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Interventions and Management

1. Constraint-Induced Movement Therapy for Infants With or at Risk for Cerebral Palsy: A Scoping Review

Monik Castillo Dionisio, Alexandra L Terrill

Am J Occup Ther. 2022 Mar 1;76(2):7602205120. doi: 10.5014/ajot.2022.047894.

Importance: Although research shows that older children with hemiplegic cerebral palsy (CP) benefit from constraint-induced movement therapy (CIMT), the efficacy of CIMT among the infant population is unknown. **Objective:** To explore the existing evidence on CIMT for infants age 24 mo and younger with CP or at risk for CP. **Data sources:** PubMed, CINAHL, Cochrane, and ProQuest were searched. **Study Selection and Data Collection:** Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) and American Occupational Therapy Association guidelines were used for abstracting data and assessing data quality. Two frameworks guided this scoping review. **Studies met the following criteria:** infants with hemiplegic CP, published in English between 2000 and 2020, peer reviewed, and addressed changes in upper limb function of the hemiplegic upper limb. **Findings:** Eight articles met the inclusion criteria: 2 randomized controlled trials, 1 retrospective cohort design, 1 pretest-posttest study, 2 single-subject studies, and 2 case studies. **In this scoping review, we examined CIMT protocols for an overview of dosage, constraint, administration, parent training and education, and objective outcome measures. Conclusions and relevance:** Existing literature supports CIMT protocols for infants with CP. However, much variability exists in protocol design and appropriate outcome measures among studies. Higher level research is needed to support the efficacy of CIMT among infants with CP. **What This Article Adds:** This scoping review summarizes the existing literature on CIMT for infants with hemiplegic CP. This information can help guide therapists to implement CIMT protocols in the clinical setting and to identify additional research to establish practice standards.

PMID: [35179556](#)

2. Accelerometry-Based Metrics to Evaluate the Relative Use of the More Affected Arm during Daily Activities in Adults Living with Cerebral Palsy

Isabelle Poitras, Jade Clouâtre, Alexandre Campeau-Lecours, Catherine Mercier

Sensors (Basel). 2022 Jan 28;22(3):1022. doi: 10.3390/s22031022.

Adults living with cerebral palsy (CP) report bimanual and unimanual difficulties that interfere with their participation in activities of daily living (ADL). There is a lack of quantitative methods to assess the impact of these motor dysfunctions on the relative use of each arm. The objective of this study was to evaluate the concurrent and discriminative validity of accelerometry-based metrics when used to assess bimanual and unimanual functions. **Methods:** A group of control subjects and hemiplegic adults living with CP performed six ADL tasks, during which they were wearing an Actigraph GT9X on each wrist and being filmed. Four bimanual and unimanual metrics were calculated from both accelerometry-based and video-

based data; these metrics were then compared to one other with an intraclass correlation coefficient (ICC). Some of these metrics were previously validated in other clinical population, while others were novel. The discriminative validity was assessed through comparisons between groups and between tasks. Results: The concurrent validity was considered as good to excellent (ICC = 0.61-0.97) depending on the experience of the raters. The tasks made it possible to discriminate between groups. Conclusion: The proposed accelerometry-based metrics are a promising tool to evaluate bimanual and unimanual functions in adults living with CP.

PMID: [35161767](#)

3. Dorsal rhizotomy in cerebral palsy: how root sectioning is influenced by intraoperative neuromonitoring

A Joud, M Sindou, I Stella, A Wiedemann, O Klein

Neurochirurgie. 2022 Feb 9;S0028-3770(22)00037-6. doi: 10.1016/j.neuchi.2022.01.009. Online ahead of print.

Introduction and objective: Dorsal rhizotomy is a controversial procedure for treating spasticity in children with cerebral palsy, particularly regarding the influence of intraoperative neuromonitoring (ION). The objective of this study was to evaluate the influence of ION in adjusting root sectioning compared the preoperative program established by the multidisciplinary team. **Material and methods:** Twenty-four consecutive children with spastic diplegia or quadriplegia, operated on between 2017 and 2020 in the University Hospital of Nancy, France, were studied. All underwent the same procedure: Keyhole Intralaminar Dorsal rhizotomy (KIDr) with enlarged multilevel interlaminar openings to access all roots from L2 to S2. The Ventral Root (VR) was stimulated to map radicular myotomes, and the Dorsal Root (DR) to test excitability of the segmental circuitry. Muscle responses were observed independently by the physiotherapist and by EMG-recordings. The study compared final root sectioning per radicular level and per side after ION versus the preoperative program determined by the multidisciplinary team. **Results:** ION resulted in significant differences in final percentage root sectioning ($p < 0.05$), with a decrease for L2 and L3 and an increase for L5. ION modified the symmetry of sectioning, with 32% instead of 5% in preoperative program. Only 5 children showed change in GMFC score 6 months after surgery. **Conclusion:** The use of ION during dorsal rhizotomy led to important modifications of root sectioning during surgery, which justifies individual control of each root, level by level and side by side, to optimize the therapeutic effect.

PMID: [35150726](#)

4. Use of Vancomycin Powder in Spinal Deformity Surgery in Cerebral Palsy Patients is Associated With Proteus Surgical Site Infections

Jerry Y Du, Anne M Dumaine, Walter Klyce, Firoz Miyanji, Paul D Sponseller, Michael P Glotzbecker, Harms Study Group

J Pediatr Orthop. 2022 Feb 14. doi: 10.1097/BPO.0000000000002079. Online ahead of print.

Purpose: Surgical site infection (SSI) rates in pediatric spinal deformity surgery for cerebral palsy (CP) patients are higher than that in idiopathic scoliosis. The use of vancomycin powder is associated with decreased risk of SSI in neuromuscular patients. Prior studies in adult and pediatric early-onset scoliosis patients have shown that vancomycin powder alters microbacterial profile in patients that develop SSI. However, the effects of topical vancomycin powder on microbiology in spinal deformity surgery for CP patients has not been studied. **Methods:** An international multicenter database of CP neuromuscular scoliosis patients was used in this retrospective cohort study. All patients that underwent posterior spinal instrumented fusion for CP neuromuscular scoliosis from 2008 to 2019 were queried, and 50 cases complicated by postoperative SSI were identified. Intraoperative antibiotic details were documented in 49 cases (98.0%). Microbiology details were documented in 45 cases (91.8%). Microbiology for patients that received topical vancomycin powder were compared with patients that did not. A multivariate regression model was used to control for potential confounders. **Results:** There were 45 patients included in this study. There were 27 males (60.0%) and 18 females (40.0%). Mean age at surgery was 14.8 ± 2.4 years. There were 24 patients that received topical vancomycin powder (53.3%). The mean time from index surgery to SSI was 4.3 ± 11.3 months. On univariate analysis of microbiology cultures by vancomycin powder cohort, there were no significant differences in culture types. *Proteus* spp. trended on significance with association with vancomycin powder use ($P = 0.078$). When controlling for potential confounders on multivariate analysis, intraoperative topical vancomycin powder was associated with increased risk for proteus infection (adjusted odds ratio: 262.900, 95% confidence interval: 1.806-38,267.121, $P = 0.028$). **Discussion:** In CP patients undergoing pediatric spinal deformity surgery, the use of vancomycin powder was independently associated with increased risk for proteus infections. Further study into antibiotic regimens for spinal deformity surgery in the CP population should be performed. Level of evidence: Level III-retrospective cohort study.

PMID: [35153286](#)

5. Blade plate versus locking plate fixation of proximal femoral varus osteotomy in children with cerebral palsy

Afolayan K Oladeji, Jason Cummings, Arya Minaie, Andrew J Landau, Joe Eric Gordon, Pooya Hosseinzadeh

J Pediatr Orthop B. 2022 Feb 14. doi: 10.1097/BPB.0000000000000962. Online ahead of print.

Objective: The hip is commonly affected in children with cerebral palsy (CP), requiring proximal femoral varus derotational osteotomies. Novel locking plates afford a popular alternative to traditional blade plates. The purpose of this study was to compare the effectiveness of blade plate versus locking plate fixation in children with CP undergoing proximal femoral osteotomy. **Methods:** We conducted a retrospective review of patients who underwent proximal femoral osteotomy over an 8-year period. Incidence of healing, failure of the procedure, and loss of varus correction were compared between the two groups (blade plate vs. locking plate). Independent samples t-tests and Chi-square analysis were employed to compare differences between continuous and categorical variables, respectively. **Results:** A total of 268 hips [137 right (51.1%)] met inclusion criteria. Ninety-eight hips (36.6%) were fixed with blade plates [170 (63.4%) locking plates]. Although those in the blade plate cohort were more likely to achieve complete radiographic healing by 6 weeks postoperatively (41.09% vs. 18.84%; $P < 0.050$), there was no significant difference ($P > 0.050$) between the two groups for healing at 3, 6, and 12 months ($P > 0.050$). There was no significant difference between the two cohorts regarding the number of patients experiencing migration percentage at least 50% at 6 (3.06% vs. 3.53%) and 12 (3.06% vs. 5.88%) months ($P > 0.050$) or in those undergoing revision surgery at 12 (5.33% vs. 1.18%) and 24 (2.04% vs. 1.76%) months ($P > 0.050$). **Conclusion:** The findings of this retrospective study show similar outcomes between blade plate and proximal femoral locking plates in proximal femoral varus osteotomy in children with CP.

PMID: [35170574](#)

6. Clinicians' Experiences of Instrumented Gait Analysis in Management of Patients with Cerebral Palsy: A Qualitative Study

Anna Hebda-Boon, Bairu Zhang, Augustine Amankwah, Adam P Shortland, Dylan Morrissey

Phys Occup Ther Pediatr. 2022 Feb 15;1-13. doi: 10.1080/01942638.2022.2037808. Online ahead of print.

Aim: To identify the interaction of instrumented gait analysis (IGA) training, expertise, and application in gait-related management of cerebral palsy. **Methods:** Semi-structured interviews with 20 purposively sampled clinicians with varying professional backgrounds, expertise, and training, analyzed using the framework method. **Results:** Fifteen sub-themes were identified within three domains: training, equipment/outputs, and roles/reasons under the core theme IGA practice. Findings were illustrated using the Experience/Equipment/Roles/Training (Exp-ERT) Framework which identifies four user categories - based on influencing factors, beset by barriers, with experience reported as a common enabling factor. Clinicians who encountered barriers in one of the domains were categorized as either "frustrated" or "hesitant" users. Those who were no longer using IGA for clinical decisions were designated "confident non-users". Finally, the 'confident experts' reported the required level of training and access to interpret IGA outputs for clinical decision-making. Expertise gained at any level of clinical practice was shown to initiate advancement within domains. **Conclusions:** Clinicians encounter a multitude of barriers to IGA practice that can result in failure to progress or impact on clinical decision-making. The Exp-ERT Framework emerges strongly from the data and could serve as an evaluation tool to diagnose barriers to confident expertise and support IGA-related professional development planning.

PMID: [35168473](#)

7. Prediction of Communication Impairment in Children With Bilateral Cerebral Palsy Using Multivariate Lesion- and Connectome-Based Approaches: Protocol for a Multicenter Prospective Cohort Study

Jie Hu, Jingjing Zhang, Yanli Yang, Ting Liang, Tingting Huang, Cheng He, Fuqin Wang, Heng Liu, Tijiang Zhang

Front Hum Neurosci. 2022 Jan 31;16:788037. doi: 10.3389/fnhum.2022.788037. eCollection 2022.

Background: Bilateral cerebral palsy (BCP) is the most common type of CP in children and is often accompanied by different degrees of communication impairment. Several studies have attempted to identify children at high risk for communication impairment. However, most prediction factors are qualitative and subjective and may be influenced by rater bias.

Individualized objective diagnostic and/or prediction methods are still lacking, and an effective method is urgently needed to guide clinical diagnosis and treatment. The aim of this study is to develop and validate an objective, individual-based model for the prediction of communication impairment in children with BCP by the time they enter school. Methods: A multicenter prospective cohort study will be conducted in four Chinese hospitals. A total of 178 children with BCP will undergo advanced brain magnetic resonance imaging (MRI) at baseline (corrected age, before the age of 2 years). At school entry, communication performance will be assessed by a communication function classification system (CFCS). Three-quarters of children with BCP will be allocated as a training cohort, whereas the remaining children will be allocated as a test cohort. Multivariate lesion- and connectome-based approaches, which have shown good predictive ability of language performance in stroke patients, will be applied to extract features from MR images for each child with BCP. Multiple machine learning models using extracted features to predict communication impairment for each child with BCP will be constructed using data from the training cohort and externally validated using data from the test cohort. Prediction accuracy across models in the test cohort will be statistically compared. Discussion: The findings of the study may lead to the development of several translational tools that can individually predict communication impairment in children newly diagnosed with BCP to ensure that these children receive early, targeted therapeutic intervention before they begin school. Trial registration: The study has been registered with the Chinese Clinical Trial Registry (ChiCTR2100049497).

PMID: [35173593](#)

8. The Effects of Combining High-Top Shoes with Twister Wrap Orthoses on Balance Parameters of Children with Spastic Diplegic Cerebral Palsy

Mehrdad Davoudi, Mobina Khosravi Farsani, Taher Babae, Hamideh Ranjbar, Seyyed Mohammadreza Shokouhyan, Alireza Ghaznavi, Mehdi Rezaei

J Biomed Phys Eng. 2022 Feb 1;12(1):91-100. doi: 10.31661/jbpe.v0i0.2106-1358. eCollection 2022 Feb.

Background: Cerebral palsy (CP) is a non-progressive encephalopathy before, during, or after childbirth with almost the most common type, i.e. spastic diplegic, leading to a frequent walking problem, In-toeing. Orthoses can reduce the consequences of CP. Objective: This study aimed to evaluate the effect of combining twister wrap orthoses (TWO) with high-top shoes on the balance parameters of children with spastic diplegic CP. Material and methods: In this quasi-experimental study, twenty children (aged 6.8 ± 0.5 years) with spastic diplegic CP with in-toeing gait participated. The tests were conducted in three conditions: 1) in bare-foot, 2) with high-top shoes, and 3) with high-top shoes plus TWO and the orthoses effects on balance parameters were compared. Results: High-top shoes positively decreased center of pressure (COP) sway in the anterior-posterior (AP) direction. Both high-top shoes and high-top shoes with TWO conditions compared to bare-foot conditions significantly improved standing balance by decreasing the ellipse area. Pairwise, wearing TWO and high-top shoes significantly reduced the COP sway in the medial-lateral (ML) and AP directions with a significant difference between using the combined orthoses and the shoe without TWO in ML of COP displacement. Conclusion: High-top shoes alone and the combination of high-top shoes with TWO conditions may enhance the stability of children with spastic diplegia more than barefoot. Although the use of combined orthoses induced significant improvement in the ML direction of COP displacement.

PMID: [35155297](#)

9. Game-Based Dual-Task Exercise Program for Children with Cerebral Palsy: Blending Balance, Visuomotor and Cognitive Training: Feasibility Randomized Control Trial

Tony Szturm, Sanjay Tejraj Parmar, Kavisha Mehta, Deepthi R Shetty, Anuprita Kanitkar, Rasit Eskicioglu, Neha Gaonkar

Randomized Controlled Trial Sensors (Basel). 2022 Jan 19;22(3):761. doi: 10.3390/s22030761.

The objective of this exploratory randomized controlled trial (RCT) was to provide evidence for the feasibility and therapeutic value of a novel game-based dual-task balance exercise program in children with cerebral palsy (CP). Twenty children with CP were recruited and randomized into two groups: (a) the conventional balance training group (CG) and (b) the experimental group (XG), which received a game-based dual-task (DT) balance exercise program. Both groups received their respective therapy programs for 12 weeks at a frequency of three sessions per week. Semi-structured interviews with the parents and children and qualitative analysis were conducted to evaluate the children's experiences with the game-based exercise program. The quantitative analysis included (a) the Pediatric Balance Scale (PBS), (b) Gross Motor Function Measure-88 (GMFM-88), and (c) computerized measures of standing balance performance during various dual-task conditions. Compliance was 100% for all 20 participants. Four themes captured the range of each participant's experiences and opinions: (a) reasons for

participation, (b) likes and dislikes with the technologies, (c) positive effects of the program, and (d) future expectations. Children in the XG demonstrated greater improvements in PBS, GMFM, and DT balance measures as compared to children in the CG. The findings demonstrate feasible trial procedures and acceptable DT-oriented training with a high compliance rate and positive outcomes. These findings support further research and development and progression to the next phase of a full-scale RCT to evaluate the clinical effectiveness of the game-based DT balance exercise program for children with CP.

PMID: [35161508](#)

10. Gaming Technology for Pediatric Neurorehabilitation: A Systematic Review

Marco Iosa, Cristiano Maria Verrelli, Amalia Egle Gentile, Martino Ruggieri, Agata Polizzi

Review Front Pediatr. 2022 Jan 28;10:775356. doi: 10.3389/fped.2022.775356. eCollection 2022.

Introduction: The emergence of gaming technologies, such as videogames and virtual reality, provides a wide variety of possibilities in intensively and enjoyably performing rehabilitation for children with neurological disorders. Solid evidence-based results are however required to promote the use of different gaming technologies in pediatric neurorehabilitation, while simultaneously exploring new related directions concerning neuro-monitoring and rehabilitation in familiar settings. **Aim of the study and methods:** In order to analyze the state of the art regarding the available gaming technologies for pediatric neurorehabilitation, Scopus and Pubmed Databases have been searched by following: PRISMA statements, PICO classification, and PEDro scoring. **Results:** 43 studies have been collected and classified as follows: 11 feasibility studies; six studies proposing home-system solutions; nine studies presenting gamified robotic devices; nine longitudinal intervention trials; and eight reviews. Most of them rely on feasibility or pilot trials characterized by small sample sizes and short durations; different methodologies, outcome assessments and terminologies are involved; the explored spectrum of neurological conditions turns out to be scanty, mainly including the most common and wider debilitating groups of conditions in pediatric neurology: cerebral palsy, brain injuries and autism. **Conclusion:** Even though it highlights reduced possibilities of drawing evidence-based conclusions due to the above outlined biases, this systematic review raises awareness among pediatricians and other health professionals about gaming technologies. Such a review also points out a definite need of rigorous studies that clearly refer to the underlying neuroscientific principles.

PMID: [35155305](#)

11. The diagnostic levels of evidence of instrumented devices for measuring viscoelastic joint properties and spasticity; a systematic review

Levinia Lara van der Velden, Maaïke Anna Catharina de Koff, Gerard Maria Ribbers, Ruud Willem Selles

Review J Neuroeng Rehabil. 2022 Feb 11;19(1):16. doi: 10.1186/s12984-022-00996-7.

Background: Many diagnostic robotic devices have been developed to quantify viscoelastic properties and spasticity of patients with upper motor neuron lesions. However, in clinical practice, subjective and nonvalid clinical scales are still commonly used. To understand the limited use of diagnostic robotic devices assessing viscoelastic joint properties and spasticity in clinical practice, we evaluate the diagnostic level of evidence of studies on these devices. **Method:** A systematic literature review was performed using multiple databases. Two of the authors independently screened all articles. Studies investigating human subjects diagnosed with stroke or cerebral palsy, measured with a mechanical device to assess viscoelastic joint properties and/or spasticity of an extremity. All articles were assigned a diagnostic level of evidence, which was established with a classification strategy based on the number of participants and the design of the study, from a Level 0 (less than 10 subjects) to a Level IV, reporting the long-term clinical consequences in daily care. **Results:** Fifty-nine articles were included. Most studies measured the upper limb (64%) in stroke patients (81%). The highest level of evidence found was Level IIa (53%); these studies correlated the test values of the robotic device with a clinical test or within subgroups. Level 0 (30%) and Level I (17%; determining the range of values of the robotic test) were also common. None of the studies tested their device for diagnostic accuracy (Level III), clinical added value (Level IV). **Conclusion:** The diagnostic evidence needed for implementing robotic devices in clinical practice is lacking. Our findings indicate that more effort should be invested in studying diagnostic accuracy (Level III) or added value for clinical care (Level IV); only these studies can provide clinicians with evidence that robotic devices have added value above the currently-used clinical scales.

PMID: [35148805](#)

12. The long-term burden of congenital cytomegalovirus: Hospitalisation and mortality in a population-based matched cohort study

Hayley Smithers-Sheedy, Gulam Khandaker, Camille Raynes-Greenow, Lloyd Flack, Philip N Britton, Sarah McIntyre, Nadia Badawi, David Burgner, Claire Galea, Cheryl A Jones

Eur J Paediatr Neurol. 2022 Feb 6;37:82-86. doi: 10.1016/j.ejpn.2022.01.019. Online ahead of print.

Aim: Congenital cytomegalovirus (cCMV) infection can result in considerable morbidity and mortality. However, the impact of cCMV on health system utilisation beyond infancy is poorly defined. Here we sought to describe the burden of cCMV health service use and mortality using linked population-based datasets. **Methods:** The design was a matched cohort study using record-linked administrative datasets. Participants included all children aged 0-15 years identified through the New South Wales (NSW), Australia, Admitted Patient Data Collection who were hospitalised with an admission code for cCMV infection between 2001 and 2011. Participants were then matched by age, sex and birth postcode quintile using Socio-Economic Indexes for Area, to children randomly selected from the NSW Perinatal Data Collection, excluding those with central nervous system infections or cerebral palsy. We calculated rate ratios (RR) for hospital admissions adjusted for preterm birth, the median length of stay, cost weights of admission and odds ratio (OR) for deaths. **Results:** There were 130 children with cCMV matched to 2672 children. Neonates (aged <1 month) with cCMV had twice the rate of admissions (RR 2.4 95%CI 2.0, 2.8) and children aged \geq 5yr to < 15yrs with cCMV almost eight times the rate of admissions (RR 7.8 95%CI 5.1, 11.5). Children with cCMV had significantly higher cost weights of admissions and an increased risk of mortality (OR 18.4 95%CI 7.8, 43.6). **Conclusion:** Throughout childhood, children with cCMV had higher rates of hospital admissions, higher admission cost weights and an increased risk of mortality compared with matched peers without cCMV.

PMID: [35151079](#)

13. Adverse motor outcome after paediatric ischaemic stroke: A nationwide cohort study

Katarina Svensson, Anna Walås, Jenny Bolk, Peter Bang, Heléne E K Sundelin

Paediatr Perinat Epidemiol. 2022 Feb 16. doi: 10.1111/ppe.12869. Online ahead of print.

Background: Various frequencies of adverse motor outcomes (cerebral palsy and hemiplegia) after paediatric ischaemic stroke have been reported. Few reports on the risks of adverse motor outcomes in nationwide cohorts and contributing risk factors are available. **Objectives:** To assess risk of adverse motor outcome and potential risk factors thereof after paediatric ischaemic stroke in a nationwide cohort. **Methods:** This nationwide matched cohort study identified 877 children <18 years of age diagnosed with ischaemic stroke through the Swedish national health registers from 1997 to 2016. These children, exposed to ischaemic stroke, alive 1 week after stroke, were matched for age, sex and county of residence with 10 unexposed children. Using Cox regression, we estimated the risk of adverse motor outcomes in children with stroke compared to that in unexposed children. Logistic regression was applied to compare the characteristics of children with and without adverse motor outcomes after stroke. **Results:** Out of the 877 children with ischaemic stroke, 280 (31.9%) suffered adverse motor outcomes compared with 21 (0.2%) of the 8770 unexposed: adjusted hazard ratio (aHR) 167.78 (95% confidence interval (CI) 107.58, 261.66). There were no differences between risk estimates of adverse motor outcome according to age at stroke: perinatal stroke (aHR 124.11, 95% CI 30.45, 505.84) and childhood stroke (aHR 182.37, 95% CI 113.65, 292.64). An association between adverse motor outcome and childhood stroke aOR 1.56 (95% CI 1.05, 2.31) was found when analysing only children with ischaemic stroke. No associations were found between adverse motor outcome and sex, gestational age or parental age at birth. **Conclusions:** The risk of adverse motor outcome is substantial after paediatric ischaemic stroke, especially childhood stroke, confirming results of previous smaller studies. This study found no associations between sex, gestational age or parental age and adverse motor outcome after paediatric ischaemic stroke.

PMID: [35172018](#)

14. Racial-ethnic inequities in age at death among adults with/without intellectual and developmental disability in the United States

Scott D Landes, Janet M Wilmoth, Katherine E McDonald, Alyssa N Smith

Prev Med. 2022 Feb 9;156:106985. doi: 10.1016/j.ypmed.2022.106985. Online ahead of print.

To identify potential differences in racial-ethnic inequities in mortality between adults with/without intellectual and developmental disability, we compared patterns in age at death by race-ethnic status among adults who did/did not have intellectual and developmental disability reported on their death certificate in the United States. Data were from the 2005-2017 U.S. Multiple Cause-of-Death Mortality files. Average age at death by racial-ethnic status was compared between adults, age 18 and older, with/without different types of intellectual and developmental disability reported on their death certificate (N = 32,760,741). A multiple descent pattern was observed among adults without intellectual or developmental disability, with age at death highest among Whites, followed by Asians, Hispanics and Blacks, then American Indians. In contrast, a bifurcated pattern was observed among adults with intellectual disability, with age at death highest among Whites, but lower and similar among all racial-ethnic minority groups. The severity of racial-ethnic inequities in age at death was most pronounced among adults with cerebral palsy. Policy makers and public health experts should be aware that racial-ethnic inequities are different for adults with intellectual and developmental disability - all minorities with intellectual and developmental disability are at greater risk of premature death than their White counterparts.

PMID: [35150747](#)

15. 'There is family tension, but they understand...': familial and sibling relationships following the diagnosis of cerebral palsy in children in Ghana

Joslin Alexei Dogbe, Joana D A Kyeremateng, Maxwell Pephrah Opoku, William Nketsia, Charles Hammond

Int J Dev Disabil. 2019 Feb 10;68(1):35-46. doi: 10.1080/20473869.2019.1573572. eCollection 2022.

The challenges faced by parents raising children with cerebral palsy (CP) have been well explored in the literature. However, little attention has been paid to the experiences of parents raising children with CP in low-income countries, such as Ghana. Objective: Therefore, the objective of this study was to explore parents' experiences of raising children with CP, specifically focusing on the relationships between spouses and between children with CP and their typically developing siblings. Method: Qualitative semi-structured interviews were conducted with 40 parents, who were purposively selected from the largest tertiary hospital in Ghana. Results: The results revealed that typically developing children accept their siblings with CP as their equals and even take up domestic responsibilities to lessen the burden on their parents. However, the parents reported experiencing marital and extended family conflict, financial burden and negative attitudes from spouses, resulting in family tensions. Conclusion: The implications of these findings for policy-making have also been discussed.

PMID: [35173962](#)

16. Altered gene expression levels of genes related to muscle function in adults with cerebral palsy

Jessica Pingel, Jasper Vandenrijt, Marie-Louise Kampmann, Jeppe Dyrberg Andersen

Tissue Cell. 2022 Jan 29;76:101744. doi: 10.1016/j.tice.2022.101744. Online ahead of print.

Cerebral palsy (CP) is the most common cause of movement disorders in children. Next generation sequencing (NGS) studies have previously shown that expression levels are fundamentally different in children with CP compared to typically developing (TD). However, given that children are in full development, we might expect gene expression levels to change once maturity is reached. Therefore, the main purpose of this study was to investigate gene expression levels of 93 target genes in adults with CP using NGS on muscle biopsies of the gastrocnemius, taken from 22 participants (n = 12 adults with CP; n = 10 TD adults). Subsequently, we carried out NGS of the mitochondrial genome to identify mtDNA variants, and additionally we studied the mitochondrial content using transmission electron microscopy images of the gastrocnemius muscle. Finally, we compared systemic ion levels between TD adults and adults with CP. Differential gene expression levels were found in genes involved in muscle contraction (MYH1 and MYBPC2), mitochondrial function (kATP5J, CYCS and NDUFB6), calcium handling (CAMK2B and ATP2A), metabolism (LPL), muscle signaling (MYC, CREB1, ACVR2B, LMNA and TRIM54), and ECM (TNC). There was no statistical significant difference between CP and TD for mtDNA variant frequencies and mitochondrial content. The ion levels of Ca²⁺, Na⁺ and K⁺ were statistically significantly reduced while the Cl⁻ levels were significant increased in adults with CP compared to TD adults. These results highlight that most transcriptional differences are related to muscle function in adults with CP and that mitochondrial function might be altered but not mitochondrial content.

PMID: [35151178](#)

17. Accounting for arterial and capillary blood gases for calculation of cerebral blood flow in preterm infants

Silke Brodtkorb, Irina Sidorenko, Varvara Turova, Esther Rieger-Fackeldey, Ursula Felderhoff-Müser, Andrey Kovtanyuk, Renée Lampe

Eur J Pediatr. 2022 Feb 12. doi: 10.1007/s00431-022-04392-0. Online ahead of print.

One of the most feared neurological complications of premature birth is intraventricular hemorrhage, frequently triggered by fluctuations in cerebral blood flow (CBF). Although several techniques for CBF measurement have been developed, they are not part of clinical routine in neonatal intensive care. A promising tool for monitoring of CBF is its numerical assessment using standard clinical parameters such as mean arterial pressure, carbon dioxide partial pressure (pCO₂) and oxygen partial pressure (pO₂). A standard blood gas analysis is performed on arterial blood. In neonates, capillary blood is widely used for analysis of blood gas parameters. The purpose of this study was the assessment of differences between arterial and capillary analysis of blood gases and adjustment of the mathematical model for CBF calculation to capillary values. The statistical analysis of pCO₂ and pO₂ values collected from 254 preterm infants with a gestational age of 23-30 weeks revealed no significant differences between arterial and capillary pCO₂ and significantly lower values for capillary pO₂. The estimated mean differences between arterial and capillary pO₂ of 15.15 mmHg (2.02 kPa) resulted in a significantly higher CBF calculated for capillary pO₂ compared to CBF calculated for arterial pO₂. Two methods for correction of capillary pO₂ were proposed and compared, one based on the mean difference and another one based on a regression model. Conclusion: Capillary blood gas analysis with correction for pO₂ as proposed in the present work is an acceptable alternative to arterial sampling for the assessment of CBF. What is Known: • Arterial blood analysis is the gold standard in clinical practice. However, capillary blood is widely used for estimating blood gas parameters. • There is no significant difference between the arterial and capillary pCO₂ values, but the capillary pO₂ differs significantly from the arterial one. What is New: • The lower capillary pO₂ values yield significantly higher values of calculated CBF compared to CBF computed from arterial pO₂ measurements. • Two correction methods for the adjustment of capillary pO₂ to arterial pO₂ that made the difference in the calculated CBF insignificant have been proposed.

PMID: [35150310](#)

18. Cord Blood Proteomic Biomarkers for Predicting Adverse Neurodevelopmental Outcomes in Monoamniotic Twins

Young Mi Jung, Seung Mi Lee, Sohee Oh, Hyun-Suk Shin, Chan-Wook Park, Joong Shin Park, Dohyun Han, Jong Kwan Jun

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Monoamniotic twins have a high risk of mortality and perinatal morbidity due to cord entanglement and vascular anastomosis. Despite efforts to reduce the mortality rate through intensive fetal surveillance and timed delivery, poor long-term neurodevelopmental outcomes remain an unsolved problem. This study aimed to identify novel biomarkers predicting abnormal neurodevelopmental outcomes in monoamniotic twins with cord blood samples taken at the time of delivery. Abnormal neurodevelopmental outcomes were defined as (1) a severe brain lesion on neonatal brain ultrasound, (2) developmental delay, (3) cerebral palsy, and/or (4) blindness or deafness. Cord blood was analyzed with mass spectrometry-based proteomics according to the neurodevelopmental outcomes. Statistical analysis was performed to determine the differentially expressed proteins between neonates with normal and abnormal neurodevelopmental outcomes. Several candidate proteins were further validated with enzyme-linked immunosorbent assays. A total of 20 neonates (10 pairs) of monoamniotic twins were included in the proteomic analysis, of which 25% had abnormal neurodevelopmental outcomes. Eighteen proteins were differentially expressed in neonates with abnormal neurodevelopmental outcomes. The upregulated proteins in the neonates with adverse neurodevelopmental outcome were immunoglobulin (Ig)-gamma-4 chain C region, apolipoprotein E, and alpha-fetoprotein. In contrast, Ig-lambda chain V region 4A, Ig-heavy variable 3, Ig-kappa chain C region, Ig-mu chain C region, C1q, ceruloplasmin, and Ig-lambda chain V-I region were decreased. In the validation experiment, the cord blood concentration of ceruloplasmin was significantly lower in neonates with adverse neurodevelopmental outcomes than in those without. Therefore, ceruloplasmin could be a useful predictive biomarker of adverse neurodevelopmental outcomes in monoamniotic twins.

PMID: [35165813](#)

19. Parent-recorded videos of infant spontaneous movement: Comparisons at 3-4 months and relationships with 2-year developmental outcomes in extremely preterm, extremely low birthweight and term-born infants

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Background: Infants born extremely preterm (EP, <28-week gestational age) or extremely low birthweight (ELBW, <1000 g) are at risk of developmental delay and cerebral palsy (CP). The General Movements Assessment (GMA) and its extension, the Motor Optimality Score, revised (MOS-R) (assesses movement patterns and posture), may help to identify early delays. **Objectives:** To compare differences in the MOS-R scored from parent-recorded videos between infants born EP/ELBW and term-born infants, to determine relationships between the MOS-R and 2-year cognitive, language and motor outcomes and if any relationships differ between birth groups and the association of the GMA (fidgety) with CP. **Methods:** A geographical cohort (EP/ELBW and term-control infants) was assessed using the MOS-R inclusive of the GMA at 3- to 4-month corrected age (CA), and the Bayley Scales of Infant and Toddler Development, 3rd edition (Bayley-III) at 2-year CA. Differences in mean total MOS-R between groups, relationships between MOS-R and 2-year outcomes and relationships between GMA (fidgety) and CP in infants born EP/ELBW were estimated using linear/logistic regression. **Results:** Three hundred and twelve infants (147 EP/ELBW; 165 term) had complete MOS-R and Bayley-III assessments. Mean MOS-R was lower in infants born EP/ELBW than controls (mean difference -3.2, 95% confidence interval [CI] -4.2, -2.3). MOS-R was positively related to cognitive (β [regression coefficient] = 0.71, 95% CI 0.27, 1.15), language (β = 0.96, 95% CI 0.38, 1.54) and motor outcomes (β = .89, 95% CI 0.45, 1.34). There was little evidence for interaction effects between birth groups for any outcome. Absent/abnormal fidgety movements were related to CP in children born EP/ELBW (risk ratio 5.91, 95% CI 1.48, 23.7). **Conclusions:** Infants born EP/ELBW have lower MOS-R than infants born at term. A higher MOS-R is related to better outcomes for 2-year development, with similar relationships in both birth groups. Absent/abnormal fidgety movements are related to CP in EP/ELBW survivors.

PMID: [35172019](#)

20. Mothers' experience of having a child with cerebral palsy. A systematic review

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Aim: To explore, synthesise and present findings of qualitative studies describing the experiences of mothers raising a child with cerebral palsy. **Design:** A systematic literature review of the qualitative evidence. **Methods:** A systematic search for qualitative studies published in the following databases: CINAHL (EBSCO), Medline via OVID, SCOPUS, and Google Scholar. The authors independently assessed eligibility, appraised methodological quality using the Critical Appraisal Skills Program tool for qualitative Research (CASP). An inductive thematic analysis method was adopted to synthesise major findings and to construct core concepts and themes. **Results:** Five overarching themes reflecting the experiences and perceptions of mothers raising and caring for a child with cerebral palsy are developed: 1) adapting and making sacrifices; 2) guilt and cultural blame; 3) social stigma and marginalisation; 4) physical, environmental, and financial challenges and 5) healthcare experiences.

PMID: [35158294](#)

21. Diagnostic Evaluation of the Functional Muscle-Bone Unit in Children With Cerebral Palsy With and Without Low Trauma Fractures

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Children and adolescents with cerebral palsy (CP) are at increased risk of low trauma fractures (LTF) due to low bone mineral content (BMC). The risk of LTFs might be overestimated by only age- and sex-adjusted Z-scores for BMC because Z-score based DXA techniques do not take into account other relevant parameters like height, muscle and fat mass. This study aimed to present an update of the functional muscle-bone unit-algorithm (uFMBU-A) to evaluate bone health in children with CP in

order to predict the risk of LTF taking into account the parameters sex, age, height, muscle and fat mass. We performed a monocentric retrospective analysis of 177 DXA-scans of children and adolescents with CP aged 8-19. Six of these 177 patients had sustained at least 1 LTF. Age-, sex- and size adjusted Z-scores of total body less head (TBLH)-BMC, lean body mass and fat mass were calculated. The uFMBU-A was applied to the study group and results were compared with established Z-score based DXA-measurements and algorithm based diagnostic techniques concerning the prediction of LTF risk. The uFMBU-A had the greatest diagnostic odds ratio (13.3 [95% CI 2.41; 72.9]) of the evaluated predictors with a sensitivity of 50.0% (95% CI 11.8; 88.2), specificity of 93% (95% CI 88.1; 96.3). The uFMBU-A was the most accurate method of the evaluated parameters to predict LTF in children with CP and is recommended when evaluating bone health.

PMID: [35168895](#)

22. Incidence of Deep Vein Thrombosis in Cerebral Palsy Following an Orthopaedic Surgical Event

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Purpose: The purpose of this study was to identify the incidence of venous thromboembolism (VTE) and characterize the demographics, comorbidities, and risk factors for patients with cerebral palsy (CP) having orthopaedic surgery. **Methods:** All patients diagnosed with CP who underwent an orthopaedic surgical procedure at one institution between 2008 and 2017 were identified. Diagnosis codes and associated patient events were recovered from the electronic medical record. Each VTE event was reviewed to ascertain an actual VTE episode related to a surgical event. **Results:** The review included 2583 orthopaedic surgical events in 1371 patients. Of the initial 88 cases identified, 28 cases had a deep thrombosis documented. Six cases of VTE occurred within 3 months following the surgical event. Three of these cases had thigh thrombosis, and 2 patients had upper arm thrombosis, and 1 patient had a superior vena cava thrombosis. On further workup, 5 of these 6 patients were identified as having a congenital hypercoagulable condition. **Conclusions:** VTE is a relatively rare occurrence after orthopaedic surgery in pediatric patients with CP, but when it occurs, a full hematologic workup for a congenital hypercoagulable condition is indicated. Based on the low incidence of thigh thrombosis, routine pharmacological or intermittent mechanical calf compression is not recommended. A careful clinical and family history should be performed to identify patients with possible genetic hypercoagulable conditions who would merit prophylaxis. **Level of evidence:** Level IV.

PMID: [35180727](#)

23. The Contributions of Extracellular Matrix and Sarcomere Properties to Passive Muscle Stiffness in Cerebral Palsy

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Cerebral palsy results from an upper motor neuron lesion and significantly affects skeletal muscle stiffness. The increased stiffness that occurs is partly a result of changes in the microstructural components of muscle. In particular, alterations in extracellular matrix, sarcomere length, fibre diameter, and fat content have been reported; however, experimental studies have shown wide variability in the degree of alteration. Many studies have reported changes in the extracellular matrix, while others have reported no differences. A consistent finding is increased sarcomere length in cerebral palsy affected muscle. Often many components are altered simultaneously, making it difficult to determine the individual effects on muscle stiffness. In this study, we use a three dimensional modelling approach to isolate individual effects of microstructural alterations typically occurring due to cerebral palsy on whole muscle behaviour; in particular, the effects of extracellular matrix volume fraction, stiffness, and sarcomere length. