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Interventions and Management

1. Hip surgery and radiology reporting for children with cerebral palsy prior to initiation of a hip surveillance program

Kyra Kane, Marshall Siemens, Shane Wunder, Jacqueline Kraushaar, J Alexandra Mortimer, Muhammed Siddiqui

J Pediatr Rehabil Med. 2021 Jun 5. doi: 10.3233/PRM-201506. Online ahead of print.

Purpose: Hip displacement impacts quality of life for many children with cerebral palsy (CP). While early detection can help avoid dislocation and late-stage surgery, formalized surveillance programs are not ubiquitous. This study aimed to examine: 1) surgical practices around pediatric hip displacement for children with CP in a region without formalized hip surveillance; and 2) utility of MP compared to traditional radiology reporting for quantifying displacement. **Methods:** A retrospective chart review examined hip displacement surgeries performed on children with CP between 2007-2016. Surgeries were classified as preventative, reconstructive, or salvage. Pre- and post-operative migration percentage (MP) was calculated for available radiographs using a mobile application and compared using Wilcoxon Signed Ranks test. MPs were also compared with descriptions in the corresponding radiology reports using directed and conventional content analyses. **Results:** Data from 67 children (115 surgical hips) was included. Primary surgery types included preventative (63.5% hips), reconstructive (36.5%), or salvage (0%). For the 92 hips with both radiology reports and radiographs available, reports contained a range of descriptors that inconsistently reflected the retrospectively-calculated MPs. **Conclusion:** Current radiology reporting practices do not appear to effectively describe hip displacement for children with CP. Therefore, standardized reporting of MP is recommended.

PMID: [34092658](https://pubmed.ncbi.nlm.nih.gov/34092658/)

2. The Femoral Head-Shaft Angle Is Not a Predictor of Hip Displacement in Children Under 5 Years With Cerebral Palsy: A Population-based Study of Children at GMFCS Levels III-V

Terje Terjesen, Joachim Horn

J Pediatr Orthop. 2021 Jun 8. doi: 10.1097/BPO.0000000000001875. Online ahead of print.

Background: The aim of this study was to evaluate whether the femoral head-shaft angle (HSA) is a predictor of hip displacement in children with cerebral palsy (CP). **Methods:** The patients were recruited from a population-based hip surveillance program. Inclusion criteria were age under 5 years, bilateral CP, Gross Motor Function Classification System (GMFCS) levels III-V, and migration percentage (MP) of both hips <40% at the primary radiograph. With these criteria, 101 children (61 boys) were included. GMFCS was level III in 26 patients, level IV in 23, and level V in 52. An anteroposterior radiograph of the pelvis was taken at diagnosis and at the last follow-up. Only the worst hip of each patient (the hip with the largest MP) was used for the analyses. **Results:** The mean age at the primary radiograph was 2.4 years (range, 0.8 to 4.9 y).

The mean primary HSA was 171.0 degrees (range, 152 to 190 degrees). The mean follow-up time was 4.3 years (range, 0.9 to 11.8 y). The mean MP at the primary radiograph was 17.5% (range, 0% to 39%) and at the last follow-up 41.9% (range, 0% to 100%). At that point, MP was <40% in 54 hips and \geq 40% in 47 hips. There was no significant difference in primary HSA between patients with final MP<40% and those with final MP \geq 40% (170.8 and 171.3 degrees, respectively; P=0.761). At the last follow-up, the mean HSA was significantly larger in hips with final MP \geq 40% than in hips with final MP<40% (171.1 vs. 167.4 degrees; P=0.029). Conclusions: There was a markedly increased valgus position of the proximal femur in nonambulatory children with CP. However, the primary HSA in children below 5 years of age was not a predictor of later hip displacement, defined as MP \geq 40%.

PMID: [34101699](#)

3. IncobotulinumtoxinA for the treatment of lower-limb spasticity in children and adolescents with cerebral palsy: A phase 3 study

Florian Heinen, Petr Kaňovský, A Sebastian Schroeder, Henry G Chambers, Edward Dabrowski, Thorin L Geister, Angelika Hanschmann, Francisco J Martinez-Torres, Irena Pulte, Marta Banach, Deborah Gaebler-Spira

J Pediatr Rehabil Med. 2021 Jun 5. doi: 10.3233/PRM-210040. Online ahead of print.

Purpose: Investigate the efficacy and safety of multipattern incobotulinumtoxinA injections in children/adolescents with lower-limb cerebral palsy (CP)-related spasticity. **Methods:** Phase 3 double-blind study in children/adolescents (Gross Motor Function Classification System - Expanded and Revised I-V) with unilateral or bilateral spastic CP and Ashworth Scale (AS) plantar flexor (PF) scores \geq 2 randomized (1:1:2) to incobotulinumtoxinA (4, 12, 16 U/kg, maximum 100, 300, 400 U, respectively) for two 12- to 36-week injection cycles. Two clinical patterns were treated. Pes equinus (bilateral or unilateral) was mandatory; if unilateral, treatment included flexed knee or adducted thigh. **Endpoints:** Primary: AS-PF change from baseline to 4 weeks; Coprimary: investigator-rated Global Impression of Change Scale (GICS)-PF at 4 weeks; Secondary: investigator's, patient's, and parent's/caregiver's GICS, Gross Motor Function Measure-66 (GMFM-66). **Results:** Among 311 patients, AS-PF and AS scores in all treated clinical patterns improved from baseline to 4-weeks post-injection and cumulatively across injection cycles. GICS-PF and GICS scores confirmed global spasticity improvements. GMFM-66 scores indicated better motor function. No significant differences between doses were evident. Treatment was well-tolerated, with no unexpected treatment-related adverse events or neutralising antibody development. **Conclusion:** Children/adolescents with lower-limb spasticity experienced multipattern benefits from incobotulinumtoxinA, which was safe and well-tolerated in doses up to 16 U/kg, maximum 400 U.

PMID: [34092664](#)

4. Generic scaled versus subject-specific models for the calculation of musculoskeletal loading in cerebral palsy gait: Effect of personalized musculoskeletal geometry outweighs the effect of personalized neural control Hans Kainz, Mariska Wesseling, Ilse Jonkers

Biomech (Bristol, Avon). 2021 Jun 1;87:105402. doi: 10.1016/j.clinbiomech.2021.105402. Online ahead of print.

