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Interventions and Management

1. Improvements in Upper Extremity Function Following Intensive Training Are Independent of Corticospinal Tract Organization in Children With Unilateral Spastic Cerebral Palsy: A Clinical Randomized Trial

Kathleen M Friel, Claudio L Ferre, Marina Brandao, Hsing-Ching Kuo, Karen Chin, Ya-Ching Hung, Maxime T Robert, Veronique H Flamand, Ana Smorenburg, Yannick Bleyenheuft, Jason B Carmel, Talita Campos, Andrew M Gordon

Front Neurol. 2021 May 3;12:660780. doi: 10.3389/fneur.2021.660780. eCollection 2021.

Background/Objectives: Intensive training of the more affected upper extremity (UE) has been shown to be effective for children with unilateral spastic cerebral palsy (USCP). Two types of UE training have been particularly successful: Constraint-Induced Movement Therapy (CIMT) and Bimanual training. Reorganization of the corticospinal tract (CST) early during development often occurs in USCP. Prior studies have suggested that children with an ipsilateral CST controlling the affected UE may improve less following CIMT than children with a contralateral CST. We tested the hypothesis that improvements in UE function after intensive training depend on CST laterality. **Study Participants and Setting:** Eighty-two children with USCP, age 5 years 10 months to 17 years, University laboratory setting. **Materials/Methods:** Single-pulse transcranial magnetic stimulation (TMS) was used to determine each child's CST connectivity pattern. Children were stratified by age, sex, baseline hand function and CST connectivity pattern, and randomized to receive either CIMT or Bimanual training, each of which were provided in a day-camp setting (90 h). Hand function was tested before, immediately and 6 months after the intervention with the Jebsen-Taylor Test of Hand Function, the Assisting Hand Assessment, the Box and Block Test, and ABILHAND-Kids. The Canadian Occupational Performance Measure was used to track goal achievement and the Pediatric Evaluation of Disability Inventory was used to assess functioning in daily living activities at home. **Results:** In contrast to our hypothesis, participants had statistically similar improvements for both CIMT and Bimanual training for all measures independent of their CST connectivity pattern (contralateral, ipsilateral, or bilateral) ($p < 0.05$ in all cases). **Conclusions/Significance:** The efficacy of CIMT and Bimanual training is independent of CST connectivity pattern. Children with an ipsilateral CST, previously thought to be maladaptive, have the capacity to improve as well as children with a contralateral or bilateral CST following intensive CIMT or Bimanual training. Clinical Trial Registration: www.ClinicalTrials.gov, identifier NCT02918890.

PMID: [34012418](https://pubmed.ncbi.nlm.nih.gov/34012418/)

2. The Neurological Hand Deformity Classification: Construct validity, test-retest, and inter-rater reliability

Simon Garbellini, Melinda Randall, Michael Steele, Catherine Elliott, Christine Imms

J Hand Ther. 2021 Mar 29;S0894-1130(21)00045-4. doi: 10.1016/j.jht.2021.03.005. Online ahead of print.

Background: The Neurological Hand Deformity Classification (NHDC) is an impairment-based tool that classifies hand deformity into one of two ordinal scales: flexion or extension deformities. Classification is made from live observation or from recorded video footage. Differentiation between the levels is determined by wrist position and wrist and finger

movement. Purpose: To examine aspects of validity and reliability of the NHDC. Study design: A measurement study design. Methods: Data from a convenience sample of 127 children with cerebral palsy, 66 males; 61 females, ranging in age from 8 months to 15 years, across all Manual Ability Classification System levels I to V, were analyzed. Construct validity was assessed by testing predetermined hypotheses of relationships between the NHDC and measures of body function and activity measures with observed performance using the Chi Squared Test of Independence and Spearman Correlation Coefficient. Test-retest and inter-rater reliability were assessed by calculating agreement between repeated measures and paired raters using weighted kappa and Cohen's kappa with 95% confidence intervals. Results: Predicted hypotheses for the NHDC were met in nine of 10 Spearman's rho correlations with body structure measures and in 2 of 7 correlations with activity measures. Test-retest for flexion deformities: $\kappa_w = 0.84$; 95% CI 0.70-0.98; and extension deformities: $\kappa = 1.0$; 95% CI 1.0-1.0 was good to excellent; inter-rater reliability for flexion deformities: $\kappa_w = 0.76$; 95% CI 0.67-0.85; and extension deformities $\kappa = 0.75$; 95% CI 0.43-1.0 was moderate to excellent. Conclusion: Expected relationships between the NHDC and other measures, stability between repeated measures and acceptable between-rater agreement supports confidence classifying hand deformity in children with cerebral palsy with the NHDC.

PMID: [34011469](#)

3. Comparative cost-utility analysis of postoperative discharge pathways following posterior spinal fusion for scoliosis in non-ambulatory cerebral palsy patients

K Aaron Shaw, Vahé Heboyan, Nicholas D Fletcher, Joshua S Murphy

Spine Deform. 2021 May 18. doi: 10.1007/s43390-021-00362-y. Online ahead of print.

Purpose: Accelerated postoperative discharge (AD) pathways have demonstrated numerous benefits for patients with adolescent idiopathic scoliosis undergoing PSF. Although early evidence supports the application of AD pathways over more traditional discharge (TD) approaches for patients with neuromuscular scoliosis, the economic impact of these pathways has not been investigated. Methods: A decision-analysis model was constructed using a hypothetical 15-year-old male with non-ambulatory CP with a 65-degree thoracolumbar scoliosis and pelvic obliquity undergoing operative treatment with PSF from T2-pelvis with pedicle screw fixation. The literature was reviewed to estimate costs, probabilities, and quality-adjusted life years (QALYs) for identified complication profiles for discharge pathways. QALYs were constructed using age-matched values for US population average, applying a CP diagnosis corrective value. A probabilistic sensitivity analysis was performed using a second-order Monte Carlo simulations. Incremental cost-utility ratio and incremental net monetary benefit (NMB) were calculated. One-way sensitivity analyses were performed by selective variable variation. Results: AD pathway resulted in an average cost and effectiveness of \$67,069 and 15.4 QALYs compared with \$81,312 and 15.4 QALYs for TD. AD resulted in a 2.1% greater NMB with a cost-effectiveness ratio of \$4361/QALY compared with \$5290/QALY in the TD. The cost-effectiveness of TD was inversely sensitive to implant cost variation while the AD maintained effectiveness despite cost variations. Conclusion: This cost-utility analysis demonstrated that the implementation of an AD pathway following PSF for non-ambulatory CP scoliosis is economically more effective, providing a 17.5% cost reduction with enhanced value of care evidenced by a 2.1% greater NMB over a TD pathway. The cost-effectiveness of the AD was maintained despite implant cost variations.

