing loss, vision loss and developmental dyslexia were selected from 3,456 identified articles. Global prevalence estimates were derived from cohorts in high-income countries in all cases except epilepsy and were calculated from nine to 56 countries. Sensory impairments were the most prevalent disabilities (approximately 13%) and cerebral palsy was the least prevalent disability (approximately 0.2-0.3%) based on the eligible reviews. Pooled estimates for geographical regions were available for vision loss and developmental dyslexia. All studies had a moderate to high risk of bias. GBD prevalence estimates were lower for all disabilities except cerebral palsy and intellectual disability. Conclusion: Available estimates from systematic reviews and meta-analyses do not provide representative evidence on the global and regional prevalence of developmental disabilities among children and adolescents

due to limited geographical coverage and substantial heterogeneity in methodology across studies. Population-based data for all regions using other approaches such as reported in the GBD Study are warranted to inform global health policy and intervention.

PMID: 36891340

16. Diagnostic Yield of Exome Sequencing in Cerebral Palsy and Implications for Genetic Testing Guidelines: A Systematic Review and Meta-analysis
Pedro J Gonzalez-Mantilla, Yirui Hu, Scott M Myers, Brenda M Finucane, David H Ledbetter, Christa L Martin, Andres Moreno-De-Luca

Importance: Exome sequencing is a first-tier diagnostic test for individuals with neurodevelopmental disorders, including intellectual disability/developmental delay and autism spectrum disorder; however, this recommendation does not include cerebral palsy. Objective: To evaluate if the diagnostic yield of exome or genome sequencing in cerebral palsy is similar to that of other neurodevelopmental disorders. Data sources: The study team searched PubMed for studies published between 2013 and 2022 using cerebral palsy and genetic testing terms. Data were analyzed during March 2022. Study selection: Studies performing exome or genome sequencing in at least 10 participants with cerebral palsy were included. Studies with fewer than 10 individuals and studies reporting variants detected by other genetic tests were excluded. Consensus review was performed. The initial search identified 148 studies, of which 13 met inclusion criteria. Data extraction and synthesis: Data were extracted by 2 investigators and pooled using a random-effects meta-analysis. Incidence rates with corresponding 95% CIs and prediction intervals were calculated. Publication bias was evaluated by the Egger test. Variability between included studies was assessed via heterogeneity tests using the I2 statistic. Main outcomes and measures: The primary outcome was the pooled diagnostic yield (rate of pathogenic/likely pathogenic variants) across studies. Subgroup analyses were performed based on population age and on the use of exclusion criteria for patient selection. Results: Thirteen studies were included consisting of 2612 individuals with cerebral palsy. The overall diagnostic yield was 31.1% (95% CI, 24.2%-38.6%; I2 = 91%). The yield was higher in pediatric populations (34.8%; 95% CI, 28.3%-41.5%) than adult populations (26.9%; 95% CI, 1.2%-68.8%) and higher among studies that used exclusion criteria for patient selection (42.1%; 95% CI, 36.0%-48.2%) than those that did not (20.7%; 95% CI, 12.3%-30.5%). Conclusions and relevance: In this systematic review and meta-analysis, the genetic diagnostic yield in cerebral palsy was similar to that of other neurodevelopmental disorders for which exome sequencing is recommended as standard of care. Data from this meta-analysis provide evidence to support the inclusion of cerebral palsy in the current recommendation of exome sequencing in the diagnostic evaluation of individuals with neurodevelopmental disorders.

PMID: 36877506

17. All Patients With a Cerebral Palsy Diagnosis Merit Genomic Sequencing
Clare van Eyk, Suzanna C MacLennan, Alastair H MacLennan

JAMA Pediatr. 2023 Mar 6. doi: 10.1001/jamapediatrics.2023.0015. Online ahead of print.

No abstract available

PMID: 36877500

18. Ideal Mode of Auditory Stimulation in Preterm Neonates in Neonatal Intensive Care Unit: A Systematic Review
Pallavi Palaskar, Shruti D Ramekar, Namrata Sant, Rinkle J Malani

Review Cureus. 2023 Feb 1;15(2):e34496. doi: 10.7759/cureus.34496. eCollection 2023 Feb.

The objective of this review was to find out the best mode of auditory stimulation for preterm neonates admitted to the neonatal intensive care unit. We also aimed to find out the different effects of different types of auditory stimulation in these neonates. Advanced neonatal care and technological advances in neonatal intensive care units have led to increased survival of preterm-

born neonates, but this in turn leads to increased incidences of disabilities like cerebral palsy, visual impairment, delayed social development, etc. Early intervention is provided to facilitate further development and prevent delays in all domains. Auditory stimulation is proven to benefit these neonates to stabilize their vitals and improve their auditory performance in later life. Different modes of auditory stimulation have been studied worldwide, but none of the studies has presented the ideal mode of auditory stimulation for these preterm neonates. In this review, we have discussed the effects produced by different types of auditory stimulation and compared their pros and cons. For conducting a systematic review, a search strategy adopted by MEDLINE is used. A total of 78 articles published between 2012 and 2017, on the effects of auditory stimulation on preterm infants' performance were reviewed. Out of that, eight studies that met the inclusion criteria and focused on short-term and long -term effects were included in this systematic review. Search terms included preterm neonates, auditory stimulation, and early intervention. Randomized controlled trials and cohort studies were included. Auditory stimulation by maternal sound provides physiological and autonomic stability, but the behavioral states of preterm neonates improved with auditory stimulation by music therapy with lullabies. Maternal singing during kangaroo care may be recommended for providing physiological stability.

PMID: 36874338

19. Cerebral palsy and postnatal steroids
Shabih Manzar

Acta Paediatr. 2023 Mar 5. doi: 10.1111/apa.16742. Online ahead of print.

No abstract available

PMID: 36872848

20. Cerebral palsy and Postnatal Steroids - Reply
K A Hodgson, B J Manley, J L Y Cheong

Acta Paediatr. 2023 Mar 5. doi: 10.1111/apa.16743. Online ahead of print.

No abstract available

PMID: 36872528

21. Harnessing neuroplasticity to improve motor performance in infants with cerebral palsy: a study protocol for the GAME randomised controlled trial
Catherine Morgan, Nadia Badawi, Roslyn N Boyd, Alicia J Spittle, Russell C Dale, Adrienne Kirby, Rod W Hunt, Koa Whittingham, Kerstin Pannek, Rachael L Morton, William Tarnow-Mordi, Michael C Fahey, Karen Walker, Kristina Prelog, Catherine Elliott, Jane Valentine, Andrea Guzzetta, Shannon Olivey; GAME study team; Iona Novak

BMJ Open. 2023 Mar 10;13(3):e070649. doi: 10.1136/bmjopen-2022-070649.

Introduction: Cerebral palsy (CP) is the most common physical disability of childhood worldwide. Historically the diagnosis was made between 12 and 24 months, meaning data about effective early interventions to improve motor outcomes are scant. In high-income countries, two in three children will walk. This evaluator-blinded randomised controlled trial will investigate the efficacy of an early and sustained Goals-Activity-Motor Enrichment approach to improve motor and cognitive skills in infants with suspected or confirmed CP. Methods and analysis: Participants will be recruited from neonatal intensive care units and the community in Australia across four states. To be eligible for inclusion infants will be aged 3-6.5 months corrected for prematurity and have a diagnosis of CP or 'high risk of CP' according to the International Clinical Practice Guideline criteria. Eligible participants whose caregivers consent will be randomly allocated to receive usual care or weekly sessions at home from a GAME-trained study physiotherapist or occupational therapist, paired with a daily home programme, until age 2. The

study requires 150 participants per group to detect a 0.5 SD difference in motor skills at 2 years of age, measured by the Peabody Developmental Motor Scales-2. Secondary outcomes include gross motor function, cognition, functional independence, social-emotional development and quality of life. A within-trial economic evaluation is also planned. Ethics and dissemination: Ethical approval was obtained from the Sydney Children's Hospital Network Human Ethics Committee in April 2017 (ref number HREC/17/SCHN/37). Outcomes will be disseminated through peer-reviewed journal publications, presentations at international conferences and consumer websites. Trial registration number: ACTRN12617000006347.

PMID: 36898755

22. Vibration Therapy as an Early Intervention for Children Aged 2-4 Years with Cerebral Palsy: A Feasibility Study
Alena Adaikina, José G B Derraik, Janice Taylor, Gina L O'Grady, Paul L Hofman, Silmara Gusso

Phys Occup Ther Pediatr. 2023 Mar 5;1-18. doi: 10.1080/01942638.2023.2181723. Online ahead of print.

Aims: To evaluate the feasibility and acceptability of vibration therapy (VT) in preschool children with cerebral palsy (CP) and obtain preliminary data on its potential effectiveness. Methods: Nine children aged 2.5-4.8 years (4 boys) with CP GMFCS levels I-III participated in a single-group feasibility study, undergoing a 12-week control period without intervention, followed by 12 weeks of home-based VT (four times/week, 9 min/day, frequency 20 Hz). We assessed adherence to VT protocol, adverse events, and family acceptability of VT. Clinical assessments included motor function (GMFM-66), body composition (DXA), mobility (10-meter walk/run test), and health-related quality of life (PedsQL). Results: VT was well tolerated and acceptable to families, with high adherence levels reported (mean = 93%). There were no observed between-period differences (ΔControl vs ΔVT) except for an improvement in the PedsQL "Movement & Balance" dimension with VT ($p = 0.044$). Nonetheless, changes after the VT but not the Control period were suggestive of potential treatment benefits for mobility, gross motor function, and body composition (lean mass and legs bone mineral density).Conclusion: Home-based VT is feasible and acceptable for preschool children with CP. Our preliminary data suggest potential health benefits from VT for these children, supporting larger randomized trials to assess its effectiveness properly. Clinical trial registration number: Australian New Zealand Clinical Trials Registry (ACTRN12618002027291).

PMID: 36872600

23. Clinical measures for children with cerebral palsy: Challenges and approaches
Peter Rosenbaum

Dev Med Child Neurol. 2023 Mar 7. doi: 10.1111/dmcn.15573. Online ahead of print.

No abstract available

PMID: 36882987

24. Pediatric Outpatient Occupational Therapy: Transitioning From In-Person Services to Telehealth
Kelly Tanner, Jinyu Xu, Sara O'Rourke, Rajesh Ganta, Xu Zhang

OTJR (Thorofare N J). 2023 Mar 6;15394492231155090. doi: 10.1177/15394492231155090. Online ahead of print.

The COVID-19 pandemic necessitated rapid adoption of telehealth for outpatient pediatric occupational therapy practice. The dose of therapy may have varied across diagnostic and geographical groups despite efforts to ensure access for all patients. The objective of the study was to describe the visit length of outpatient pediatric occupational therapy practice for three diagnostic groups at one institution both during and prior to the COVID-19 pandemic. Retrospective review of electronic health records for two time periods using both practitioner-entered and telecommunications data. Data were analyzed using descriptive statistics and generalized linear mixed model. Prior to the pandemic, average treatment length did not vary by primary diagnosis. During the pandemic, average visit length varied by primary diagnosis, with feeding disorder (FD) visits

significantly shorter than cerebral palsy (CP) and autism spectrum disorder (ASD) visits. During the pandemic, visit length was associated with rurality for the whole sample and for patients with ASD and CP, but not FD. Patients with FD may have been seen for shorter durations during telehealth visits. The technology gap may affect services for patients living in rural communities.

PMID: 36879452

25. Gross motor function profile of children with cerebral palsy in a low-resource setting: A call for reflection on the model of care
Triveni Shetty, Sailakshmi Ganesan, Ashok Johari, Rajani Mullerpatan

J Pediatr Rehabil Med. 2023 Mar 3. doi: 10.3233/PRM-220039. Online ahead of print.

Purpose: The current study aimed to explore Gross Motor Function Measure (GMFM) profiles among children with cerebral palsy (CP) at various Gross Motor Function Classification System (GMFCS) levels in a low-resource setting. Seventy-one ambulatory children with CP (61% males), were studied after signed informed consent was obtained from parents and assent from children older than 12 years. Methods: Ambulatory capacity of children with CP was classified using GMFCS levels. Functional ability of all participants was measured using GMFM-88. Results: Children with CP in a low-resource setting had 12-44% lower GMFM scores in dimensions of standing, walking, running, and jumping with reference to children from high-resource settings with similar ambulatory capacity reported previously. The most affected components across various GMFCS levels were 'sitting on a large and small bench from floor,' 'arm-free squatting,' 'half-kneeling,' 'kneel-walking,' and 'single-limb hop'. Conclusion: Knowledge of GMFM profiles can guide clinicians and policymakers in low-resource settings for strategic rehabilitation planning and extend the focus of rehabilitation from restoration of body structure and function to the wider domain of social participation in leisure, sport, work, and the community at large. Additionally, providing tailored rehabilitation based on a profile of motor function can ensure an economically, environmentally, and socially sustainable future.

PMID: 36872801

26. Improvement in social capital and health of children with cerebral palsy: Evidence from resource-poor settings
Bayu Arie Fianto

Dev Med Child Neurol. 2023 Mar 5. doi: 10.1111/dmcn.15566. Online ahead of print.

No abstract available

PMID: 36872462

27. Correction: Osteopenia in children with cerebral palsy can be treated with oral alendronate
Muhammet Sukru Paksu, Sebahattin Vurucu, Abdulbaki Karaoglu, Alper Ozgur Karacalioglu, Ahmet Bolat, Ozgur Yesilyurt, Bulent Unay, Ridvan Akin

Published Erratum Childs Nerv Syst. 2023 Mar 7. doi: 10.1007/s00381-023-05890-8. Online ahead of print.

No abstract available

PMID: 36879175

28. Health care transition for cerebral palsy with intellectual disabilities: A systematic review
M Osako, Y Yamaoka, C Takeuchi, Y Mochizuki, T Fujiwara

Rev Neurol (Paris). 2023 Mar 2;S0035-3787(23)00820-2. doi: 10.1016/j.neurol.2022.11.013. Online ahead of print.

Objective: Today, most individuals with cerebral palsy are adults who need a paediatric-to-adult health care transition. However, many remain in paediatric care for treatment of adult-onset health issues. Therefore, a systematic review based on the 'Triple Aim' framework was performed to determine the status of paediatric-to-adult health care transition for people with cerebral palsy. A comprehensive evaluation of transitional care was proposed for using this framework. It consists of 'experience of care', meaning satisfaction with the care, 'population health', meaning the well-being of patients, and 'cost', meaning cost-effectiveness. Method: Electronic database (PubMed) searches were performed. The inclusion criteria were original articles published between 1990 and 2020. The search terms used in this study were ('cerebral palsy' AND 'transition to adult health care') OR ('cerebral palsy' AND 'transition'). The study type had to be epidemiological, case report, case-control, and cross-sectional, but not qualitative. The outcomes of the studies were categorised into 'care experience', 'population health', and 'cost', according to the Triple Aim framework. Results: Thirteen articles met the abovementioned inclusion criteria. Few studies have examined the effect of the intervention of transition for young adults with cerebral palsy. Participants in some studies had no intellectual disability. Young adults were dissatisfied with the 'care experience', 'population health', and 'cost' and had unmet health needs and inadequate social participation. Interpretation: Further transition intervention studies with a comprehensive assessment and proactive involvement of individuals are warranted. The presence of an intellectual disability should be considered.

PMID: 36870883

Prevention and Cure

29. Umbilical cord-derived mesenchymal stromal cell therapy to prevent the development of neurodevelopmental disorders related to low birth weight
Masahiro Tsuji, Takeo Mukai, Yoshiaki Sato, Yasue Azuma, Saki Yamamoto, Florence Cayetanot, Laurence Bodineau, Atsuto Onoda, Tokiko Nagamura-Inoue, Jacques-Olivier Coq

Sci Rep. 2023 Mar 7;13(1):3841. doi: 10.1038/s41598-023-30817-3.

Low birth weight (LBW) increases the risk of neurodevelopmental disorders (NDDs) such as attention-deficit/hyperactive disorder and autism spectrum disorder, as well as cerebral palsy, for which no prophylactic measure exists. Neuroinflammation in fetuses and neonates plays a major pathogenic role in NDDs. Meanwhile, umbilical cord-derived mesenchymal stromal cells (UC-MSCs) exhibit immunomodulatory properties. Therefore, we hypothesized that systemic administration of UC-MSCs in the early postnatal period may attenuate neuroinflammation and thereby prevent the emergence of NDDs. The LBW pups born to dams subjected to mild intrauterine hypoperfusion exhibited a significantly lesser decrease in the monosynaptic response with increased frequency of stimulation to the spinal cord preparation from postnatal day 4 (P4) to P6, suggesting hyperexcitability, which was improved by intravenous administration of human UC-MSCs (1 × 105 cells) on P1. Three-chamber sociability tests at adolescence revealed that only LBW males exhibited disturbed sociability, which tended to be ameliorated by UC-MSC treatment. Other parameters, including those determined via open-field tests, were not significantly improved by UC-MSC treatment. Serum or cerebrospinal fluid levels of pro-inflammatory cytokines were not elevated in the LBW pups, and UC-MSC treatment did not decrease these levels. In conclusion, although UC-MSC treatment prevents hyperexcitability in LBW pups, beneficial effects for NDDs are marginal.

PMID: 36882440

30. Characteristics of Children With Cerebral Palsy in the Post-Therapeutic Hypothermia Era
Heather Pekeles, Fatema Al Amrani, Marta Perez-Morgui, Pia Wintermark, Michael Shevell

J Child Neurol. 2023 Mar 5;8830738231159162. doi: 10.1177/08830738231159162. Online ahead of print.

Objectives: To explore the profile of children with cerebral palsy secondary to intrapartum asphyxia treated with therapeutic hypothermia after birth and to compare characteristics of children treated with therapeutic hypothermia with mild vs severe cerebral palsy outcome. Study Design: We identified all children treated with therapeutic hypothermia for intrapartum asphyxia in a single-center tertiary-level neonatal intensive care unit from 2008 to 2018 with a cerebral palsy outcome. We collected perinatal and outcome measures from patient charts. We searched the literature for characteristics of children with cerebral palsy prior to therapeutic hypothermia (historical cohort) to compare to our cohort. We subdivided our cohort into mild vs severe cerebral palsy and compared neonatal characteristics to identify predictors of severe phenotype. Results: Thirty of 355 cooled neonates (8%) developed cerebral palsy. More children had spastic quadriparesis and epilepsy, and fewer had visual impairment in the post-therapeutic hypothermia era compared to the historical cohort, but had similar Gross Motor Function Classification System scores. In our cohort, more children had severe (19 of 30, 63%) compared to mild cerebral palsy (11 of 30, 37%). The severe group had higher mean birth weight, lower 5- and 10-minute Apgar scores, and more often white matter injury with associated deep gray matter injury or near-total injury pattern ($P < .05$). Conclusions: Our data demonstrated more infants with severe rather than mild cerebral palsy in our cohort treated with therapeutic hypothermia. Birthweight, 5- and 10-minute Apgar scores, and magnetic resonance imaging (MRI) findings were significantly different between mild and severe phenotype groups. Our findings can guide clinicians how to better weigh these factors, when counseling parents in the neonatal period.

PMID: 36872628