Background: Musculoskeletal modelling is used to assess musculoskeletal loading during gait. Linear scaling methods are used to personalize generic models to each participant's anthropometry. This approach introduces simplifications, especially when used in paediatric and/or pathological populations. This study aimed to compare results from musculoskeletal simulations using various models ranging from linear scaled to highly subject-specific models, i.e., including the participant's musculoskeletal geometry and electromyography data. **Methods:** Magnetic resonance images (MRI) and gait data of one typically developing child and three children with cerebral palsy were analysed. Musculoskeletal simulations were performed to calculate joint kinematics, joint kinetics, muscle forces and joint contact forces using four modelling frameworks: 1) Generic-scaled model with static optimization, 2) Generic-scaled model with an electromyography-informed approach, 3) MRI-based model with static optimization, and 4) MRI-based model with an electromyography-informed approach. **Findings:** Root-mean-square-differences in joint kinematics and kinetics between generic-scaled and MRI-based models were below 5° and 0.15 Nm/kg, respectively. Root-mean-square-differences over all muscles was below 0.2 body weight for every participant. Root-mean-square-differences in joint contact forces between the different modelling frameworks were up to 2.2 body weight. Comparing the simulation results from the typically developing child with the results from the children with cerebral palsy showed similar root-mean-square-differences for all modelling frameworks. **Interpretation:** In our participants, the impact of MRI-based models on joint contact forces was higher than the impact of including electromyography. Clinical reasoning based on overall root-mean-square-differences in musculoskeletal simulation results between healthy and pathological participants are unlikely to be affected by the modelling choice.

PMID: [34098149](#)

5. Do physical activity interventions influence subsequent attendance and involvement in physical activities for children with cerebral palsy: a systematic review

Gaela Kilgour, Brooke Adair, Ngaire Susan Stott, Michael Steele, Amy Hogan, Christine Imms

Disabil Rehabil. 2021 Jun 7;1-17. doi: 10.1080/09638288.2021.1909151. Online ahead of print.

Purpose: To investigate if children with cerebral palsy have sustained attendance and involvement in physical activities after completing physical activity interventions. **Methods:** The Preferred Reporting Items for Systematic Reviews and Meta-Analyses guidelines were followed. Seven databases were searched for the period 2001-2020 with hand-searching of pertinent reference lists. Criteria for study inclusion were participants aged 0-18 years and $\geq 50\%$ with cerebral palsy; follow-up ≥ 1 month beyond completion of the physical activity intervention; and measurement of attendance and/or involvement in any physical activity post-intervention. Study selection, data extraction, and risk of bias assessments (Physiotherapy Evidence Database (PEDro) or tool for non-randomised studies) were completed independently by paired reviewers. Results were compiled by narrative synthesis. **Results:** Thirteen studies were included (11 randomised controlled trials (RCTs), two non-randomised case series; intervention sample sizes: 6-34). All study participants had cerebral palsy and were aged 4-16.7 years. PEDro scores for the RCTs ranged from 5 to 10; 10 did not blind one or more therapist, participant, or assessor. Two case series showed high risk of bias. Twelve studies reported on attendance, with positive changes in three studies. At 4-14 weeks post-intervention, two studies demonstrated positive changes were maintained. Four studies included involvement outcomes; one reporting positive changes in physical activity involvement four weeks after intervention completion. **Conclusions:** Physical activity attendance may be influenced by physical activity interventions in the short term, but more robust research designs are required to investigate whether gains can be sustained. Activity involvement, which may influence ongoing participation, is under-researched. **Implications for Rehabilitation:** Positive changes in attendance and involvement following physical activity interventions appear short term at best. Physical activity interventions should have longer follow-up periods to determine the effect on sustained physical activity participation. Careful selection and reporting of attendance and involvement outcome measures is required. The optimal physical activity intervention to increase attendance or involvement in physical activities remains uncertain.

PMID: [34097836](#)

6. Nintendo Wii Balance Board therapy for postural control in children with cerebral palsy: a systematic review and meta-analysis

Desirée Montoro-Cárdenas, Irene Cortés-Pérez, Noelia Zagalaz-Anula, María C Osuna-Pérez, Esteban Obrero-Gaitán, Rafael Lomas-Vega

Review Dev Med Child Neurol. 2021 Jun 8. doi: 10.1111/dmcn.14947. Online ahead of print.

Aim: To analyse the efficacy of Nintendo Wii therapy (NWT) on functional balance in children with cerebral palsy (CP). **Method:** A systematic review with meta-analysis (PROSPERO identification number CRD42020169510) was performed using randomized controlled trials (RCTs) that examined the effect of NWT on functional, dynamic, and static balance in children with CP, assessed with the Pediatric Balance Scale, the Timed Get Up and Go Test, and the One Leg Stance Test respectively. The pooled effect was calculated using the Cohen's standardized mean difference (SMD). **Results:** Eleven RCTs with 270 children (when sex was reported: 43% females, 57% males) with CP (mean age [SD] 10y 1mo [1y 1mo], range 5-16y) were included. On functional balance, we found very low-quality evidence with a large effect of NWT compared with no intervention (SMD 0.95, 95% confidence interval [CI] 0.02-1.89) and moderate-quality evidence for using NWT plus conventional physical therapy (CPT) versus CPT (SMD 0.78, 95% CI 0.20-1.35) in sessions of approximately 30 minutes (SMD 0.86, 95% CI 0.20-1.52) and interventions lasting longer than 3 weeks (SMD 1.03, 95% CI 0.58-1.47). For dynamic balance, very low-quality evidence for a medium effect for using NWT plus CPT versus CPT (SMD 0.70, 95% CI 0.12-1.29) was found. **Interpretation:** NWT can be considered an effective treatment for improving functional and dynamic balance in children with CP, especially when combined with CPT in 30-minute sessions with interventions lasting longer than 3 weeks.

PMID: [34105150](#)

7. Humanoid Robot Based Platform to Evaluate the Efficacy of Using Inertial Sensors for Spasticity Assessment in Cerebral Palsy

Nicholas J Cooney, Atul Singh Minhas

IEEE J Biomed Health Inform. 2021 Jun 11;PP. doi: 10.1109/JBHI.2021.3088133. Online ahead of print.

Spasticity is commonly present in individuals with cerebral palsy (CP) and manifests itself as shaky movements, muscle tightness and joint stiffness. Accurate and objective measurement of spasticity is investigated using inertial measurement unit (IMU) sensors. However, use of current IMU-based devices is limited to clinics in urban areas where experienced and trained health professionals are available to collect spasticity data. Designing these devices based on the wearable internet of things based architectures with edge computing will expand their use to home, aged care or remote clinics enabling less-experienced health professionals or care givers to collect spasticity data. However, these new designs require rigorous testing during their prototyping stage and collection of supporting data for regulatory approvals. This work demonstrates that a humanoid robot can act as an accurate model of the movements of CP individuals performing pendulum test during their spasticity assessment. Utilizing this model, we present a robust platform to evaluate new designs of IMU-based spasticity measurement devices.

PMID: [34115599](#)

8. Reliability of Processing 3-D Freehand Ultrasound Data to Define Muscle Volume and Echo-intensity in Pediatric Lower Limb Muscles with Typical Development or with Spasticity

Britta Hanssen, Nathalie De Beukelaer, Simon-Henri Schless, Francesco Cenni, Lynn Bar-On, Nicky Peeters, Guy Molenaers, Anja Van Campenhout, Christine Van den Broeck, Kaat Desloovere

Ultrasound Med Biol. 2021 Jun 7;S0301-5629(21)00207-6. doi: 10.1016/j.ultrasmedbio.2021.04.028. Online ahead of print.

This investigation assessed the processor reliability of estimating muscle volume and echo-intensity of the rectus femoris, tibialis anterior and semitendinosus. The muscles of 10 typically developing children (8.15 [1.40] y) and 15 children with spastic cerebral palsy (7.67 [3.80] y; Gross Motor Function Classification System I = 5, II = 5, III = 5) were scanned with 3-D freehand ultrasonography. For the intra-processor analysis, the intra-class correlations coefficients (ICCs) for muscle volume ranged from 0.943-0.997, with relative standard errors of measurement (SEM%) ranging from 1.24%-8.97%. For the inter-processor analysis, these values were 0.853 to 0.988 and 3.47% to 14.02%, respectively. Echo-intensity had ICCs >0.947 and relative SEMs <4% for both analyses. Muscle volume and echo-intensity can be reliably extracted for the rectus femoris, semitendinosus and tibialis anterior in typically developing children and children with cerebral palsy. The need for a single processor to analyze all data is dependent on the size of the expected changes or differences.

PMID: [34112554](#)

9. Trends in Prevalence and Severity of Pre/Perinatal Cerebral Palsy Among Children Born Preterm From 2004 to 2010: A SCPE Collaboration Study

Catherine Arnaud, Virginie Ehlinger, Malika Delobel-Ayoub, Dana Klapouszczak, Oliver Perra, Owen Hensey, David Neubauer, Katalin Hollódy, Daniel Virella, Gija Rackauskaite, Andra Greitane, Kate Himmelmann, Els Ortibus, Ivana Dakovic, Guro L Andersen, Antigone Papavasiliou, Elodie Sellier, Mary Jane Platt, Inge Krägeloh-Mann

Front Neurol. 2021 May 20;12:624884. doi: 10.3389/fneur.2021.624884. eCollection 2021.

Aim: To report on prevalence of cerebral palsy (CP), severity rates, and types of brain lesions in children born preterm 2004 to 2010 by gestational age groups. **Methods:** Data from 12 population-based registries of the Surveillance of Cerebral Palsy in Europe network were used. Children with CP were eligible if they were born preterm (<37 weeks of gestational age) between 2004 and 2010, and were at least 4 years at time of registration. Severity was assessed using the impairment index. The findings of postnatal brain imaging were classified according to the predominant pathogenic pattern. Prevalences were estimated per 1,000 live births with exact 95% confidence intervals within each stratum of gestational age: ≤27, 28-31, 32-36 weeks. Time trends of both overall prevalence and prevalence of severe CP were investigated using multilevel negative binomial regression models. **Results:** The sample comprised 2,273 children. 25.8% were born from multiple pregnancies. About 2-thirds had a bilateral spastic CP. 43.5% of children born ≤27 weeks had a high impairment index compared to 37.0 and 38.5% in the two other groups. Overall prevalence significantly decreased (incidence rate ratio per year: 0.96 [0.92-1.00]) in children born 32-36 weeks. We showed a decrease until 2009 for children born 28-31 weeks but an increase in 2010 again,

and a steady prevalence (incidence rate ratio per year = 0.97 [0.92-1.02] for those born ≤ 27 weeks. The prevalence of the most severely affected children with CP revealed a similar but not significant trend to the overall prevalence in the corresponding GA groups. Predominant white matter injuries were more frequent in children born < 32 weeks: 81.5% (≤ 27 weeks) and 86.4% (28-31 weeks), compared to 63.6% for children born 32-36 weeks. Conclusion: Prevalence of CP in preterm born children continues to decrease in Europe excepting the extremely immature children, with the most severely affected children showing a similar trend.

PMID: [34093391](#)

10. Caregiver-reported health-related quality of life of New Zealand children born very and extremely preterm

Gordon X H Liu, Jane E Harding , PIANO Study Team

PLoS One. 2021 Jun 8;16(6):e0253026. doi: 10.1371/journal.pone.0253026. eCollection 2021.

Background: Children born preterm, particularly at earlier gestations, are at increased risk for mortality and morbidity, but later health-related quality of life (HRQoL) is less well described. Neurodevelopmental impairment and socio-economic status may also influence HRQoL. Our aim was to describe the HRQoL of a cohort of New Zealand children born very and extremely preterm, and how this is related to neurodevelopmental impairment, gestational age, and socio-economic deprivation. Methods: Children born < 30 weeks' gestation or < 1500 g birthweight were assessed at 7 years' corrected age. Caregivers completed the Child Health Questionnaire Parent Form (CHQ-PF50), and the Health Utilities Index Mark 2 (HUI-2). Neurodevelopmental impairment was defined as Wechsler full scale intelligence quotient below -1 standard deviation (SD), Movement Assessment Battery for Children total score ≤ 15 percentile, cerebral palsy, deafness, or blindness. Results: Data were collected for 127 children, of whom 60 (47%) had neurodevelopmental impairment. Overall, HRQoL was good: mean (SD) CHQ-PF50 physical summary score = 50.8 (11.1), psychosocial summary score = 49.3 (9.1) [normative mean 50 (10)]; HUI-2 dead-healthy scale = 0.92 (0.09) [maximum 1.0]. Neurodevelopmental impairment, lower gestational age, and higher socio-economic deprivation were all associated with reduced HRQoL. However, on multivariable analysis, only intelligence quotient and motor function were associated with psychosocial HRQoL, while intelligence quotient was associated with physical HRQoL. Conclusions: Most seven-year-old children born very and extremely preterm have good HRQoL. Further improvements will require reduced neurodevelopmental impairment.

PMID: [34101760](#)

11. Assessment of family needs of children with cerebral palsy in Northern-Nigeria: A cross-sectional study

Abdullahi B Umar, Abdulsalam M Yakasai, Musa S Danazumi, Usman T Shehu, Umaru M Badaru, Bashir Kaka

J Pediatr Rehabil Med. 2021 May 28. doi: 10.3233/PRM-200696. Online ahead of print.

Purpose: This study aimed to identify the needs and priorities of parents of children with cerebral palsy (CP) in order to improve care by increasing family participation in rehabilitation programmes. Methods: This cross-sectional questionnaire-based study was conducted between January to March 2019. Convenience sampling was used to recruit 43 family members (18 years and above) of children with CP who came to the physiotherapy departments for rehabilitation services for their children. Fisher's exact test was used to analyse the association between socio-demographic characteristics and each of the need items. Results: Five different items were identified to be the family needs that were most frequently met: a) 'need for active involvement in the child's treatment and therapies' (n= 40; 93.0%), b) 'need for the provision of standard medical care' (n= 39; 90.7%), c) 'need for questions to be answered honestly' (n= 38; 88.4%), d) 'need for healthcare professionals to respect the child's wishes' (n= 36; 83.7%), e) 'need for mothers to discuss their feelings (depression, stress etc.) with someone who has similar experience' (n= 36; 83.7%). Conversely, three items were the most unmet family needs: a) 'need to have professionals to consult whenever the child needs help' (n= 39; 90.7%), b) 'need to be informed about the child's prognosis' (n= 41; 95.3%), and c) 'need to have financial support to provide the child with adequate care' (n= 43; 100%). Conclusion: All participants overwhelmingly reported that their financial needs were their highest priority. The multiple needs of families of children with disabilities must be assessed and considered in rehabilitation services when treating children with CP.

PMID: [34092657](#)

12. Prediction of outcome from MRI and general movements assessment after hypoxic-ischaemic encephalopathy in low-income and middle-income countries: data from a randomised controlled trial

Karoline Aker, Niranjana Thomas, Lars Adde 4 5, Beena Koshy, Miriam Martinez-Biarge, Ingeborg Nakken, Caroline S Padankatti, Ragnhild Støen

Arch Dis Child Fetal Neonatal Ed. 2021 Jun 10;fetalneonatal-2020-321309. doi: 10.1136/archdischild-2020-321309. Online ahead of print.

Objective: To evaluate the accuracy of neonatal MRI and general movements assessment (GMA) in predicting neurodevelopmental outcomes in infants with hypoxic-ischaemic encephalopathy (HIE). **Design:** Secondary analyses of a randomised controlled trial (RCT). **Setting:** Tertiary neonatal intensive care unit in India. **Methods:** Fifty infants with HIE were included in an RCT of therapeutic hypothermia (25 cooled and 25 non-cooled). All infants underwent brain MRI at day 5, GMA at 10-15 weeks and outcome assessments including Bayley Scales of Infant and Toddler Development, third edition, at 18 months. Associations between patterns of brain injury, presence/absence of fidgety movements (FMs) and outcomes were assessed. **Results:** Seventeen of 47 (36%) had adverse outcome (5 (21%) cooled vs 12 (52%) non-cooled, $p=0.025$). Eight infants died (four before an MRI, another three before GMA). Two developed severe cerebral palsy and seven had Bayley-III motor/cognitive composite score <85 . Twelve (26%) had moderately/severely abnormal MRI and nine (23%) had absent FMs. The positive predictive value (95% CI) of an adverse outcome was 89% (53% to 98%) for moderate/severe basal ganglia and thalamic (BGT) injury, 83% (56% to 95%) for absent/equivocal signal in the posterior limb of the internal capsule (PLIC) and 67% (38% to 87%) for absent FMs. Negative predictive values (95% CI) were 85% (74% to 92%) for normal/mild BGT injury, 90% (78% to 96%) for normal PLIC and 86% (74% to 93%) for present FMs. **Conclusions:** Neonatal MRI and GMA predicted outcomes with high accuracy in infants with HIE. The GMA is a feasible low-cost method which can be used alone or complementary to MRI in low-resource settings to prognosticate and direct follow-up. Trial registration number: CTRI/2013/05/003693.

PMID: [34112719](#)

13. Addressing disparities among children with cerebral palsy: Optimizing enablement, functioning, and participation

Deirdre Flanagan, Deborah Gaebler, Emma-Lorraine B Bart-Plange, Michael E Msall

J Pediatr Rehabil Med. 2021 Jun 5. doi: 10.3233/PRM-210015. Online ahead of print.

Purpose: Recognizing health disparities among children with cerebral palsy (CP) is necessary for understanding potential risk factors for CP and for implementing early and effective preventative and intervention treatments. However, there is currently little and conflicting evidence regarding the direct impact of contextual factors such as socioeconomic status (SES) for children with CP in the United States. These contextual factors include the complex social determinants of health on prematurity, comprehensive informed obstetric management for minority and vulnerable populations, and cumulative adversity disproportionately experienced by children, by gender, minority status, immigration, poverty, and structural racism. **Methods:** This study presents results from a review of health disparities among children with CP, using registry and population surveillance data from Australia, Canada, Scandinavia, the United Kingdom, Ireland, Turkey, and the United States. **Results:** The review confirmed that there are significant health disparities among children with CP, both in terms of prevalence and severity, based on factors such as SES, neighborhood disadvantage, maternal education, gender, and minority status. **Conclusion:** Strategies need to be implemented in the United States to promote enablement and functioning among children with CP who face additional health disparities. This requires a greater understanding of population groups at increased risk, comprehensive assessment and care for young children with motor delays, and systematic population counts of children and adults with CP using registries and systems of neurodevelopmental surveillance across health, education, and community rehabilitation. These efforts also require sensitivity to structural and persistent racism, stigma, trauma-informed care, and culturally sensitive community engagement. Additional efforts are also required to improve outcomes over the life course for individuals living a life with CP from a framework of enablement, self-direction, equity and social justice.

PMID: [34092660](#)

14. Decreased clinical response to therapy in pediatric patients with cerebral palsy: Current trends and challenges

Matthew McLaughlin, Didem Inanoglu

J Pediatr Rehabil Med. 2021 Jun 5. doi: 10.3233/PRM-210028. Online ahead of print.

PMID: [34092663](#)

15. Disrupted access to therapies and impact on well-being during the COVID-19 pandemic for children with motor impairment and their caregivers

Ellen N Sutter, Linda Smith Francis, Sunday M Francis, Daniel H Lench, Samuel T Nemanich, Linda E Krach, Theresa Sukal-Moulton, Bernadette T Gillick

Am J Phys Med Rehabil. 2021 Jun 3. doi: 10.1097/PHM.0000000000001818. Online ahead of print.

Objectives: Determine the impact of the COVID-19 pandemic on access to rehabilitation therapies, and the impact of changes in therapy access on the physical and mental well-being of children with motor impairment and their caregivers. **Design:** Caregivers of children <18 years old with childhood-onset motor impairment (primarily cerebral palsy) completed an anonymous survey through the online platform REDCap between May 5 and July 13, 2020. **Results:** The survey was completed by 102 participants. Before the pandemic, 92 of 102 children (90%) were receiving one or more therapies; at the time surveyed, 55 children (54%) were receiving any therapies ($p < .001$). Greater than 40% of the sample reported increased child stress, decreased physical activity, and/or decline in mobility/movement. Participants who reported a decrease in number of therapies at the time surveyed more frequently reported lower satisfaction with treatment delivery ($p < .001$), decline in child's mobility ($p = .001$), and increased caregiver stress ($p = .004$). Five qualitative themes were identified from open-ended question responses related to therapies and well-being. **Conclusions:** Access to pediatric rehabilitation therapies was disrupted during COVID-19. Disrupted access may be related to impact on physical and mental health. With the expansion of telehealth, caregiver and child feedback should be incorporated to optimize benefit.

PMID: [34091465](#)

16. Neonatal Cranial Ultrasound Findings Among Infants Born Extremely Preterm: Associations With Neurodevelopmental Outcomes at Ten Years of Age

Heather Campbell, Jennifer Check, Karl C K Kuban, Alan Leviton, Robert M Joseph, Jean A Frazier, Laurie M Douglass, Kyle Roell, Elizabeth N Allred, Lynn Ansley Fordham, Stephen R Hooper, Hernan Jara, Nigel Paneth, Irina Mokrova, Hongyu Ru, Hudson P Santos Jr, Rebecca C Fry, T Michael O'Shea

J Pediatr. 2021 Jun 3;S0022-3476(21)00510-2. doi: 10.1016/j.jpeds.2021.05.059. Online ahead of print.

Objective: To examine the association between neonatal cranial ultrasound abnormalities among infants born extremely preterm and neurodevelopmental outcomes at ten years of age. **Study design:** In a multi-center birth cohort of infants born at < 28 weeks' gestation, 889 of 1198 survivors were evaluated for neurological, cognitive, and behavioral outcomes at 10 years of age. Sonographic markers of white matter damage (WMD) included echolucencies in the brain parenchyma and moderate to severe ventricular enlargement. Neonatal cranial ultrasound findings were classified as: intraventricular hemorrhage (IVH) without WMD, IVH with WMD, WMD without IVH, and neither IVH nor WMD. **Results:** WMD without IVH was associated with an increased risk of cognitive impairment (OR 3.5, 95% CI 1.7, 7.4), cerebral palsy (OR 14.3, 95% CI 6.5, 31.5), and epilepsy (OR 6.9; 95% CI 2.9, 16.8). Similar associations were found for WMD accompanied by IVH. Isolated IVH was not significantly associated these outcomes. **Conclusions:** Among children born extremely preterm, cranial ultrasound abnormalities, particularly those indicative of WMD, are predictive of neurodevelopmental impairments at 10 years of age. The strongest associations were found with cerebral palsy.

PMID: [34090894](#)

17. Spectrum of Movement Disorders and Correlation with Functional Status in Children with Cerebral Palsy

Narayanawamy Suresh, Divyani Garg, Sanjay Pandey, Rajeev Kumar Malhotra, Ritu Majumdar, Sharmila B Mukherjee, Suvasini Sharma

Indian J Pediatr. 2021 Jun 7. doi: 10.1007/s12098-021-03785-7. Online ahead of print.

Objectives: To detail the spectrum of movement disorders (MD) among children with cerebral palsy (CP) and assess impact on functional status. **Methods:** In this cross-sectional study, children with CP were recruited and examined for various MDs. Tone abnormality was assessed using Hypertonia Assessment Tool (HAT), functional status using Gross Motor Function

Classification System Expanded and Revised (GMFCS E&R), Manual Ability Classification System (MACS), and Communication Function Classification System (CFCS). These scores were classified into mild-moderate (level I-III)/severe (level IV-V) categories. Results: A total of 113 children (mean age 4.9 ± 3.4 y, 66.4% boys) were enrolled. MDs were noted in 52 (46%) children; the most frequent were dystonia (28%), chorea (14%), choreoathetosis (8%). Of 64 children with quadriparetic CP, 27 (42.2%) demonstrated MDs. Of 19 children with hemiparetic CP, 2 (10.5%) had MDs. Of 16 children with dyskinetic CP, 15 (93%) had MDs. Children with dyskinetic CP had significantly higher frequency of MDs ($p = 0.001$). There was no difference in occurrence of all MDs or dystonia alone amongst the two categories (mild-moderate/severe) of GMFCS E&R levels, CFCS levels or MACS levels. Conclusion: Although diverse MDs occur frequently in CP, these do not correlate with the broad functional status of the child. The study is limited by small sample size.

PMID: [34097232](#)

18. Neuromotor examination in unilateral cerebral palsy: Bilateral impairments in different levels of motor integration

Deisiane Oliveira Souto, Thalita Karla Flores Cruz, Patrícia Lemos Bueno Fontes, Korbinian Moeller, Vitor Geraldi Haase

Appl Neuropsychol Child. 2021 Jun 8;1-11. doi: 10.1080/21622965.2021.1932498. Online ahead of print.

Unilateral cerebral palsy (UCP) usually results in damage to the unilateral pyramidal system. However, the clinical presentation of neuromotor deficits also suggests lesions to the extrapyramidal and cerebellar systems bilaterally. In this study, we developed and tested a behavioral neuromotor examination protocol assessing impairments at three levels of motor integration for children with UCP, also considering impairments of the non-paretic upper limb as well as the influences of the laterality of the lesion. We included 30 children with UCP (10.79 ± 2.61 years) and 60 healthy children (8.27 ± 1.57 years) in the study. All children were assessed on general cognitive ability and classified according to the manual ability classification system (MACS). Our neuromotor examination protocol incorporated specific tasks for each level of motor integration: pyramidal, extrapyramidal and cerebellar. Children with UCP and controls did not differ with respect to general cognitive abilities and sex but children with UCP were significantly older. Controls performed significantly better than children with UCP on neuromotor tasks at all levels of motor integration. Additionally, performance of the non-plegine hand in children with UCP was significantly inferior to controls. With the exception of fine motor skills (pyramidal level), children with right and left UCP did not differ. Our behavioral neuromotor examination was sensitive to reveal impairments at all three levels of motor integration bilaterally in children with UCP-although more subtle for the non-paretic limb.

PMID: [34100328](#)

19. Muscle regeneration in spastic muscles of children with cerebral palsy

Mariz Vainzof, Juliana Gurgel-Giannetti

Dev Med Child Neurol. 2021 Jun 6. doi: 10.1111/dmcn.14953. Online ahead of print.

PMID: [34091893](#)

20. Lactoferrin versus iron hydroxide polymaltose complex for the treatment of iron deficiency anemia in children with cerebral palsy: a randomized controlled trial

Omneya M Omar, Hala Assem, Doaa Ahmed, Marwa S Abd Elmaksoud

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Iron deficiency anemia (IDA) is common among children with cerebral palsy (CP), and studies on the efficacy of lactoferrin (Lf) in the treatment of IDA are limited. This study aimed to compare the efficacy of Lf with that of iron hydroxide polymaltose complex (IPC) in the treatment of IDA in children with CP. This randomized controlled study, conducted at Alexandria University Children's Hospital, enrolled 70 children aged 1-10 years with CP and IDA; 35 children randomly received IPC, whereas the other 35 received Lf. Four children withdrew from the study; thus, only 66 children were analyzed (32 in the IPC group and 34 in the Lf group). At baseline, the hemoglobin level and other blood parameters were similar between the two intervention groups. After four weeks of treatment, both the IPC and Lf groups showed significant improvements in hemoglobin (Hb), serum ferritin (SF), serum iron, total iron-binding capacity, mean corpuscular volume, and mean corpuscular hemoglobin from baseline. Upon comparing the two treatment groups, adjusted mean Hb and SF changes in

the Lf group were significantly higher than that of the IPC group ($p = 0.001$ and $p = 0.033$, respectively), and constipation was less likely to occur in the Lf group than the IPC group ($p = 0.049$). Conclusion: Lactoferrin is effective and superior to IPC as an oral iron replacement therapy in children with CP and IDA, as it has fewer side effects. What is Known: • Lactoferrin (LF) is a natural glycoprotein capable of treating iron deficiency anemia (IDA). • Studies on the efficacy of Lf in the treatment of IDA in children with cerebral palsy (CP) are limited. What is New? • This trial compared the efficacy of Lf and iron hydroxide polymaltose complex (IPC) as treatments of IDA in children with CP. • Lf is effective and even better than IPC as a treatment of IDA in children with CP, as it has fewer side effects.