PMID: [34008146](#)

4. Quantitative magnetic resonance imaging of spinal cord microstructure in adults with cerebral palsy

Julien Cohen-Adad

Dev Med Child Neurol. 2021 May 15. doi: 10.1111/dmcn.14927. Online ahead of print.

PMID: [33990941](#)

5. Facilitating new movement strategies: Equine assisted physiotherapy for children with cerebral palsy

Brita Cecilie Norrud, Målfrid Råheim, Tobba Therkildsen Sudmann, Margareta Håkanson

J Bodyw Mov Ther. 2021 Apr;26:364-373. doi: 10.1016/j.jbmt.2020.12.022. Epub 2021 Feb 10.

Background: Equine Assisted Physiotherapy (EAPT) offers children with cerebral palsy (CP) opportunities for new movement

experiences, and may influence movement qualities. Descriptions of how, and to what extent EAPT affects trunk control is missing. The aim of this study was to explore if, and how changes in trunk control and changes in other movement aspects were observable in children with CP during EAPT, and if potential changes in trunk control could be measured. Method: A multiple case study with a mixed methods design was completed. Two children with CP, GMFCS grade I, were observed using video during a period of six months, and tested with Trunk Impairment Scale modified Norwegian Version. Skilled physiotherapists analyzed the videos qualitatively, and triangulated recurring changes in movement with the results from the test. Results: Riding bareback, improvements in trunk control were observed and measured. However, riding in a saddle led to reduced trunk control. Other observable movement changes were: from asymmetry to symmetry, adaptation to rhythm, mastery of riding skills, and reduced loss of postural control. Increased instances of adapting own movements in spontaneous dialogue with the horse, were observed. Instructions and feedback from the therapist influenced the dialogue with the horse both positively and negatively. Conclusion: This study describes in detail how balance and symmetry can be stimulated during EAPT in a body characterized by imbalance and asymmetry. During EAPT, the children gained the possibility to explore new movement qualities. Equipment and feedback influenced movement qualities.

PMID: [33992271](#)

6. Variability in Postoperative Immobilization and Rehabilitation Following Reconstructive Hip Surgery in Nonambulatory Children With Cerebral Palsy

Stacey D Miller, Maria Juricic, Nandy Fajardo, Judy So, Benjamin J Shore, Unni G Narayanan, Kishore Mulpuri

J Pediatr Orthop. 2021 May 13. doi: 10.1097/BPO.0000000000001850. Online ahead of print.

Background: Despite being a common procedure, there are no standard protocols for postoperative immobilization and rehabilitation following reconstructive hip surgery in children with cerebral palsy (CP). The purpose of this study was to investigate variability in postoperative management and physical therapy (PT) recommendations among orthopaedic surgeons treating hip displacement in children with CP. Methods: An invitation to participate in an anonymous, online survey was sent to 44 pediatric orthopaedic surgeons. The case of a child undergoing bilateral femoral varus derotation osteotomies and adductor tenotomies was presented. Surgeons were asked to consider their typical practice and the case scenario when answering questions related to immobilization, weight-bearing, and rehabilitation. Recommendations with increasingly complex surgical interventions and different age or level of motor function were also assessed. Results: Twenty-eight orthopaedic surgeons from 9 countries with a mean 21.3 years (range: 5 to 40 y) of experience completed the survey. Postoperative immobilization was recommended by 86% (24/28) of respondents with 7 different methods of immobilization identified. All but 1 (23/24) reported immobilizing full time. Most (20/23) reported using immobilization for 4 to 6 weeks. Return to weight-bearing varied from 0 to 6 weeks for partial weight-bearing and 0 to 12 weeks for full weight-bearing. PT in the first 1 to 2 weeks postoperatively was reported as unnecessary by 29% (8/28) of surgeons. PT for range of motion, strengthening, and return to function was recommended by 96% (27/28) of surgeons, starting at a mean of 2.6 weeks postoperatively (range: 0 to 16 wk). Only 48% (13/27) reported all of their patients would receive PT for these goals in their practice setting. Inpatient rehabilitation was available for 75% (21/28) but most surgeons (17/21) reported this was accessed by 20% or fewer of their patients. Conclusions: Postoperative immobilization and PT recommendations were highly variable among surgeons. This variability may influence surgical outcomes and complication rates and should be considered when evaluating procedures. Further study into the impact of postoperative immobilization and rehabilitation is warranted.

PMID: [33999564](#)

7. Clinical gait analysis and physical examination don't correlate with physical activity of children with cerebral palsy. Cross-sectional study

Anne-Laure Guinet, Khouri Néjib, Desailly Eric

Int Biomech. 2020 Dec;7(1):88-96. doi: 10.1080/23335432.2020.1812429.

Gait analysis and physical clinical measures are usually performed in children with cerebral palsy to help the surgeons make therapeutic decision. However, the level of physical activity in daily life is not systematically assessed. The aim of this cross sectional study was to examine the correlations between: three-dimensional gait analysis kinematic and spatiotemporal parameters, clinical measures and physical activity. Participants were 30 children with cerebral palsy (10-18 y), with GMFCS I-III. Daily physical activity was measured with an Actigraph GT3X accelerometer in free living environment during seven consecutive days. The percent of time spent in sedentary, in moderate to vigorous physical activity and the number of steps per day were computed from the accelerometer data. Kinematics parameters did not correlate with physical activity. Moderate correlations were found between spatio-temporal parameters and physical activity, for instance timing of toe-off ($r = -0.40$, $p =$

0.03). Few physical examination parameters were correlated with physical activity, such as the hip flexors selective motor control ($r = 0.69$ with moderate to vigorous activity and $r = 0.70$ with steps per day, $p < 0.05$). The physical activity profile cannot be sufficiently determined by a combination of clinical measures.

PMID: [33998383](#)

8. The effect of dual-task conditions on gait and balance performance in children with cerebral palsy: A systematic review and meta-analysis of observational studies

Meysam Roostaei, Parvin Raji, Giovanni Morone, Bahman Razi, Khosro Khademi-Kalantari

Review J Bodyw Mov Ther. 2021 Apr;26:448-462. doi: 10.1016/j.jbmt.2020.12.011. Epub 2020 Dec 9.

Dual-task conditions are commonly experienced in daily routines. The aim of the present systematic review is to investigate the effect of dual-task conditions on gait and balance performance in children with cerebral palsy (CP) and to perform meta-analyses where applicable. Five databases, "ProQuest", "PubMed", "OTSeeker", "Scopus", and "PEDro" from the incipient date of databases up to Aug 24, 2020 were searched for studies focusing on the effects of dual-task conditions on gait and balance performance in children with CP. After removing irrelevant articles and applying inclusion and exclusion criteria, nine articles were included in the present systematic review and meta-analysis. The results of the meta-analysis showed that walking speed was slower during dual-task conditions compared to single-task conditions in children with CP (WMD = -0.29 m/s, 95% CI = -0.34, -0.24, $P \leq 0.001$) and walking speed decreased in children with CP during dual-task conditions in comparison with the typical development (TD) control group (WMD = -0.19 m/s, 95% CI = -0.23 to -0.15, $P \leq 0.001$). The results of subgroup analysis based on the type of task indicated that adding concurrent tasks to walking degrades walking speed under varied dual-task conditions. Additionally, theoretical synthesis of the literature demonstrated that other gait and balance variables are changed by performing cognitive and motor secondary tasks differently. Although these changes may be compensatory strategies to retain their stability, there was not sufficient evidence to reach a firm conclusion. Research gaps and recommendations for future studies are discussed.

PMID: [33992282](#)

9. Surgery versus botulinum neurotoxin A to reduce drooling and improve daily life for children with neurodevelopmental disabilities: a randomized controlled trial

Stijn Bekkers, Ineke M J Pruijn, Jan J W van der Burg, Karen van Hulst, Saskia E Kok, Corinne P Delsing, Arthur R T Scheffer, Frank J A van den Hoogen

Dev Med Child Neurol. 2021 May 16. doi: 10.1111/dmcn.14924. Online ahead of print.

Aim: To compare the effect of bilateral submandibular duct ligation and botulinum neurotoxin A (BoNT-A) on drooling severity and its impact on daily life and care in children and adolescents with moderate-to-severe drooling. **Method:** This was a randomized, interventional, controlled trial in which 53 children and adolescents (31 males, 22 females, mean age 11y, range 8-22y, SD 2y 10mo) with cerebral palsy (58.5%) or other non-progressive developmental disorders (41.5%) were randomized to BoNT-A ($n=26$) or bilateral submandibular duct ligation ($n=27$). A parent questionnaire on the severity of drooling in specific positions and daily activities and the impact of drooling on daily life and care was filled out at baseline and 8 and 32 weeks posttreatment. **Results:** Both BoNT-A and bilateral submandibular duct ligation had a positive effect on daily care, damage to electronic equipment and/or furniture, social interactions, and self-esteem. However, bilateral submandibular duct ligation had a significant greater and longer-lasting short- (8wks) and medium-term (32wks) effect on daily care, reducing damage to electronic devices, and improving social interactions and satisfaction with life in general. **Interpretation:** This randomized controlled trial confirms reduced drooling by both BoNT-A and bilateral submandibular duct ligation, but provides new evidence on improved well-being through a reduction in drooling. Even though there is a greater risk of complications and morbidity after bilateral submandibular duct ligation, compared to BoNT-A there was a significantly greater and longer-lasting positive effect on most outcomes.

PMID: [33997959](#)

10. Efforts targeted malnutrition among children with cerebral palsy in care homes and hospitals- a qualitative exploration study

Nanna Marie Hedegaard Rosendahl, Rikke Caroline Jensen, Mette Holst

J Hum Nutr Diet. 2021 May 15. doi: 10.1111/jhn.12916. Online ahead of print.

Introduction: Children with cerebral palsy (CP) are at risk of becoming malnourished. Malnutrition risk increases with increased Gross Motor Function Classification. This study aimed to investigate and explore existing nutritional efforts and practice among staff in Danish hospitals and care homes working with children with CP. **Methods:** Ten qualitative interviews were generated with multidisciplinary staff working in hospital wards, care homes and neuropaediatric teams(NPT). Analysis was inspired by Braun and Clarke's thematic analysis. **Results:** The analysis generated the following six themes: Weighing and anthropometric measurements; Existing nutritional evaluation and intervention in practice; Cross-sectoral communication and interdisciplinarity; Involvement of the children's parents; Knowledge and professional intuition; Time and priority. Weighing is the most common nutritional evaluation, supplemented by weight/height curves in the NPT. No actual screening is done. In care homes without healthcare staff nutritional focus primarily regards the meal and how the children are able to eat. In care homes with health staff as well as hospital departments, the focus is primarily on weight. Written communication is a necessity for the nutritional collaboration around the child to work, but collaboration between IT-systems are lacking. Interdisciplinarity and parental involvement are key components for managing nutrition issues among children with CP. The specialist dietician was much required, but difficult to reach. **Conclusion:** Despite the great focus on nutrition and nutritional status among children with CP in all of the inquired contexts, existing efforts for assessment reveal primarily on weight and intuition. No actual screening is done. Collaboration between sectors and professionals is challenged by lacking IT-systems and systematic procedures.

PMID: [33991366](#)

11. Sleep problems in children with cerebral palsy and their parents

Raquel Y Hulst, Jan Willem Gorter, Jeanine M Voorman, Eveline Kolk, Sanne Van Der Vossen, Johanna M A Visser-Meily, Marjolijn Ketelaar, Sigrid Pillen, Olaf Verschuren

Dev Med Child Neurol. 2021 May 15. doi: 10.1111/dmcn.14920. Online ahead of print.

