

Cerebral Palsy Alliance is delighted to bring you this free weekly bulletin of the latest published research into cerebral palsy. Our organisation is committed to supporting cerebral palsy research worldwide - through information, education, collaboration and funding. Find out more at cerebralpalsy.org.au/our-research

Professor Nadia Badawi AM
CP Alliance Chair of Cerebral Palsy Research

[Subscribe to CP Research News](#)

Interventions and Management

1. Altered upper limb kinematics in individuals with dyskinetic cerebral palsy in comparison with typically developing peers - A statistical parametric mapping study

Inti Vanmechelen, Kaat Desloovere, Helga Haberfehlner, Brian Martens, Jeroen R Vermeulen, Annemieke I Buizer, Jean-Marie Aerts, Hilde Feys, Elegast Monbaliu

Gait Posture. 2023 Jun 17;S0966-6362(23)00159-5. doi: 10.1016/j.gaitpost.2023.06.010. Online ahead of print.

Background: Dyskinetic cerebral palsy (DCP) is clinically characterized by involuntary movements and abnormal postures, which can aggravate with activity. While upper limb movement variability is often detected in the clinical picture, it remains unknown how movement patterns of individuals with DCP differ from typically developing (TD) peers. Research question: Do individuals with DCP show i) higher time-dependent standard deviations of upper limb joint angles and ii) altered upper limb kinematics in time and/or amplitude during functional upper limb tasks in comparison with TD individuals? Methods: Three-dimensional upper limb movement patterns were cross-sectionally compared in 50 individuals with and without DCP during three functional tasks: reach forward (RF), reach and grasp vertical (RGV) and reach sideways (RS). Mean and point-wise standard deviations of angular waveform of the upper limb joint angles were compared between groups to evaluate differences in time and/or amplitude using traditional and non-linear registration statistical parametric mapping. Results: Thirty-five extremities from 30 individuals (mean age 17y4m, range 5-25 y; MACS level I(n = 2); II(n = 15); III(n = 16); IV(n = 2)) with DCP and twenty TD individuals (mean age 16y8m, range 8-25 y) were evaluated. The DCP compared to TD group showed higher point-wise standard deviations at the level of all joints, which was time-dependent and varied between tasks. Mean wrist and elbow flexion was higher for the DCP group during RF (0-83 % wrist; 57-100 % elbow), RGV (0-82 % wrist; 12-100 % elbow) and RS (0-43 % wrist; 70-100 % elbow). Significance: This is the first study exploring the movement patterns of individuals with DCP during reaching using quantitative measures. Analyzing these individual movement patterns by statistical parametric mapping (SPM) allows us to focus on both specific joint or on specific timing during the movement cycle. The individual information that this method yields can guide individual therapy aiming to improve reaching function in different parts of the movement cycle or evaluate intervention effects on upper extremity treatment.

PMID: [37344269](#)

2. Stimulating the motor development of very premature infants: effects of early crawling training on a mini-skateboard

Marie-Victorine Dumuids-Vernet, Vincent Forma, Joëlle Provasi, David Ian Anderson, Elodie Hinnekens, Evelyne Soyez, Mathilde Strassel, Léa Guéret, Charlotte Hym, Viviane Huet, Lionel Granjon, Lucie Calamy, Gilles Dassieu, Laurence Boujenah, Camille Dollat, Valérie Biran, Marianne Barbu-Roth

Front Pediatr. 2023 Jun 6;11:1198016. doi: 10.3389/fped.2023.1198016. eCollection 2023.

Aim: To examine the effects of an early home-based 8-week crawling intervention performed by trained therapists on the motor and general development of very premature infants during the first year of life. Methods: At term-equivalent age, immediately following discharge from the Neonatal Intensive Care Unit (NICU), we randomly allocated 44 premature infants

born before 32 weeks' gestation without major brain damage to one of three conditions in our intervention study: crawling on a mini-skateboard, the Crawliskate (Crawli), prone positioning control (Mattress), or standard care (Control). The Crawli and Mattress groups received 5 min daily at-home training administered by trained therapists for 8 consecutive weeks upon discharge from the NICU. The outcomes of greatest interest included gross motor development (Bayley-III) at 2, 6, 9, and 12 months (primary outcome) corrected age (CA), mature crawling at 9 months CA and general development at 9 and 12 months CA [Ages and Stages Questionnaires-3 (ASQ-3)]. The study was registered at www.clinicaltrials.gov; registration number: NCT05278286. Results: A 3 (Condition) \times 4 (Age) repeated measures ANOVA revealed that Crawli group infants had significantly higher Bayley-III gross motor development scores than Mattress and Control group infants. Crawli group infants also scored significantly higher on groups of Bayley-III items related to specific motor skills than infants in the other groups, including crawling at 9 months CA. We found significant differences in favor of the Crawli group in separate one-way ANOVAs at each of the ages we examined. A 3 (Condition) \times 2 (Age) repeated measures ANOVA revealed that the Crawli group scored significantly higher than the Control group for the ASQ-3 total score and communication score and significantly higher for the fine motor score than the Control and Mattress groups. We found additional significant differences in favor of the Crawli group for other dimensions of the ASQ-3 in separate one-way ANOVAs at 9 and 12 months CA. Interpretation: Early crawling training on a Crawliskate provides an effective way to promote motor and general development in very premature infants. The findings also provide clear evidence for a link between newborn crawling and more mature crawling later in development.

PMID: [37346892](https://pubmed.ncbi.nlm.nih.gov/37346892/)

3. Risk Factors of Infectious Complications in Pediatric Patients With Cerebral Palsy After Spinal Arthrodesis

Michael Miskiewicz, Sofia Hidalgo Perea, Matthew Magruder, Amr Abdelgawad

Clin Spine Surg. 2023 Jun 13. doi: 10.1097/BSD.0000000000001471. Online ahead of print.