PMID: [34101010](#)

21. Blood pressure in adults with cerebral palsy: a systematic review and meta-analysis of individual participant data

Suzie Noten, Rita J G van den Berg-Emons, Deborah E Thorpe, Patricia C Heyn, Christina M Marciniak, Patrick G McPhee, Robert P Lamberts, Nelleke G Langerak, Olaf Verschuren, Tommi Salokivi, Katherine M Morrison, Mark D Peterson, Chonnanid Limsakul, Henk J Stam, Grigorios Papageorgiou, Jorie Versmissen, Wilma M A Van Der Slot

J Hypertens. 2021 Jun 7. doi: 10.1097/HJH.0000000000002912. Online ahead of print.

Objectives: This systematic review and meta-analysis was designed to determine the overall mean blood pressure and prevalence of hypertension among a representative sample of adults living with cerebral palsy by combining individual participant data. Additional objectives included estimating variations between subgroups and investigating potential risk factors for hypertension. **Methods:** Potential datasets were identified by literature searches for studies published between January 2000 and November 2017 and by experts in the field. Samples of adults with cerebral palsy ($n \geq 10$, age ≥ 18 years) were included if blood pressure data, cerebral palsy-related factors (e.g. cerebral palsy subtype), and sociodemographic variables (e.g. age, sex) were available. Hypertension was defined as at least 140/90 mmHg and/or use of antihypertensive medication. **Results:** We included data from 11 international cohorts representing 444 adults with cerebral palsy [median (IQR) age of the sample was 29.0 (23.0-38.0); 51% men; 89% spastic type; Gross Motor Function Classification System levels I-V]. Overall mean SBP was 124.9 mmHg [95% confidence interval (CI) 121.7-128.1] and overall mean DBP was 79.9 mmHg (95% CI 77.2-82.5). Overall prevalence of hypertension was 28.7% (95% CI 18.8-39.8%). Subgroup analysis indicated higher blood pressure levels or higher prevalence of hypertension in adults with cerebral palsy above 40 years of age, men, those with spastic cerebral palsy or those who lived in Africa. BMI, resting heart rate and alcohol consumption were risk factors that were associated with blood pressure or hypertension. **Conclusion:** Our findings underscore the importance of clinical screening for blood pressure in individuals with cerebral palsy beginning in young adulthood.

PMID: [34102658](#)

22. Women with cerebral palsy: A qualitative study about their experiences with sexual and reproductive health education and services

Susan Hayden Gray, Rachel Byrne, Sinead Christensen, David Williams, Molly Wylie, Eileen Fowler, Deborah Gaebler-Spira, Christina Marciniak, Laurie Glader

J Pediatr Rehabil Med. 2021 May 28. doi: 10.3233/PRM-210020. Online ahead of print.

Purpose: To explore the recalled experiences of women with CP regarding sexual health education and services they received. **Methods:** Semi-structured interviews and focus groups were conducted at four academic tertiary hospitals with 33 adult women with CP. Templates were used to ask about four key content domains: appointment planning (including planning for a gynecologic exam), accessibility of services, experiences with providers, and recommendations for improvement. Sessions were transcribed verbatim and analyzed to generate a coding dictionary. Blinded coding was carried out for each transcript, with duplicate coding used to confirm identified themes. Iterative analysis was used to identify and consolidate coding and key themes. **Results:** Similar barriers were discussed at the four sites, including lack of accessible exam tables, hospital staff unfamiliar with physical disabilities, and assumptions that women with CP are not sexually active. Many described the sexual education they received as brief, omitted, or mistimed. Self-advocacy was crucial, and recommended strategies ranged from pre-gynecologic exam medication to visit checklists. **Conclusion:** Reproductive health education for young women with CP is frequently inadequate. Medical professionals lack relevant knowledge and awareness; medical facilities lack necessary infrastructure. Recommendations for improvements are made.

PMID: [34092661](#)

23. Genetic testing in individuals with cerebral palsy

Halie J May, Jennifer A Fasheun, Jennifer M Bain, Evan H Baugh, Louise E Bier, Anya Revah-Politi, New York Presbyterian Hospital/Columbia University Irving Medical Center Genomics Team; David P Roye Jr, David B Goldstein, Jason B Carmel

Dev Med Child Neurol. 2021 Jun 10. doi: 10.1111/dmcn.14948. Online ahead of print.

AIM To determine which patients with cerebral palsy (CP) should undergo genetic testing, we compared the rate of likely causative genetic variants from whole-exome sequencing in individuals with and without environmental risk factors. **METHOD** Patients were part of a convenience and physician-referred cohort recruited from a single medical center, and research whole-exome sequencing was completed. Participants were evaluated for the following risk factors: extreme preterm birth, brain bleed or stroke, birth asphyxia, brain malformations, and intrauterine infection. **RESULTS** A total of 151 unrelated individuals with CP (81 females, 70 males; mean age 25y 7mo [SD 17y 5mo], range 3wks-72y) participated. Causative genetic variants were identified in 14 participants (9.3%). There was no significant difference in diagnostic rate between individuals with risk factors (10 out of 123; 8.1%) and those without (4 out of 28; 14.3%) (Fisher's exact $p=0.3$). **INTERPRETATION** While the rate of genetic diagnoses among individuals without risk factors was higher than those with risk factors, the difference was not statistically significant at this sample size. The identification of genetic diagnoses in over 8% of cases with risk factors suggests that these might confer susceptibility to environmental factors, and that further research should include individuals with risk factors.

PMID: [34114234](#)

24. Genome-Wide Association Study Identifies Genetic Risk Factors for Spastic Cerebral Palsy

Andrew T Hale, Oluwatoyin Akinnusotu, Jing He, Janey Wang, Natalie Hibshman, Chevis N Shannon, Robert P Naftel

Neurosurgery. 2021 Jun 7;nyab184. doi: 10.1093/neuros/nyab184. Online ahead of print.

Background: Although many clinical risk factors of spastic cerebral palsy (CP) have been identified, the genetic basis of spastic CP is largely unknown. Here, using whole-genome genetic information linked to a deidentified electronic health record (BioVU) with replication in the UK Biobank and FinnGen, we perform the first genome-wide association study (GWAS) for spastic CP. **Objective:** To define the genetic basis of spastic CP. **Methods:** Whole-genome data were obtained using the multi-ethnic genotyping array (MEGA) genotyping array capturing single-nucleotide polymorphisms (SNPs), minor allele frequency (MAF) > 0.01, and imputation quality score (r^2) > 0.3, imputed based on the 1000 genomes phase 3 reference panel. Threshold for genome-wide significance was defined after Bonferroni correction for the total number of SNPs tested ($P < 5.0 \times 10^{-8}$). Replication analysis (defined as $P < .05$) was performed in the UK Biobank and FinnGen. **Results:** We identify 1 SNP (rs78686911) reaching genome-wide significance with spastic CP. Expression quantitative trait loci (eQTL) analysis suggests that rs78686911 decreases expression of GRIK4, a gene that encodes a high-affinity kainate glutamatergic receptor of largely unknown function. Replication analysis in the UK Biobank and FinnGen reveals additional SNPs in the GRIK4 loci associated with CP. **Conclusion:** To our knowledge, we perform the first GWAS of spastic CP. Our study indicates that genetic variation contributes to CP risk.

PMID: [34098570](#)

25. All cases of cerebral palsy warrant genomic screening

Alastair H Maclennan

Dev Med Child Neurol. 2021 Jun 10. doi: 10.1111/dmcn.14951. Online ahead of print.

PMID: [34114233](#)

26. Intrauterine growth restriction followed by oxygen support uniquely interferes with genetic regulators of myelination

Jill Chang, Robert H Lurie, Abhineet Sharma, Mirrah Bashir, Camille M Fung, Robert W Dettman, Maria L V Dizon

eNeuro. 2021 Jun 7;ENEURO.0263-20.2021. doi: 10.1523/ENEURO.0263-20.2021. Online ahead of print.

Intrauterine growth restriction (IUGR) and oxygen exposure in isolation and combination adversely affect the developing brain, putting infants at risk for neurodevelopmental disability including cerebral palsy. Rodent models of IUGR and postnatal hyperoxia have demonstrated oligodendroglial injury with subsequent white matter injury (WMI) and motor dysfunction. Here we investigate transcriptomic dysregulation in IUGR with and without hyperoxia exposure to account for the abnormal brain structure and function previously documented. We performed RNA sequencing and analysis using a mouse model of IUGR and found that IUGR, hyperoxia, and the combination of IUGR with hyperoxia (IUGR/hyperoxia) produced distinct changes in gene expression. IUGR in isolation demonstrated the fewest differentially expressed genes compared to control. In contrast, we detected several gene alterations in IUGR/hyperoxia; genes involved in myelination were strikingly downregulated. We also identified changes to specific regulators including TCF7L2, BDNF, SOX2, and DGCR8, through Ingenuity Pathway Analysis, that may contribute to impaired myelination in IUGR/hyperoxia. Our findings show that IUGR with hyperoxia induces unique transcriptional changes in the developing brain. These indicate mechanisms for increased risk for WMI in IUGR infants exposed to oxygen and suggest potential therapeutic targets to improve motor outcomes. Significance Statement: This study demonstrates that perinatal exposures of IUGR and/or postnatal hyperoxia result in distinct transcriptomic changes in the developing brain. In particular, we found that genes involved in normal developmental myelination, myelin maintenance, and remyelination were most dysregulated when IUGR was combined with hyperoxia. Understanding how multiple risk factors lead to WMI is the first step in developing future therapeutic interventions. Additionally, because oxygen exposure is often unavoidable after birth, an understanding of gene perturbations in this setting will increase our awareness of the need for tight control of oxygen use to minimize future motor disability.

PMID: [34099489](#)

27. Locomotion is impacted differently according to the perinatal brain injury model: Meta-analysis of preclinical studies with implications for cerebral palsy

Sabrina da Conceição Pereira, Raul Manhães-de-Castro, Diego Bulcão Visco, Glayciele Leandro de Albuquerque, Caio Matheus Santos da Silva Calado, Vanessa da Silva Souza, Ana Elisa Toscano

Review J Neurosci Methods. 2021 Jun 8;360:109250. doi: 10.1016/j.jneumeth.2021.109250. Online ahead of print.

Background: Different approaches to reproduce cerebral palsy (CP) in animals, contribute to the knowledge of the pathophysiological mechanism of this disease and provide a basis for the development of intervention strategies. Locomotion and coordination are the main cause of disability in CP, however, few studies highlight the quantitative differences of CP models, on locomotion parameters, considering the methodologies to cause brain lesions in the perinatal period. Methods: Studies with cerebral palsy animal models that assess locomotion parameters were systematically retrieved from Medline/ PubMed, SCOPUS, LILACS, and Web of Science. Methodological evaluation of included studies and quantitative assessment of locomotion parameters were performed after eligibility screening. Results: CP models were induced by hypoxia-ischemia (HI), Prenatal ischemia (PI), lipopolysaccharide inflammation (LPS), intraventricular haemorrhage (IVH), anoxia (A), sensorimotor restriction (SR), and a combination of different models. Overall, 63 studies included in qualitative synthesis showed a moderate quality of evidence. 16 studies were included in the quantitative meta-analysis. Significant reduction was observed in models that combined LPS with HI related to distance traveled (SMD -7.24 95 % CI [-8.98, -5.51], Z = 1.18, p < 0.00001) and LPS with HI or anoxia with sensory-motor restriction (SMD -6.01, 95 % CI [-7.67, -4.35], Z = 7.11), or IVH (SMD -4.91, 95 % CI [-5.84, -3.98], Z = 10.31, p < 0.00001) related to motor coordination. Conclusion: The combination of different approaches to reproduce CP in animals causes greater deficits in locomotion and motor coordination from the early stages of life to adulthood. These findings contribute to methodological refinement, reduction, and replacement in animal experimentation, favoring translational purposes.

PMID: [34116077](#)

28. Biochemical and structural basis of the passive mechanical properties of whole skeletal muscle

Richard L Lieber, Ben Binder-Markey

J Physiol. 2021 Jun 7. doi: 10.1113/JP280867. Online ahead of print.

Passive skeletal muscle mechanical properties of whole muscle are not as well understood as muscle's active mechanical properties. Both the structural basis for passive mechanical properties and the properties themselves are challenging to determine because it is not clear which structures within skeletal muscle actually bear passive loads and there are not

established standards by which to make mechanical measurements. Evidence suggests that titin bears the majority of the passive load within the single muscle cell. However, at larger scales, such as fascicles and muscles, there is emerging evidence that the extracellular matrix (ECM) bears the majority of the load. Complicating the ability to quantify and compare across size scales, muscles, and species, definitions of muscle passive properties such as stress, strain, modulus and stiffness can be made relative to many reference parameters. These uncertainties make a full understanding of whole muscle passive mechanical properties and modeling these properties very difficult. Future studies defining the specific load bearing structures and their composition and organization are required to fully understand passive mechanics of the whole muscle and develop therapies to treat disorders in which passive muscle properties are altered such as muscular dystrophy, traumatic laceration, and contracture due to upper motor neuron lesion as seen in spinal cord injury, stroke and cerebral palsy. Abstract figure legend Schematic representation of passive load bearing at various skeletal muscle scales. In the sarcomere (upper left), load is borne by the giant elastic protein titin and, to a lesser extent, desmin. The myofilaments actin and myosin are responsible for active force production. Sarcomeres in series (myofibrils) connect to the fiber surface at specialized focal adhesions called costameres at which desmin and other cytoskeletal proteins converge (upper right). Muscle fibers are embedded in a connective tissue matrix (lower left) composed primarily of basal lamina collagen type IV. At larger scales, passive load is borne in the perimysium (lower right) by perimysial cables that seem to be composed of collagen types 1 and 3 and this is where the majority of passive load is borne. This article is protected by copyright. All rights reserved.

PMID: [34101193](#)

29. Preventing the human factor: organizational aspects linked to fetal asphyxia

Roberta Amadori, Sara Grandioso, Elena Osella, Carmela Melluzza, Carmen I Aquino, Viviana Stampini, Raffaele Tinelli, Daniela Surico

Minerva Obstet Gynecol. 2021 Jun 7. doi: 10.23736/S2724-606X.21.04815-6. Online ahead of print.

Background: Perinatal asphyxia can cause cerebral palsy and hypoxic - ischemic encephalopathy. They are public health problems because they cause permanent disability. Methods: This is a retrospective, analytical, observational study. 162 cases of mothers whose children experienced fetal asphyxia were compared to 361 controls where this condition did not occur. The variables analyzed were classified as: prepartum, intrapartum and organizational. Results: Assisted Reproductive Technology obtained pregnancies, smoking, maternal body mass index, lack of one to one assistance during labor, birth on a day of high volume activity increased the risk of fetal asphyxia, as well as other traditionally linked factors like shoulder dystocia or age over 35 years. Conclusions: Cerebral palsy cannot always be prevented because it is a syndrome with a multitude of potential causes. But a small number of cases is likely to be linked to acute intrapartum events that could be limited by changing organizational policies such as staff training and implementing team work and discussion. Our paper proposes strategies to try and modify organizational risk factors and therefore limit the incidence of fetal asphyxia.

PMID: [34096693](#)

30. What Happens to Our Neuromuscular Patients in Adulthood: Pathway to Independence and Maximal Function

Steven E Koop, M Wade Shrader

J Pediatr Orthop. 2021 Jul 1;41(Suppl 1):S87-S89. doi: 10.1097/BPO.0000000000001779.

Background: Children with neuromuscular disorders regularly seek care from pediatric orthopaedic surgeons. These conditions can have a significant impact on the growth and development of children and their function and well-being as adults. Questions exist about the long-term outcomes of musculoskeletal interventions performed during childhood. Methods: A search of recent literature pertaining to the musculoskeletal and functional consequences of cerebral palsy, spina bifida, Duchenne muscular dystrophy, and spinal muscle atrophy was performed. Information from those articles was combined with the experience of the authors and their institutions. Results: Neuromuscular conditions can result in limb and spine deformities that lead to impaired physical function. Orthopaedic interventions during childhood can improve function and well-being and can be durable into adulthood. Unfortunately, many individuals with these conditions transition to adult health care that lacks the informed, collaborative multidisciplinary care they received as children. This can lead to unmet health care needs and a shortage of long-term natural history and outcome studies that would inform the care of children today. Conclusions: Adults with childhood-onset neuromuscular conditions need, and deserve, dedicated health care systems that include the best aspects of the care they received as children. Pediatric orthopaedic surgeons have a role in promoting the development of such systems and a responsibility to learn from their adult patients. Level of evidence: Expert Opinion.

PMID: [34096544](#)

31. A resource guide to understanding cerebral palsy: Commentary on collaboration to support health literacy and shared decision making

Elizabeth Chan, Cynthia Frisina, Deborah Gaebler-Spira

J Pediatr Rehabil Med. 2021 May 28. doi: 10.3233/PRM-210026. Online ahead of print.

PMID: [34092662](#)

32. The cerebral palsy research network: Building a learning health network for cerebral palsy

Amy F Bailes, Jacob Kean, Paul H Gross, Unni Narayanan, Garey Noritz, Ed Hurvitz, Jeffrey Leonard, Michele Shusterman, Mary Gannotti

J Pediatr Rehabil Med. 2021 Jun 5. doi: 10.3233/PRM-210011. Online ahead of print.

Purpose: The purpose of this study was to measure the growth of the Cerebral Palsy (CP) Research Network towards becoming a Learning Health Network in order to guide future development. **Methods:** Thirteen CP Research Network leaders completed the Network Maturity Grid (NMG) which consists of six domains with eight to 10 components each. The six domains are Systems of Leadership, Governance and Management, Quality Improvement, Engagement and Community, Data and Analytics, and Research. Radar mapping was utilized to display mean scores on a 5-point ordinal scale (1 = not started to 5 = idealized state) across domains and for individual components within domains. Consensus was reached for top priorities for the next 3-5 years. **Results:** Domain scores ranged from 2.4 in Quality Improvement to 3.2 in System of Leadership. The lowest scoring component was clinician clinical decision support and the highest was common purpose. The following priority areas of focus were agreed upon moving forward: development of leaders, financial sustainability, quality improvement education and training, patient reported data, data quality and validation, and primary data collection. **Conclusion:** Results from this project will be utilized for strategic planning to improve the network. Conducting regular self-assessments of the network with the NMG will be useful in achieving the network's ultimate goal to improve care and outcomes for individuals with CP.

PMID: [34092659](#)

33. Quality appraisal of systematic reviews of interventions for children with cerebral palsy reveals critically low confidence

Kat Kolaski, Lynne Romeiser Logan, Katherine D Goss, Charlene Butler

Dev Med Child Neurol. 2021 Jun 6. doi: 10.1111/dmcn.14949. Online ahead of print.

Aim: To evaluate the methodological quality of recent systematic reviews of interventions for children with cerebral palsy in order to determine the level of confidence in the reviews' conclusions. **Method:** A comprehensive search of 22 databases identified eligible systematic reviews with and without meta-analysis published worldwide from 2015 to 2019. We independently extracted data and used A Measurement Tool to Assess Systematic Reviews-2 (AMSTAR-2) to appraise methodological quality. **Results:** Eighty-three systematic reviews met strict eligibility criteria. Most were from Europe and Latin America and reported on rehabilitative interventions. AMSTAR-2 appraisal found critically low confidence in 88% (n=73) because of multiple and varied deficiencies. Only 7% (n=6) had no AMSTAR-2 critical domain deficiency. The number of systematic reviews increased fivefold from 2015 to 2019; however, quality did not improve over time. **Interpretation:** Most of these systematic reviews are considered unreliable according to AMSTAR-2. Current recommendations for treating children with CP based on these flawed systematic reviews need re-evaluation. Findings are comparable to reports from other areas of medicine, despite the general perception that systematic reviews are high-level evidence. The required use of current widely accepted guidance for conducting and reporting systematic reviews by authors, peer reviewers, and editors is critical to ensure reliable, unbiased, and transparent systematic reviews.

PMID: [34091900](#)