Aim: To describe: (1) the frequency and types of sleep problems, (2) parent-rated satisfaction with their child's and their own sleep, and (3) child factors related to the occurrence of sleep problems in children with cerebral palsy (CP) and their parents. The secondary objective was to compare the sleep outcomes of children with CP with those from typically developing children and their parents. **Method:** The Sleep section of the 24-hour activity checklist was used to assess the sleep of children with CP and their parents and the sleep of typically developing children and their parents. **Results:** The sleep outcomes of 90 children with CP (median age 5y, range 0-11y, 53 males, 37 females, 84.4% ambulatory) and 157 typically developing peers (median age 5y, range 0-12y; 79 males, 78 females) and their parents were collected. Children with CP were more likely to have a sleep problem than typically developing children. Non-ambulatory children with CP were more severely affected by sleep problems than ambulatory children. The parents of non-ambulatory children were less satisfied about their child's and their own sleep. Waking up during the night, pain/discomfort in bed, and daytime fatigue were more common in children with CP and more prevalent in children who were non-ambulatory. **Interpretation:** These findings highlight the need to integrate sleep assessment into routine paediatric health care practice.

PMID: [33990937](#)

12. Psychological, cardiometabolic, musculoskeletal morbidity and multimorbidity among adults with cerebral palsy and spina bifida: a retrospective cross-sectional study

Mark D Peterson, Paul Lin, Neil Kamdar, Edward A Hurvitz, Elham Mahmoudi

Am J Phys Med Rehabil. 2021 May 17. doi: 10.1097/PHM.0000000000001787. Online ahead of print.

Background: Individuals living with cerebral palsy (CP) or spina bifida (SB) are at heightened risk for a number of chronic health conditions such as secondary comorbidities, that may develop or be influenced by the disability, the presence of impairment, and/or the process of aging. However, very little is known about the prevalence and/or risk of developing secondary-comorbidities among individuals living with CP or SB throughout adulthood. The objective of this study was to compare the prevalence of psychological, cardiometabolic, musculoskeletal morbidity, and multimorbidity among adults with and without CP or SB. **Methods:** Privately-insured beneficiaries were included if they had an ICD-9-CM diagnostic code for CP or SB (n = 29,841). Adults without CP or SB were also included (n = 5,384,849). Prevalence estimates of common

psychological, cardiometabolic, and musculoskeletal morbidity and multimorbidity (≥ 2 conditions) were compared. Results: Adults living with CP or SB had a higher prevalence of all psychological disorders and psychological multimorbidity (14.6% vs 5.4%), all cardiometabolic disorders and cardiometabolic multimorbidity (22.4% vs. 15.0%), and all musculoskeletal disorders and musculoskeletal multimorbidity (12.2% vs. 5.4%), as compared to adults without CP or SB, and differences were to a clinically meaningful extent. Conclusions: Adults with CP or SB have a significantly higher prevalence of common psychological, cardiometabolic, and musculoskeletal morbidity and multimorbidity, as compared to adults without CP or SB. Efforts are needed to facilitate the development of improved clinical screening algorithms and early interventions to reduce risk of disease onset/progression in these higher risk populations.

PMID: [34001837](#)

13. Critical periods of bone health across the lifespan for individuals with cerebral palsy: Informing clinical guidelines for fracture prevention and monitoring

Daniel G Whitney, Edward A Hurvitz, Michelle S Caird

Bone. 2021 May 18;150:116009. doi: 10.1016/j.bone.2021.116009. Online ahead of print.

Background: Skeletal fragility is a major burden for individuals with cerebral palsy (CP), but little is known clinically about when to prevent fractures or monitor bone health for this population. Critical periods of bone health (CPBH) are important windows for intervention to augment bone growth or mitigate bone loss. However, CPBH from the general population may not align with the needs or timing of skeletal fragility for individuals with CP. The objective of this study was to identify discrepancies when evaluating individuals with CP using CPBH across the lifespan from the general population, and propose new CP-specific CPBH. Methods: Data from 2016 administrative claims databases were used, including the Optum's De-identified Clinformatics® Data Mart Database and a random 20% sample of the Medicare fee-for-service database from the Centers for Medicare and Medicaid Services. Sex-stratified fracture prevalence was compared between individuals with and without CP across the lifespan starting at 2 years of age using age groups to capture important stages of development and 3-4-year age bands in adulthood (up to >80 years). Sex-specific CPBH from the general population included: rapid bone accrual, peak bone mass, menopause, and elderly. Results: There were 23,861 individuals with CP and 9,976,161 individuals without CP. CPBH from the general population did not align with the timing of skeletal fragility for CP. For example, fractures were rare and decreased throughout the CPBH of peak bone mass for males without CP, but males with CP had a greater relative fracture risk (2.9-5.6-fold higher) and a substantially increased rate of fracture (CP-slope 14× higher than non-CP-slope). For females with CP, fracture risk was increased by 18-21 years, with additional inflection points (e.g., decade before menopause and again by 57-60 years). For males with CP, fracture risk in mid-life exhibited a pattern similar to elderly males without CP. Conclusions: This study identified discrepancies in evaluating fracture risk for individuals with CP if using established CPBH from the general population. We therefore propose new CP- and sex-specific CPBH for fracture monitoring and prevention.

PMID: [34020079](#)

14. Akwenda intervention programme for children and youth with cerebral palsy in a low-resource setting in sub-Saharan Africa: protocol for a quasi-randomised controlled study

Gillian Saloojee, Francis Ekwana, Carin Andrews, Diane L Damiano, Angelina Kakooza-Mwesige, Hans Forsberg

BMJ Open. 2021 Mar 8;11(3):e047634. doi: 10.1136/bmjopen-2020-047634.

Introduction: Cerebral palsy (CP) is the most common childhood-onset motor disorder accompanied by associated impairments, placing a heavy burden on families and health systems. Most children with CP live in low/middle-income countries with little access to rehabilitation services. This study will evaluate the Akwenda CP programme, a multidimensional intervention designed for low-resource settings and aiming at improving: (1) participation, motor function and daily activities for children with CP; (2) quality of life, stress and knowledge for caregivers; and (3) knowledge and attitudes towards children with CP in the communities. Methods: This quasi-randomised controlled clinical study will recruit children and youth with CP aged 2-23 years in a rural area of Uganda. Children will be allocated to one of two groups with at least 44 children in each group. Groups will be matched for age, sex and motor impairment. The intervention arm will receive a comprehensive, multidimensional programme over a period of 11 months comprising (1) caregiver-led training workshops, (2) therapist-led practical group sessions, (3) provision of technical assistive devices, (4) goal-directed training and (5) community communication and advocacy. The other group will receive usual care. The outcome of the intervention will be assessed before and after the intervention and will be measured at three levels: (1) child, (2) caregiver and (3) community. Standard analysis methods for randomised controlled trial will be used to compare groups. Retention of effects will be examined at 12-month follow-up. Ethics and dissemination: The study has been approved by the Uganda National Council for Science and

Technology (SS 5173) and registered in accordance with WHO and ICMJE standards. Written informed consent will be obtained from caregivers. Results will be disseminated among participants and stakeholders through public engagement events, scientific reports and conference presentations. Trial registration number: Pan African Clinical Trials Registry (PACTR202011738099314) Pre-results.

PMID: [34006038](#)

15. Mothers' perception of cerebral palsy in a low-income country of West Africa: a cross-sectional study

Emmanuel Segnon Sogbossi, Damienne Houekpetodji, Toussaint G Kpadonou, Yannick Bleyenheuft

Disabil Rehabil. 2021 May 15;1-8. doi: 10.1080/09638288.2021.1919765. Online ahead of print.

Purpose: Investigating mothers' perceptions of Cerebral Palsy (CP) and the impact of caregiving on family life in Benin, a west-African low to middle-income country (LMIC). **Methods:** In this cross-sectional study we interviewed at their homes 88 mothers of children with CP about their perception of the cause of CP and stigmatization issues. Additionally, they completed the Impact on Family Scale and the Family Support Scale questionnaires. Multinomial and ordinal regressions analyses were used ($p < 0.05$). **Results:** Most mothers (78%) referred to non-biomedical causes of CP (God's will or curse). Caregivers with a limited education level were more likely to report non-biomedical beliefs. Only 28% reported having received from physicians an explanation of the causes of CP. Around 38% endorsed neighbors considered their children as cursed, which is a source of stigmatization. Mothers perceived a high impact of caregiving on their family life that correlated with non-biomedical beliefs, low education level, and the severity of CP. **Conclusion:** There is a need in Benin, and probably in West-African LMICs generally, to shift to a family-centered care system and to inform the public about the root causes of CP to improve social integration of children with CP and decrease the negative impact on their families. **Implication for rehabilitation:** Mothers with a limited education level are more likely to report non-biomedical causes of CP in this LMIC; mothers' level of education and non-biomedical beliefs of causes of CP are associated to a high impact of caregiving on family life in this LMIC; mothers of children with CP in this LMIC do not perceive health-workers helpful.

PMID: [33993806](#)

16. Clinical Profile and Outcome of Indian Children with Aromatic L-Amino Acid Decarboxylase Deficiency: A primary CSF Neurotransmitter Disorder Mimicking as Dyskinetic Cerebral Palsy

Vykuntaraju K Gowda, Hemadri Vegda, Balamurugan B Nagarajan, Sanjay K Shivappa

J Pediatr Genet. 2021 Jun;10(2):85-91. doi: 10.1055/s-0040-1714690. Epub 2020 Jul 27.

Aromatic L-amino acid decarboxylase (AADC) deficiency is a disorder of neurotransmitter synthesis. It presents with psychomotor delay, dystonia, oculogyric crisis, and autonomic features. There is paucity of literature on this disorder. Hence, we are reporting this series with an objective to study profile and outcome of Indian children with AADC deficiency. In this retrospective review, all case records of genetically confirmed cases of AADC deficiency at the pediatric neurology department in a tertiary care hospital, from March 2014 to March 2020, were analyzed. The data were extracted in a predesigned proforma and analyzed. Out of seven cases, five were males. Median age of onset of symptoms was 4 months but median age of diagnosis was 12 months. All of them had developmental delay, oculogyric crisis, dystonia, increased sweating, intermittent fever, feeding and sleep disturbance, irritability, failure to thrive, axial hypotonia with dyskinetic quadriparesis, and normal magnetic resonance imaging (MRI) of brain and electroencephalogram (EEG). All of them were treated with pyridoxal 5-phosphate, trihexyphenidyl and pramipexole and six cases, in addition, were given bromocriptine. One case was additionally treated with selegiline. One case showed good improvement, five showed partial improvement, and one case expired. In conclusion, AADC deficiency should be suspected in any child with dyskinetic quadriparesis, oculogyric crisis, autonomic disturbances like increased sweating, intermittent fever, and sleep disturbance with normal neuroimaging.

PMID: [33996177](#)

17. An etiological study of intellectually disabled children under 14 years old in Anhui Province, China

Sinan Li, Guanglei Tong

Am J Transl Res. 2021 Apr 15;13(4):2670-2677. eCollection 2021.

Objective: To explore the etiological factors of intellectually disabled children in Anhui Province using a multicenter etiological study. **Methods:** A total of 200 children aged 0 to 14 years in Anhui Province who were diagnosed with intellectual disabilities were recruited as the study cohort. Their general information (perinatal information, parental educational levels, family environments, etc.) was collected through questionnaires, and the Gesell Developmental Scale and the Wechsler Intelligence Scale were used to assess the intelligence development of the enrolled children. **Results:** Among the 528 children, 270 (51.14%) had severe intellectual disabilities and 258 (48.86%) had mild intellectual disabilities. It was found that various perinatal factors (premature birth, asphyxia, ischemic hypoxic encephalopathy, etc.), severe cerebral palsy, and psychosocial factors were the main etiological factors, accounting for 27.42%, 22.29%, and 17.16% respectively. There was a significant difference in the distribution of the etiologies between the rural and urban areas ($P < 0.01$). The educational levels of most of the parents in the rural areas were lower than the parents' educational levels in the cities. **Conclusion:** Correlation analyses are helpful for the early diagnosis of children suspected of having intellectual disabilities and they provide a scientific basis for improving the children's quality of life and their early rehabilitation treatment.

PMID: [34017427](#)

18. Early Intervention for Children Aged 0 to 2 Years With or at High Risk of Cerebral Palsy: International Clinical Practice Guideline Based on Systematic Reviews

Catherine Morgan, Linda Fetters, Lars Adde, Nadia Badawi, Ada Bancale, Roslyn N Boyd, Olena Chorna, Giovanni Cioni, Diane L Damiano, Johanna Darrah, Linda S de Vries, Stacey Dusing, Christa Einspieler, Ann-Christin Eliasson, Donna Ferriero, Darcy Fehlings, Hans Forssberg, Andrew M Gordon, Susan Greaves, Andrea Guzzetta, Mijna Hadders-Algra, Regina Harbourne, Petra Karlsson, Lena Krumlinde-Sundholm, Beatrice Latal, Alison Loughran-Fowlds, Catherine Mak, Nathalie Maitre, Sarah McIntyre, Cristina Mei, Angela Morgan, Angelina Kakooza-Mwesige, Domenico M Romeo, Katherine Sanchez, Alicia Spittle, Roberta Shepherd, Marelle Thornton, Jane Valentine, Roslyn Ward, Koa Whittingham, Alieh Zamany, Iona Novak

JAMA Pediatr. 2021 May 17. doi: 10.1001/jamapediatrics.2021.0878. Online ahead of print.

Importance: Cerebral palsy (CP) is the most common childhood physical disability. Early intervention for children younger than 2 years with or at risk of CP is critical. Now that an evidence-based guideline for early accurate diagnosis of CP exists, there is a need to summarize effective, CP-specific early intervention and conduct new trials that harness plasticity to improve function and increase participation. Our recommendations apply primarily to children at high risk of CP or with a diagnosis of CP, aged 0 to 2 years. **Objective:** To systematically review the best available evidence about CP-specific early interventions across 9 domains promoting motor function, cognitive skills, communication, eating and drinking, vision, sleep, managing muscle tone, musculoskeletal health, and parental support. **Evidence review:** The literature was systematically searched for the best available evidence for intervention for children aged 0 to 2 years at high risk of or with CP. Databases included CINAHL, Cochrane, Embase, MEDLINE, PsycInfo, and Scopus. Systematic reviews and randomized clinical trials (RCTs) were appraised by A Measurement Tool to Assess Systematic Reviews (AMSTAR) or Cochrane Risk of Bias tools. Recommendations were formed using the Grading of Recommendations Assessment, Development, and Evaluation (GRADE) framework and reported according to the Appraisal of Guidelines, Research, and Evaluation (AGREE) II instrument. **Findings:** Sixteen systematic reviews and 27 RCTs met inclusion criteria. Quality varied. Three best-practice principles were supported for the 9 domains: (1) immediate referral for intervention after a diagnosis of high risk of CP, (2) building parental capacity for attachment, and (3) parental goal-setting at the commencement of intervention. Twenty-eight recommendations (24 for and 4 against) specific to the 9 domains are supported with key evidence: motor function (4 recommendations), cognitive skills (2), communication (7), eating and drinking (2), vision (4), sleep (7), tone (1), musculoskeletal health (2), and parent support (5). **Conclusions and relevance:** When a child meets the criteria of high risk of CP, intervention should start as soon as possible. Parents want an early diagnosis and treatment and support implementation as soon as possible. Early intervention builds on a critical developmental time for plasticity of developing systems. Referrals for intervention across the 9 domains should be specific as per recommendations in this guideline.

PMID: [33999106](#)

19. Early Intervention in Cerebral Palsy and Beyond

Abdellah Tebani, Stéphane Marret

JAMA Pediatr. 2021 May 17. doi: 10.1001/jamapediatrics.2021.0884. Online ahead of print.

PMID: [33999172](#)

20. Children and Youth with Complex Cerebral Palsy: Care and Management

Cassandra Conrad, Elizabeth Harstad

J Dev Behav Pediatr. 2020 Aug 1;41(6):485. doi: 10.1097/DBP.0000000000000811.

PMID: [34008536](#)**21. Children with Cerebral Palsy and Unmet Need for Care Coordination**

Myriam Casseus, JenFu Cheng

J Dev Behav Pediatr. 2021 May 12. doi: 10.1097/DBP.0000000000000950. Online ahead of print.

Objective: To examine the physical and functional health of children with cerebral palsy (CP) and determine the prevalence and correlates of unmet need for health care coordination among this population. **Methods:** We analyzed data from the 2016 to 2018 National Survey of Children's Health (n = 102,341). Bivariate and multivariable analyses were conducted to compare the prevalence of chronic health conditions, functional disabilities, and care coordination among children with and without CP. Multivariable logistic regression models were used to estimate the adjusted odds ratio of comorbid conditions, functional disabilities, and unmet need for care coordination. Associations between select sociodemographic factors and unmet need for care coordination were assessed. **Results:** Children with CP had significantly higher prevalence of all the comorbid conditions and functional disabilities examined. The most prevalent health conditions among children with CP were allergies (34.2%), anxiety (26.5%), and asthma (25.1%). Notably, children with CP had higher odds of autism spectrum disorder (adjusted odds ratio [aOR] = 2.97; 95% confidence interval [CI] 1.40-6.30) and mental health conditions (aOR = 3.65; 95% CI 2.15-6.21). More than half (53.8%) of children with CP had unmet need for care coordination. They also had higher odds of unmet need for care coordination than children without CP (aOR = 2.63; 95% CI 1.69-4.10). **Conclusion:** Children with CP have high prevalence of chronic health conditions and are also more likely to have unmet need for care coordination. Given the complexity of CP, our study supports the need for robust efforts to ensure that all children with CP have effective care coordination.

PMID: [33990510](#)**22. Early indicators of cardiovascular disease are evident in children and adolescents with cerebral palsy**

Nevin Hammam, Harald Becher, John Andersen, Patricia J Manns, Jackie L Whittaker, Lesley Pritchard

Disabil Health J. 2021 May 5;101112. doi: 10.1016/j.dhjo.2021.101112. Online ahead of print.

Background: Cardiovascular disease (CVD) is a leading cause of early mortality among young adults with cerebral palsy. While low physical activity in childhood has been hypothesized as a potential contributor to increased CVD risk in early adulthood, little is known about timing of vascular disease progression and the presence of subclinical atherosclerosis has not been extensively evaluated in children with cerebral palsy. **Objective:** The aim of this study was to determine if measures of vascular structure and function are different between children and adolescents with and without cerebral palsy. **Methods:** In this cross-sectional study, we measured carotid intima-media thickness (CIMT), and brachial artery flow-mediated dilation (FMD) of children with and without cerebral palsy. Group means for CIMT and brachial artery FMD absolute (FMDA) and percent of relative change (FMDR%) were compared using Wilcoxon rank-sum tests. **Results:** A total of 26 children and adolescents with cerebral palsy (46.1% girls, mean age = 15.0 ± 2.0 years) and 19 controls (68.4% girls, mean age = 13.3 ± 2.6 years) participated. Children with cerebral palsy had significantly greater average CIMT (0.43 ± 0.02 mm) than children without cerebral palsy (0.41 ± 0.01 mm, p = 0.04), and lower FMDA (0.16 ± 0.15 mm vs. 0.29 ± 0.20 mm; respectively, p = 0.03). **Conclusions:** Children and adolescents with cerebral palsy may exhibit impairments in vascular structure and function which represent an increased risk of premature atherosclerosis compared to children without cerebral palsy. Additional research to identify risk factors specific to children with cerebral palsy that would support the development of effective screening processes for early identification would enable clinicians to implement targeted preventive strategies.

PMID: [34016565](#)

23. Velocity dependent measure of spasticity: Reliability in children and juveniles with neuromotor disorders

Petra Marsico, Victoria Frontzek-Weps, Hubertus J A van Hedel

J Pediatr Rehabil Med. 2021 Apr 30. doi: 10.3233/PRM-200704. Online ahead of print.

Purpose: The purpose of this study was to create a clear, standardized test description to rate spasticity severity into four categories according to the definition given by Lance [1], referred to as the Velocity Dependent Measure of Spasticity (VDMS). **Method:** Muscle groups of the upper and lower limbs of children with neuromotor disorders were evaluated on their response to passive movement in a fast-versus slow-velocity test condition. The interrater and test-retest reliability were assessed using Gwet's alpha one (95%-CI) and the percentage agreement. **Results:** Two physiotherapists independently assessed 45 children and youths (age 4-19 years). The interrater reliability of the VDMS was substantial to almost perfect (Gwet's alpha one: 0.66-0.99, n= 45) while the test-retest reliability was almost perfect as well (Gwet's alpha one: 0.83-1.00, n= 42). **Conclusion:** The VDMS can be recommended as a reliable assessment with a standardized procedure to assess spasticity of the extremities in children with neuromotor disorders.

PMID: [33998556](#)**24. The Effects of Basic Photography Education on Quality of Life, Self-Esteem, Life Satisfaction and Moods in Children with Diplegic Cerebral Palsy: A Randomized Controlled Study**

Hakan Alkan, Oya Topuz, Bekir İnce, Şahin Kapıkıran

Phys Occup Ther Pediatr. 2021 May 16;1-11. doi: 10.1080/01942638.2021.1923613. Online ahead of print.

Aims: To investigate the effects of basic photography education and practices on health-related-quality-of-life (HRQoL), self-esteem, life satisfaction, and moods of children with diplegic cerebral palsy (CP). **Methods:** Twenty children with diplegic CP who did not have an intellectual or communication disability were included in this prospective randomized controlled clinical study and divided into two equal groups using a computer-generated list of random numbers to receive either eight weeks of basic photographic training or a control group. Basic photography training including a total of 20 hours theoretical and 30 hours practice was given during the eight weeks in this study. HRQoL, self-esteem, life satisfaction, depression and anxiety were assessed at baseline and at the end of the training. **Results:** At the end of the treatment, statistically significant improvements were found for self-reported HRQoL, life satisfaction, and self-esteem in the basic photographic training group compared to the control group ($p < 0.05$). However, no statistically significant between group difference was found for parents' version of the HRQoL ($p > 0.05$). **Conclusions:** The findings suggest that basic photography training can improve HRQoL, life satisfaction, and self-esteem in children with diplegic CP.

PMID: [33998374](#)**25. Supine lying center of pressure movement characteristics as a predictor of normal developmental stages in early infancy**

Junsig Wang, Safeer F Siddicky, Tara Johnson, Namarta Kapil, Bittu Majmudar, Erin M Mannen

Technol Health Care. 2021 May 7. doi: 10.3233/THC-202754. Online ahead of print.

Background: Absent or abnormal fidgety movements in young infants are associated with subsequent diagnoses of developmental disorders such as cerebral palsy. The General Movement Assessment (GMA) is a qualitative clinical tool to visually identify infants with absent or abnormal fidgety movements associated with developmental stage, yet no quantitative measures exist to detect fidgety activity. **Objective:** To determine whether a correlation exists between quantitative Center of Pressure (CoP) measurements during supine lying and age. **Methods:** Twenty-four healthy full-term infants participated in the Institutional Review Board-approved study. Participants were placed supine in view of a GoPro camera on an AMTI force plate for two minutes. Spontaneous movements were evaluated by three trained raters using the GMA. Traditional CoP parameters (range, total path length, mean velocity, and mean acceleration of resultant CoP) were assessed, and complexity of each of the resultant CoP variables (location, velocity, and acceleration) was calculated by sample entropy. Linear regression with Pearson correlation was performed to assess the correlations between the CoP parameters and adjusted age. **Results:** Nineteen infants were deemed fidgety per the GMA and were included in further analyses. All Sample entropy measures and range of resultant CoP had significant correlations with adjusted age ($p < 0.05$). Sample entropy of resultant CoP decreased with increasing age while range of resultant CoP increased with increasing age. **Conclusion:** The results suggest that complexity of

CoP and range of CoP are good predictors of age in typical developing infants during the fidgety period. Therefore, an approach using these parameters should be explored further as a quantifiable tool to identify infants at risk for neurodevelopmental impairment.

PMID: [33998566](#)

26. Pre-eclampsia Complicated With Maternal Renal Dysfunction Is Associated With Poor Neurological Development at 3 Years Old in Children Born Before 34 Weeks of Gestation

Noriko Yoneda, Satoshi Yoneda, Sayaka Tsuda, Mika Ito, Arihiro Shiozaki, Hideki Niimi, Taketoshi Yoshida, Akitoshi Nakashima, Shigeru Saito

Front Pediatr. 2021 Apr 29;9:624323. doi: 10.3389/fped.2021.624323. eCollection 2021.

Objective: The purpose of this study was to investigate perinatal factors associated with a poor neurodevelopmental outcome in preterm infants. **Methods:** A retrospective study was conducted by searching our clinical database between January 2006 and December 2016. A total of 165 singleton children who were born between 23 and 33 weeks of gestation were included. We defined poor neurological development outcomes as follows: cerebral palsy; intellectual disability; developmental disorder including autism and attention-deficit/hyperactivity disorder; low score (<85 points) on Bayley Scales of Infant and Toddler Development, Third Edition (Bayley-III); or low score of Kyoto Scale of Psychological Development corrected at 3 years old. We diagnosed maternal renal dysfunction according to the Clinical Practice Guideline for chronic kidney disease 2018 and the Best Practice Guide 2015 for Care and Treatment of Hypertension in Pregnancy. **Results:** The rate of poor neurological development was 25/165 (15.2%): cerebral palsy (n = 1), intellectual disability (n = 1), developmental disorder (n = 2), low score of Bayley-III (n = 20), and low score of Kyoto Scale of Psychological Development (n = 1). Preeclampsia complicated with maternal renal dysfunction (P = 0.045) and delivery at <30 weeks of gestation (P = 0.007) were independent risk factors for poor neurological development. **Conclusions:** In addition to previous risk factors such as delivery at <30 weeks of gestation, preeclampsia complicated with renal dysfunction was also associated with poor neurodevelopmental outcomes corrected at 3 years old.

PMID: [33996679](#)

27. Term birth weight and neurodevelopmental outcomes

Marianna Cortese, Dag Moster, Allen J Wilcox

Epidemiology. 2021 May 13. doi: 10.1097/EDE.0000000000001350. Online ahead of print.

Background: Preterm birth is an important risk factor for neurodevelopmental disabilities. The vast majority of these disabilities occur, however, among term births. The role of fetal growth restriction specifically among term babies has been incompletely described. **Methods:** We conducted a population-based study of term birth weight and its link to a range of neurodevelopmental outcomes using Norwegian health registries. To remove the influence of preterm birth, we restricted our analyses to 1.8 million singleton babies born during a narrow range of term gestational age (39–41 weeks). Babies with malformations were excluded. We adjusted analyses simply for year of birth, as further adjustments for sex, parity, maternal age, smoking, marital status, immigrant status, and parental education had trivial influence. An additional sibling analysis controlled for unmeasured family-based confounding. **Results:** The risk of neurodevelopmental disabilities at term steadily increased at birth weights lower than 3.5 kg. Using the category of 3.5–3.9 kg as the reference, the odds reached 25-fold for cerebral palsy at the smallest weights (95% confidence interval 8.0–79), 16-fold for vision/hearing disability (4.0–65), 11-fold for intellectual impairment (6.9–17), 7-fold for schizophrenia (1.0–50), 5.4-fold for epilepsy (2.6–12), and 3.5-fold for autism spectrum (1.3–9.4) and behavioral disorders including attention deficit hyperactivity disorder (ADHD) (2.1–5.4). Associations remained robust with sibling controls. **Conclusions:** Reduced fetal growth is a powerful predictor of a wide variety of neurodevelopmental disabilities independent of preterm delivery.

PMID: [34001752](#)

28. Unravelling neuroinflammation in abusive head trauma with radiotracer imaging

Rahul M Nikam, Xuyi Yue, Vinay V Kandula, Bishnuhari Paudyal, Sigrid A Langhans, Lauren W Averill, Arabinda K Choudhary

Review *Pediatr Radiol*. 2021 May;51(6):966-970. doi: 10.1007/s00247-021-04995-z. Epub 2021 May 17.

Abusive head trauma (AHT) is a leading cause of mortality and morbidity in child abuse, with a mortality rate of approximately 25%. In survivors, the prognosis remains dismal, with high prevalence of cerebral palsy, epilepsy and neuropsychiatric disorders. Early and accurate diagnosis of AHT is challenging, both clinically and radiologically, with up to one-third of cases missed on initial examination. Moreover, most of the management in AHT is supportive, reflective of the lack of clear understanding of specific pathogenic mechanisms underlying secondary insult, with approaches targeted toward decreasing intracranial hypertension and reducing cerebral metabolism, cell death and excitotoxicity. Multiple studies have elucidated the role of pro- and anti-inflammatory cytokines and chemokines with upregulation/recruitment of microglia/macrophages, oligodendrocytes and astrocytes in severe traumatic brain injury (TBI). In addition, recent studies in animal models of AHT have demonstrated significant upregulation of microglia, with a potential role of inflammatory cascade contributing to secondary insult. Despite the histological and biochemical evidence, there is a significant dearth of specific imaging approaches to identify this neuroinflammation in AHT. The primary motivation for development of such imaging approaches stems from the need to therapeutically target neuroinflammation and establish its utility in monitoring and prognostication. In the present paper, we discuss the available data suggesting the potential role of neuroinflammation in AHT and role of radiotracer imaging in aiding diagnosis and patient management.

PMID: [33999238](#)

29. Cerebral palsy litigation after fifty years: A hoax on you

Thomas P Sartwelle, James C Johnston, Berna Arda, Mehila Zebenigus

Indian J Med Ethics. Oct-Dec 2020;V(4):1-15. doi: 10.20529/IJME.2020.093.

The worldwide cerebral palsy (CP) litigation crisis is predicated on the hoax that electronic foetal monitoring (EFM) predicts and prevents CP. There are decades of research disproving this hoax, yet EFM continues to be performed in the vast majority of labours in developed countries with resultant harm to mothers and babies alike through unnecessary caesarean sections with all of the attendant complications and ramifications of that procedure. This article reviews the history and evolution of EFM, explores the reasons for its misuse, discusses how obstetricians have abandoned their ethical mandate by failing to obtain informed consent for EFM, and proposes a realistic, practical solution that would effectively change the standard of care.

PMID: [34018953](#)

30. Proteomic changes in the hippocampus and motor cortex in a rat model of cerebral palsy: Effects of topical treatment.

Wang T, Zhang Y, Chen W, Tao J, Xue Q, Ge W, Dou W, Ma C.

Biomed Pharmacother. 2021 Jan;133:110844. doi: 10.1016/j.biopha.2020.110844. Epub 2020 Nov 11.

PMID: [33186793](#)

31. Corrigendum to 'Proteomic changes in the hippocampus and motor cortex in a rat model of cerebral palsy: Effects of topical treatment' [Biomed. Pharmacother. 133 (2021) 110844]

Tao Wang, Yusheng Zhang, Weiwu Chen, Jin Tao, Qiao Xue, Wei Ge, Wanchen Dou, Chao Ma

Published Erratum *Biomed Pharmacother*. 2021 May 15;140:111695. doi: 10.1016/j.biopha.2021.111695. Online ahead of print.

PMID: [34004509](#)