Study design: This was a retrospective study. Objective: The main objective of this study was to investigate (1) whether pediatric patients with cerebral palsy (CP) have higher rates of postoperative infectious complications after spinal fusion and (2) risk factors for postoperative infections. Summary of background data: Prior studies have shown that patients with CP undergo corrective spine surgery more often than the general population, yet typically have worse postoperative outcomes. Further investigation is needed to improve our understanding of the perioperative factors that place children with CP at greater risk of postoperative infectious complications. Patients and methods: The 2019 "American College of Surgeons National Surgical Quality Improvement Program" Pediatric database was used for patient data. The univariable analysis compared the prevalence of preoperative comorbidities and perioperative factors between children with and without CP. Multivariable logistic regression modeling was used to ascertain independent risk factors for postoperative infectious complications. Results: A total of 4445 patients were included in the study; 606 (13.63%) patients had CP and 3839 (86.37%) did not. Patients with CP were more likely to have several notable preoperative comorbidities, and the rate of developing any infectious complication was more than 7 times greater in the CP cohort than in the control cohort (14.36% vs 1.88%; $P < 0.001$). Multivariable analysis revealed CP [odds ratio (OR): 3.55, CI: 2.25-5.60; $P < 0.001$], American Society of Anesthesiologists class 3 or higher (OR: 2.10, CI: 1.29-3.42; $P = 0.003$), and hematologic disorders (OR: 2.01, CI: 1.06-3.83; $P = 0.033$) to be independent risk factors for increased postoperative infectious complications. Conclusions: CP is an independent risk factor for the development of 30-day postoperative infectious complications in pediatric patients. In addition, the American Society of Anesthesiologists class 3 or higher and hematologic disorders were risk factors for postoperative infections after spinal fusion surgery.

PMID: [37348066](https://pubmed.ncbi.nlm.nih.gov/37348066/)

4. Impact of selected risk factors on motor performance in the third month of life and motor development in the ninth month

Ewa Gajewska, Jerzy Moczko, Mariusz Naczka, Alicja Naczka, Magdalena Sobieska

PeerJ. 2023 Jun 13;11:e15460. doi: 10.7717/peerj.15460. eCollection 2023.

Background: Proper motor development can be influenced by a range of risk factors. The resulting motor performance can be assessed through quantitative and qualitative analysis of posture and movement patterns. Methods: This study was designed as the cohort follow-up of the motor assessment and aimed to demonstrate, in a mathematical way, the impact of particular risk factors on elements of motor performance in the 3rd month and the final motor performance in the 9th month of life. Four hundred nineteen children were assessed (236 male and 183 female), including 129 born preterm. Each child aged 3 months underwent a physiotherapeutic assessment of the quantitative and qualitative development, in the prone and supine positions. The neurologist examined each child aged 9 months, referring to the Denver Development Screening Test II and assessing reflexes, muscle tone and symmetry. The following risk factors were analyzed after the neurological consultation: condition at birth (5th min Apgar score), week of gestation at birth, intraventricular hemorrhage, respiratory distress syndrome, and the incidence of intrauterine hypotrophy and hyperbilirubinemia determined based on medical records. Results: A combination of several risk factors affected motor development stronger than any one of them solely, with Apgar score, hyperbilirubinemia, and intraventricular hemorrhage exhibiting the most significant impact. Conclusions: Premature birth on its own did not cause a

substantial delay in motor development. Nonetheless, its co-occurrence with other risk factors, namely intraventricular hemorrhage, respiratory distress syndrome, and hyperbilirubinemia, notably worsened motor development prognosis. Moreover, improper position of the vertebral column, scapulae, shoulders, and pelvis in the third month of life may predict disturbances in further motor development.

PMID: [37334124](#)

5. Ultrasound findings in painful spastic hip. Muscle thickness in children with cerebral palsy

Claudia Guízar-Sánchez, Cristina Hernández-Díaz, Diana Guízar-Sánchez, Ana Victoria Meza-Sánchez, Alejandra Torres-Serrano, María Elena Camacho Cruz, Lucio Ventura-Ríos

BMC Musculoskelet Disord. 2023 Jun 22;24(1):512. doi: 10.1186/s12891-023-06610-8.

Background: In cerebral palsy (CP), spasticity is the dominant symptom and hip pain is one of the most common secondary conditions. Aetiology is not clear. Musculoskeletal ultrasound (MSUS) is a low-cost, non-invasive imaging technique that allows assessment of structural status, dynamic imaging, and quick contralateral comparison. **Objective:** A retrospective case-matched-control study. To investigate associated factors with painful spastic hip and to compare ultrasound findings (focusing on muscle thickness) in children with CP vs. typically developing (TD) peers. **Setting:** Paediatric Rehabilitation Hospital in Mexico City, from August to November 2018. **Participants:** 21 children (13 male, 7 + 4.26 years) with CP, in Gross Motor Function Classification System (GMFCS) levels IV to V, with spastic hip diagnosis (cases) and 21 children age- and sex-matched (7 + 4.28 years) TD peers (controls). **Characteristically data:** Sociodemographic data, CP topography, degree of spasticity, mobility arch, contractures, Visual Analog Scale (VAS), GMFCS, measurements of the volumes of eight major muscles of the hip joint and MSUS findings of both hips. **Results:** All children with CP group reported chronic hip pain. Associated factors for hip pain (high VAS hip pain score) were degree of hip displacement (percentage of migration), Ashworth Level, GMFCS level V. No synovitis, bursitis or tendinopathy was found. Significant differences ($p < 0.05$) were found in muscle volumes in all hip muscles (right and left) except in the right and left adductor longus. **Conclusion:** Though possibly the most important issue with diminished muscle growth in CP children is the influence on their long-term function, it is likely that training routines that build muscle size may also increase muscle strength and improve function in this population. To improve the choice of treatments in this group and maintain muscle mass, longitudinal investigations of the natural history of muscular deficits in CP as well as the impact of intervention are needed.

PMID: [37349815](#)

6. Reliability and validity of assessing lower-limb muscle architecture of patients with cerebral palsy (CP) using ultrasound: A systematic review

Wei Liu, Hui Dong Wu, Yan To Ling, Queenie Tsung Kwan Shea, Vaheh Nazari, Yong-Ping Zheng, Christina Zong-Hao Ma

Review J Clin Ultrasound. 2023 Jun 18. doi: 10.1002/jcu.23498. Online ahead of print.

Aims: To investigate the reliability, validity, and level of evidence of applying ultrasound in assessing the lower-limb muscles of patients with cerebral palsy (CP). **Method:** Publications in Medline, PubMed, Web of Science, and Embase were searched on May 10, 2023, to identify and examine relevant studies investigating the reliability/validity of ultrasound in evaluating the architecture of CP lower-limb muscles systematically, following the Preferred Reporting Items for Systematic Reviews and Meta-Analysis 2020 guidelines. **Results:** Out of 897 records, 9 publications with 111 CP participants aged 3.8-17.0 years were included (8 focused on intra-rater and inter-rater reliability, 2 focused on validity, and 4 were with high quality). The ultrasound-based measurements of muscle thickness (intra-rater only), muscle length, cross-sectional area, muscle volume, fascicle length, and pennation angle showed high reliability, with the majority of intraclass correlation coefficient (ICC) values being larger than 0.9. Moderate-to-good correlations between ultrasound and magnetic resonance imaging measurements existed in muscle thickness and cross-sectional area ($0.62 \leq ICC \leq 0.82$). **Interpretation:** Generally, ultrasound has high reliability and validity in evaluating the CP muscle architecture, but this is mainly supported by moderate and limited levels of evidence. More high-quality future studies are needed.

PMID: [37334435](#)

7. Individualized orthotic alignment and footwear for balance and mobility in children with bilateral spastic cerebral palsy: A randomized trial

Kristie F Bjornson, Stefania Fatone, Michael Orendurff, Chuan Zhou, Philip M Hurvitz, Garth Shippen

Dev Med Child Neurol. 2023 Jun 20. doi: 10.1111/dmcn.15675. Online ahead of print.

Aim: To examine whether designed-to-be-rigid ankle-foot orthoses and footwear combinations with individualized alignment

and footwear designs (AFO-FC/IAFD) would be more effective than designed-to-be-rigid AFO with non-individualized alignment and footwear designs (AFO-FC/NAFD) in children with cerebral palsy (CP). Method: Nineteen children with bilateral spastic CP were randomized to AFO-FC/NAFD (n = 10) or AFO-FC/IAFD (n = 9) groups. Fifteen were male, average age 6 years 11 months (range 4 years 2 months-9 years 11 months), classified in Gross Motor Function Classification System levels II (n = 15) and III (n = 4). The Pediatric Balance Scale (PBS), Gait Outcomes Assessment List (GOAL), Patient-Reported Outcomes Measurement Information System (PROMIS), and Orthotic and Prosthetic Users' Survey (OPUS) measures of satisfaction were collected at baseline and after 3 months' wear. Results: Compared with the AFO-FC/NAFD group, those with AFO-FC/IAFD demonstrated greater change in PBS total scores (mean 12.8 [standard deviation 10.5] vs 3.5 [5.8]; p = 0.03) and GOAL total scores (3.5 [5.8] vs -0.44 [5.5]; p = 0.03). There were no significant changes in OPUS or PROMIS scores. Interpretation: After 3 months, individualized orthosis alignment and footwear designs had a greater positive effect on balance and parent-reported mobility than a non-individualized approach. No effect was documented for the PROMIS and OPUS. Results may inform orthotic management for ambulatory children with bilateral spastic CP.

PMID: [37340674](#)

8. Moving forward: The importance of tailored orthotic management in children with cerebral palsy

Cristina Bayón

Dev Med Child Neurol. 2023 Jun 22. doi: 10.1111/dmcn.15684. Online ahead of print.

No abstract available

PMID: [37349957](#)

9. Eight weeks of proprioceptive neuromuscular facilitation stretching and static stretching do not affect muscle-tendon properties, muscle strength, and joint function in children with spastic cerebral palsy

Annika Kruse, Andreas Habersack, Guido Weide, Richard T Jaspers, Martin Svehlik, Markus Tilp

Clin Biomech (Bristol, Avon). 2023 Jun 7;107:106011. doi: 10.1016/j.clinbiomech.2023.106011. Online ahead of print.

Background: While the effect of static stretching for individuals with cerebral palsy is questionable, recent results suggest that the combination with activation seems promising to improve muscle-tendon properties and function. Therefore, this study analyzed the effects of 8-week proprioceptive neuromuscular facilitation stretching on the gastrocnemius medialis muscle-tendon properties, muscle strength, and the ankle joint in children with spastic cerebral palsy in comparison to static stretching. Methods: Initially, 24 children with spastic cerebral palsy were randomly assigned to a static stretching (10.7 ± 1.8 years) or proprioceptive neuromuscular facilitation stretching group (10.9 ± 2.6 years). Plantar flexors were manually stretched at home for 300 s and ~ 250-270 s per day four times a week for eight weeks, respectively. Assessments of ankle joint function (e.g., range of motion), muscle-tendon properties, and isometric muscle strength were conducted using 3D motion capture, 2D ultrasound, dynamometry, and electromyography. A mixed analysis of variance was used for the statistical analysis. Findings: Stretching adherence was high in the proprioceptive neuromuscular facilitation stretching (93.1%) and static stretching group (94.4%). No significant changes (p > 0.05) were observed in ankle joint function, muscle-tendon properties, and isometric muscle strength after both interventions. Moreover, no differences (p > 0.05) were found between the stretching techniques. Interpretation: The findings support the idea that manual stretching (neither proprioceptive neuromuscular facilitation stretching nor static stretching) performed in isolation for eight weeks may not be appropriate to evoke significant changes in muscle-tendon properties, voluntary muscle strength, or joint function in children with spastic cerebral palsy.

PMID: [37329655](#)

10. Physical conditioning in children and adolescents with cerebral palsy: Systematic review and meta-analysis

Kamilla Passini Santos, Verônica Mirian Machado da Silva, Isabella Ferreira Dos Reis, Mariana Rodrigues Carvalho de Aquino, Mariana Ribeiro Volpini Lana, Cláudia Maria Monteiro de Freitas Teixeira

Review J Bodyw Mov Ther. 2023 Jul;35:158-163. doi: 10.1016/j.jbmt.2023.04.036. Epub 2023 Apr 24.

Introduction: Cerebral palsy (CP) can be described as a group of permanent non-progressive disorders that occur in the developing fetal or infant brain. Studies have shown that children and adolescents with CP have low cardiorespiratory fitness and higher energy expenditure during daily activities when compared to typical children. Therefore, interventions focused on the physical conditioning of this population could be critical. Objective: To evaluate the effect of physical conditioning training on distance walked and maximum oxygen consumption (VO₂ max) in individuals with CP, through a systematic review. Method: Two independent researchers performed systematic searches in the PUBMED, SciELO, PEDro, ERIC, and Cochrane databases using the search terms "physical fitness", "aerobic training" or "endurance" combined with "cerebral palsy".

Inclusion criteria: (1) population (children and adolescents with CP between 5 and 18 years old); (2) type of study (experimental studies); (3) type of intervention (physical conditioning protocols); (4) outcome (distance walked in the 6 min walking test - 6MWT, and VO₂ max). Results: 386 studies were identified and 5 articles were considered eligible. After physical conditioning training, there was an increase of 46.34 m ($p = 0.07$) and 5.93. ml. kg⁻¹. min⁻¹ ($p < 0.001$) in the 6MWT and VO₂ max, respectively. Conclusion: Physical conditioning training appears to be clinically beneficial to the cardiorespiratory fitness of children and adolescents with CP.

PMID: [37330763](#)

11. Effects of Powerchair Football: Contextual Factors That Impact Participation

Aurelien Vandenbergue, J P Barfield, Said Ahmaidi, Stephanie Williams, Thierry Weissland

Adapt Phys Activ Q. 2023 Jun 20;1-21. doi: 10.1123/apaq.2022-0124. Online ahead of print.

The aim of this study was to identify contextual factors that negatively affect activity and participation among powerchair football (PF) players. Thirty-seven semistructured interviews were conducted with PF players (Mage = 27.9 ± 8.2 years) in France (n = 18) and the United States (n = 19). Participants reported acute back and neck pain as the primary morbidities resulting from PF participation, with sustained atypical posture in the sport chair as the primary cause. Competition-related physical and mental stress were also identified as participation outcomes. Accompanying the many benefits of PF, participants recognized negative impacts of discomfort, physical fatigue, and mental fatigue. Interventions such as seating modifications, thermotherapy to combat pain, napping to combat acute physical stress, and mental preparation to manage state anxiety were all identified as prospective interventions.

PMID: [37339770](#)

12. Umbrella Systematic Review and Meta-Analysis: Physical Activity as an Effective Therapeutic Strategy for Improving Psychosocial Outcomes in Children and Adolescents

Marianna Purgato, Camilla Cadorin, Eleonora Prina, Madalena Cabral Ferreira, Lidia Del Piccolo, Markus Gerber, Mark Jd Jordans, Giovanni Ostuzzi, Justin Richards, Dorian Rudi, Francesca Vitali, Samuele Cortese, Federico Schena, Corrado Barbui

J Am Acad Child Adolesc Psychiatry. 2023 Jun 16;S0890-8567(23)00314-3. doi: 10.1016/j.jaac.2023.04.017. Online ahead of print.

Objective: Physical activity (PA) interventions are part of many interdisciplinary programs for the management of children and adolescents with or without physical or psychological conditions or disabilities. Aiming to summarize the available evidence, we conducted an umbrella review of meta-analyses of PA interventions that included psychosocial outcomes in populations of children and adolescents. Method: Literature searches were conducted in PubMed, Cochrane Central, Web of Science, Medline, SportDiscus, and PsychInfo from Jan 1, 2010 to May 6, 2022. Meta-analyses of randomised and quasi-randomised studies investigating the efficacy of PA interventions for psychosocial outcomes in children and adolescents were included. Summary effects were recalculated using common metric and random-effects models. We assessed between-study heterogeneity, predictive intervals, publication bias, small-study effects, and whether the results of the observed positive studies were more than expected due to the chance. On the basis of these calculations, strength of associations was assessed using quantitative umbrella review criteria, and credibility of evidence using the GRADE approach. Quality was assessed using the AMSTAR-II tool. This study is registered with the Open Science Framework, <https://osf.io/ap8qu>. Results: A total of 112 studies from 18 meta-analyses generating 12 new meta-analyses with 21232 children and adolescents with conditions or disabilities as ADHD, cancer, cerebral palsy, chronic respiratory diseases, depression, neuromotor impairment, obesity and in general populations were included. PA interventions were efficacious in reducing psychological symptoms in all meta-analyses across the different population groups using random-effects models. However, umbrella review criteria suggested a weak strength of association for this outcome, and GRADE credibility of evidence ranged from moderate to very low. For psychological wellbeing, three out of five meta-analyses identified significant effects, but the strength of these associations was weak, and GRADE credibility of evidence ranged from moderate to very low. Similarly, for social outcomes, meta-analyses reported a significant summary effect, but the strength of association was weak, and GRADE credibility of evidence ranged from moderate to very low. For self-esteem, one meta-analysis in children with obesity failed to show any effect. Conclusion: Even though existing meta-analyses suggested a beneficial effect of PA interventions on psychosocial outcomes across different population groups, the strength of associations was weak, and the credibility of evidence was variable, depending on the target population, outcome, and condition or disability. Randomised studies of PA interventions in children and adolescents with and without different physical and psychological conditions or disabilities should always include psychosocial outcomes as an important dimension of social and mental health.

PMID: [37331468](#)

13. In Silico Biomarkers of Motor Function to Inform Musculoskeletal Rehabilitation and Orthopedic Treatment

Ilse Jonkers, Erica Beaucage-Gauvreau, Bryce Adrian Killen, Dhruv Gupta, Lennart Scheys, Friedl De Groot

Review J Appl Biomech. 2023 Jun 22;1-10. doi: 10.1123/jab.2023-0029. Online ahead of print.

In this review, we elaborate on how musculoskeletal (MSK) modeling combined with dynamic movement simulation is gradually evolving from a research tool to a promising in silico tool to assist medical doctors and physical therapists in decision making by providing parameters relating to dynamic MSK function and loading. This review primarily focuses on our own and related work to illustrate the framework and the interpretation of MSK model-based parameters in patients with 3 different conditions, that is, degenerative joint disease, cerebral palsy, and adult spinal deformities. By selecting these 3 clinical applications, we also aim to demonstrate the differing levels of clinical readiness of the different simulation frameworks introducing in silico model-based biomarkers of motor function to inform MSK rehabilitation and treatment, with the application for adult spinal deformities being the most recent of the 3. Based on these applications, barriers to clinical integration and positioning of these in silico technologies within standard clinical practice are discussed in the light of specific challenges related to model assumptions, required level of complexity and personalization, and clinical implementation.

PMID: [37348849](#)

14. Intervention with the CO-OP Approach leads to a transfer effect over time to untrained goals for children with cerebral palsy or spina bifida

Ann-Marie Öhrvall, Caisa Hofgren, Barbro Lindquist, Lena Bergqvist, Kate Himmelmann, Arve Opheim, Douglas Sjöwall, Katarina Brock, Marie Peny-Dahlstrand

Disabil Rehabil. 2023 Jun 23;1-10. doi: 10.1080/09638288.2023.2225875. Online ahead of print.

Purpose: This study aims to investigate whether the treatment effects, in terms of goal attainment, transfer effects and impact on executive functions, of an intervention in children with cerebral palsy or spina bifida using the Cognitive Orientation to daily Occupational Performance (CO-OP) Approach are maintained over time, from immediately after the intervention to three months afterwards. **Method:** A three-month follow-up study, from an intervention using CO-OP. Thirty-four children (7-16 years) each identified four goals (one untrained to examine transfer) and participated in an eleven-session intervention. Assessments were performed at baseline, immediately after the intervention and at a three-month follow-up using the Canadian Occupational Performance Measure and the Performance Quality Rating Scale. Executive function and self-rated competence were assessed at the same timepoints. **Results:** Statistically significant and clinically relevant improvements in goal achievement were demonstrated for both trained and untrained goals after the intervention and were maintained at follow-up. The clinically relevant improvement in untrained goals continued to increase until follow-up. Self-rated competence increased after the intervention and was maintained at follow-up. **Conclusion:** The CO-OP intervention was effective in achieving and maintaining the children's own goals over time. The transfer effect was confirmed by higher goal attainment for the untrained goals.

PMID: [37353883](#)

15. Dystonia in Childhood: How Insights from Paediatric Research Enrich the Network Theory of Dystonia

Verity M McClelland, Jean-Pierre Lin

Adv Neurobiol. 2023;31:1-22. doi: 10.1007/978-3-031-26220-3_1.

Dystonia is now widely accepted as a network disorder, with multiple brain regions and their interconnections playing a potential role in the pathophysiology. This model reconciles what could previously have been viewed as conflicting findings regarding the neuroanatomical and neurophysiological characteristics of the disorder, but there are still significant gaps in scientific understanding of the underlying pathophysiology. One of the greatest unmet challenges is to understand the network model of dystonia in the context of the developing brain. This article outlines how research in childhood dystonia supports and contributes to the network theory and highlights aspects where data from paediatric studies has revealed novel and unique physiological insights, with important implications for understanding dystonia across the lifespan.

PMID: [37338693](#)

16. Effects of Dual Task Training and Transcranial Direct Current Stimulation in Children with Spastic Cerebral Palsy: A Pilot Randomized Control Trial

Luanda André Collange-Grecco, Camila Cosmo, André Luís Santos Silva, Sueli Rizzutti, Claudia Santos Oliveira, Mauro Muszkat

Dev Neurorehabil. 2023 Jun 23;1-8. doi: 10.1080/17518423.2023.2228400. Online ahead of print.

Objective: Compare the effectiveness of active and sham transcranial direct current stimulation (tDCS) during the training of a dual task in children with spastic cerebral palsy (CP). **Methods:** Thirty children with CP were submitted to ten sessions of either active (n = 15) or sham (n = 15) tDCS over the motor cortex for 20 minutes during the training of a dual task. Pre-intervention, post-intervention and follow-up evaluations involved measures of functional performance, intellectual performance, functional mobility and cortical excitability. **Results:** The combination of active tDCS and dual task training led to improvements in functional mobility as well as functional and intellectual performances one month after the end of the intervention. **Conclusion:** The combination of active tDCS and dual task training demonstrated promising effects for children with spastic CP.

PMID: [37352444](#)

17. [The quality of life of teenagers living with cerebral palsy participating in conductive education] [Article in Hungarian]

Dóra Mladoneczki-Leszko, Anna Kelemen

Orv Hetil. 2023 Jun 18;164(24):948-953. doi: 10.1556/650.2023.32775. Print 2023 Jun 18.

Introduction: For people living with cerebral palsy, it is crucial to determine and monitor their quality of life because it indirectly can predict the completion of their needs and wishes, and their health-related conditions can be subjectively judged. Cerebral palsy is one of the the most common causes of childhood-onset, probably this is the reason why most quality of life studies focus on children and not on adolescents or adults. **Objectives:** The aims of this study were to explore the quality of life of teenagers living with cerebral palsy receiving conductive education provided by the Pető András Faculty of Semmelweis University, and to map the differences and similarities between the perceptions of parents and their adolescent children. **Method:** This is a descriptive, cross-sectional study. We used the CP QoL-Teen quality of life questionnaire for adolescents living with cerebral palsy. 60 adolescents who were diagnosed with cerebral palsy and receive conductive education took part in the research together with their parents. The caregivers answered the proxy version of the CP QoL Teen questionnaire. **Results:** In the population we examined, there is no significant difference between the answers given by parents and teenagers. The highest agreement was found in the chapter of social wellbeing (p = 0.982). **Conclusions:** This study highlights the importance of social relationships in achieving a better quality of life for teenagers living with cerebral palsy. Furthermore, it also points out the high adaptability of the relationship between parents and their adolescent children. Orv Hetil. 2023; 164(24): 948-953.

PMID: [37330981](#)

18. Catalogues of EQ-5D-3L Health-Related Quality of Life Scores for 199 Chronic Conditions and Health Risks for Use in the UK and the USA

Michael Falk Hvidberg, Mónica Hernández Alava

Pharmacoeconomics. 2023 Jun 18. doi: 10.1007/s40273-023-01285-4. Online ahead of print.

Background: Health-related quality of life (HRQoL) measures are essential in economic evaluation, but sometimes primary sources are unavailable, and information from secondary sources is required. Existing HRQoL UK/US catalogues are based on earlier diagnosis classification systems, amongst other issues. A recently published Danish catalogue merged EQ-5D-3L data from national health surveys with national registers containing patient information on ICD-10 diagnoses, healthcare activities and socio-demographics. **Aims:** To provide (1) UK/US EQ-5D-3L-based HRQoL utility population catalogues for 199 chronic conditions on the basis of ICD-10 codes and health risks and (2) regression models controlling for age, sex, comorbidities and health risks to enable predictions in other populations. **Methods:** UK and US EQ-5D-3L value sets were applied to the EQ-5D-3L responses of the Danish dataset and modelled using adjusted limited dependent variable mixture models (ALDVMMs). **Results:** Unadjusted mean utilities, percentiles and adjusted disutilities based on two ALDVMMs with different control variables were provided for both countries. Diseases from groups M, G, and F consistently had the smallest utilities and the largest negative disutilities: fibromyalgia (M797), sclerosis (G35), rheumatism (M790), dorsalgia (M54), cerebral palsy (G80-G83), post-traumatic stress disorder (F431), dementia (F00-2), and depression (F32, etc.). Risk factors, including stress, loneliness, and BMI30+, were also associated with lower HRQoL. **Conclusions:** This study provides comprehensive catalogues of UK/US EQ-5D-3L HRQoL utilities. Results are relevant in cost-effectiveness analysis, for NICE submissions, and for comparing and identifying facets of disease burden.

PMID: [37330973](#)

19. Global hotspots and trends in research on preschool children's motor development from 2012 to 2022: a bibliometric analysis

Jun-Wei Wang, Sha Qu, Zhi-Cheng Zhu, Xing Zhao, Wen-Jing Song, Xue Li, Wan-Di Chen, Dong-Mei Luo

Review Front Public Health. 2023 Jun 2;11:1118674. doi: 10.3389/fpubh.2023.1118674. eCollection 2023.

Background: Motor development plays an important role in human development throughout the lifespans, from conception to death, and has received increasing scholarly attention in recent years. However, valuable comprehensive reviews and literature analysis on this topic are still lacking. Here, this bibliometric study aimed to identify global motor development research hotspots and trends on preschool children's motor development from 2012 to 2022. Methods: CiteSpace 6.1.R4 was used to visualize and analyze general bibliometric characteristics, research hotspots, and trends through a review of 2,583 articles on the motor development of preschool children, which were published from 2012 to 2022 and included in the Web of Science Core Collection. Results: Research on motor development in preschool children has been carried out into a phase of rapid development. The top five frequently occurring keywords were physical activity (n = 489), performance (n = 319), intervention (n = 222), health (n = 196), and executive function (n = 165); The top five keywords in terms of centrality are academic achievement (0.22), low birth weight (0.16), association (0.14), brain (0.13), and cerebral palsy (0.13). Thirteen keyword clusters were produced from the log-likelihood ratio (Q = 0.74, S = 0.88), and five research topics has been received focused attention in recent years. The keywords with the strongest citation bursts in the last 5 years are developing country (S = 5.92), school-aged children (S = 5.86), middle-income country (S = 3.46), efficacy (S = 5.41), readiness (S = 3.21), motor proficiency (S = 3.6), and screen time (S = 3.3), indicating newly emerging research trends. Conclusion: The results indicated that interventions involving fundamental movement skills, cognitive function, 24-h movement behaviors, neurodevelopmental disorders, and health-related fitness were hot topics in the field of motor development over the last decade. Emerging research trends generally center on school readiness, socioeconomic status, motor proficiency, and screen time.

PMID: [37333555](#)

20. Policy brief: adaptive cycling equipment for individuals with neurodevelopmental disabilities as durable medical equipment

Mary E Gannotti, Margaret E O'Neil, Maria Fragala-Pinkham, George E Gorton 3rd, Daniel G Whitney

Front Rehabil Sci. 2023 Jun 5;4:1160948. doi: 10.3389/fresc.2023.1160948. eCollection 2023.

Durable medical equipment (DME) policies require that the equipment be medically necessary; however, adaptive cycling equipment (bicycles and tricycles) are usually not deemed medically necessary. - Individuals with neurodevelopmental disabilities (NDD) are at high risk for secondary conditions, both physical and mental, that can be mitigated by increasing physical activity. - Significant financial costs are associated with the management of secondary conditions. - Adaptive cycling can provide improved physical health of individuals with NDD potentially reducing costs of comorbidities. - Expanding DME policies to include adaptive cycling equipment for qualifying individuals with NDD can increase access to equipment. - Regulations to ensure eligibility, proper fitting, prescription, and training can optimize health and wellbeing. - Programs for recycling or repurposing of equipment are warranted to optimize resources.

PMID: [37342677](#)

21. Developmental trajectories of spoken language comprehension and functional communication in children with cerebral palsy: A prospective cohort study

Emma Vaillant, Kim J Ostrom, Heleen Beckerman, R Jeroen Vermeulen, Annemieke I Buizer, Johanna J M Geytenbeek

Dev Med Child Neurol. 2023 Jun 22. doi: 10.1111/dmcn.15673. Online ahead of print.

Aim: To investigate spoken language comprehension (SLC), single-word comprehension (SWC), functional communication development, and their determinants, in children with cerebral palsy. Method: This was a prospective cohort study in the Netherlands spanning 2 years 6 months. The main outcomes were SLC and SWC, assessed by the Computer-Based instrument for Low motor Language Testing (C-BiLLT) and the Peabody Picture Vocabulary Test-III-NL (PPVT-III-NL) respectively; and functional communication, measured by a subscale of the Focus on the Outcomes of Communication Under Six-34 (FOCUS-34). Linear mixed models were used to determine developmental trajectories, which were compared with norm and reference data. Potential determinants, for example intellectual functions, speech production, functional communication level (classified with the Communication Function Classification System, CFCS), and functional mobility, were added to assess their effects. Results: Children with cerebral palsy (n = 188; mean age 59 months, range 17-110) were monitored for 2 years 6 months. Developmental trajectories for SLC (C-BiLLT) and SWC (PPVT-III-NL) were nonlinear; those for functional communication (FOCUS-34) were linear. Compared with norm and reference groups, significantly delayed SLC, SWC, and functional communication development were found. Determinants for SLC and SWC were intellectual functions and functional communication level (CFCS); and for functional communication development (FOCUS-34), speech production and arm-hand

functioning. Interpretation: Children with cerebral palsy showed delayed SLC, SWC, and functional communication development compared with norm and reference groups. Remarkably, functional mobility was not associated with the development of SLC, SWC, or functional communication.

PMID: [37349942](#)

22. Atypical clinical presentation and management of urinary stone disease in non-verbal non-ambulatory children

Nicolas Fernandez, Hailey Silverii, Pankaj Dangle, Karyn Yonekawa

J Pediatr Urol. 2023 Jun 7;S1477-5131(23)00228-0. doi: 10.1016/j.jpuro.2023.06.002. Online ahead of print.

Introduction: Management of kidney stones primarily depends on various factors such as the presence of urinary tract obstruction, pain, stone size, location, impact on renal function, and the existence of infection. Renal colic is the classic presentation of an obstructive kidney stone. However, in this study, we present a cohort of non-verbal non-ambulatory (NVNA) patients who exhibit a distinct and uncommon presentation of kidney stones due to their medical conditions. Information about kidney stone disease in this group of complex pediatric patients is scarce and their associated risk factors are not well understood. Therefore, we aim to summarize the clinical presentation, and management challenges in this unique group of NVNA patients to identify potential variables for prospective studies. **Methods:** A retrospective chart review was completed for all NVNA patients seen at the pediatric multidisciplinary kidney stone clinic between July 2020 to August 2022. Demographic variables, clinical presentation data, metabolic evaluation, radiological imaging, and surgical management was included for analysis. **Results:** A total of 224 pediatric patients were referred to the multidisciplinary stone clinic. Of those, 27 were identified to be NVNA. The most common primary diagnosis was Cerebral Palsy followed by Lennox-Gastaut syndrome. Average age at first kidney stone presentation was 11.5 years \pm 5.7 years. An obstructing stone was diagnosed in 18 (66%) patients, 4 (22.2%) of these presented with sepsis. Average stone burden was 9.2 mm (\pm 5.8 mm). Of the obstructing stones, 13 (72%) were in the kidney. All patients with an obstructing stone underwent surgical management with retrograde endoscopic approach. Metabolic 24-h-urine analysis was completed in 24 (89%) patients. 17 (62%) had an elevated urine density, 15 (55%) demonstrated calcium oxalate supersaturation, 12 (44%) met criteria for acidosis, and 7 (26%) had significant hypocitraturia. **Discussion:** NVNA patients represent a unique cohort whose clinical presentation is atypical. Limited ability to express symptoms makes early detection difficult to recognize. Twenty two percent of patients present with sepsis as their first manifestation of an acutely obstructing kidney stone. Pain is subjectively interpreted by caregivers and is an uncommon symptom. Our cohort demonstrates common risk factors for stones including propensity for chronic dehydration, slow urinary tract transit, ineffective bladder emptying, G-tube feeding, and lithogenic medications. **Conclusion:** NVNA pediatric patients have atypical kidney stone clinical presentation. Awareness of this unique group of patients should support future collaborative studies to focus on understanding these atypical presentations and reflect on improving management.

PMID: [37344296](#)

23. Psychometric properties of the caregiving difficulty scale in mothers of children with cerebral palsy

Eun-Young Park

BMC Neurol. 2023 Jun 20;23(1):237. doi: 10.1186/s12883-023-03264-w.

Background: The Caregiving Difficulty Scale is used to measure the burden of caregiving experienced by mothers of children with cerebral palsy. This study aimed to identify the psychometric properties of the Caregiving Difficulty Scale using the Rasch model. **Methods:** Data collected from 206 mothers of children with cerebral palsy were analyzed. Unidimensionality, difficulty of item, rating scale appropriateness, and reliability using the separation index of the Caregiving Difficulty Scale were verified. Unidimensionality of all 25 items was identified through the item fit. **Results:** Our analysis of item difficulty indicated that person ability and item difficulty are expressed as a similar logit extend. The use of the 5-point rating scale appeared to be appropriate. Outcome analysis revealed that the reliability was high based on the person and that the item separation level was acceptable. **Conclusions:** This study showed that the Caregiving Difficulty Scale could be a valuable tool for evaluating the caregiving burden in mothers of children with cerebral palsy.

PMID: [37340392](#)

24. RNA sequencing reveals a complete picture of a homozygous missense variant in a patient with VPS13D movement disorder: a case report and review of the literature

Elizabeth K Baker, Jingfen Han, William A Langley, Michael A Reott Jr, Barbara E Hallinan, Robert J Hopkin, Wenying Zhang

Mol Genet Genomics. 2023 Jun 20. doi: 10.1007/s00438-023-02044-y. Online ahead of print.

RNA sequencing (RNA-seq) is a complementary diagnostic tool to exome sequencing (ES), only recently clinically available to undiagnosed patients post-ES, that provides functional information on variants of unknown significance (VUS) by evaluating its effect on RNA transcription. ES became clinically available in the early 2010s and promised an agnostic platform for patients with a neurological disease, especially for those who believed to have a genetic etiology. However, the massive data generated by ES pose challenges in variant interpretation, especially for rare missense, synonymous, and deep intronic variants that may have a splicing effect. Without functional study and/or family segregation analysis, these rare variants would be likely interpreted as VUS which is difficult for clinicians to use in clinical care. Clinicians are able to assess the VUS for phenotypic overlap, but this additional information alone is usually not enough to re-classify a variant. Here, we report a case of a 14-month-old male who presented to clinic with a history of seizures, nystagmus, cerebral palsy, oral aversion, global developmental delay, and poor weight gain requiring gastric tube placement. ES revealed a previously unreported homozygous missense VUS, c.7406A > G p.(Asn2469Ser), in VPS13D. This variant has not been previously reported in genome aggregation database (gnomAD), ClinVar, or in any peer-reviewed published literature. By RNA-seq, we demonstrated that this variant mainly impacts splicing and results in a frameshift and early termination. It is expected to generate either a truncated protein, p.(Val2468fs*19), or no protein from this transcript due to nonsense-mediated mRNA decay leading to VPS13D deficiency. To our knowledge, this is the first case utilizing RNA-seq to further functionally characterize a homozygous novel missense VUS in VPS13D and confirm its impact on splicing. This confirmed pathogenicity gave the diagnosis of VPS13D movement disorder to this patient. Therefore, clinicians should consider utilizing RNA-seq to clarify VUS by evaluating its effect on RNA transcription.

PMID: [37340120](#)

25. Efficacy of videogames and exergames in pediatric neurorehabilitation: a systematic review

Agata Polizzi, Sergio Rinella, Martino Ruggieri, Amalia E Gentile, Cristiano M Verrelli, Marco Iosa

Minerva Pediatr (Torino). 2023 Jun 16. doi: 10.23736/S2724-5276.23.07146-X. Online ahead of print.

Introduction: In recent years, the rehabilitation of children with neurological disorders has taken into account the possibility of using videogaming consoles and virtual reality systems to make children's therapy more enjoyable, motivating, participated and effective. This study aims at conducting a systematic review about the use and the efficacy of digital games in pediatric neurorehabilitation. **Evidence acquisition:** In accordance with the PRISMA approach, a rather wide-ranging search was conducted on PubMed, Scopus, and Web of Science databases by using different combinations of keywords based on MeSH terms. **Evidence synthesis:** Fifty-five papers have been included into this review, namely, 38 original studies and 17 reviews. The total number of children and adolescents is 573, with 58% of them being affected by cerebral palsy. Despite a wide variability in the adopted protocols, devices, assessment tools, and a more frequent focus on motor aspects than on cognitive ones, the results of the majority of the analyzed studies support the safety (i.e., absence of severe adverse effects) and efficacy of the videogame-based therapy. **Conclusions:** Videogames, when administered by means of commercial consoles or ad-hoc digital systems, seem to be a valid support for physical therapy. Further researchers are needed to deeply investigate the role of this approach in cognitive therapy and cognitive outcomes.

PMID: [37335184](#)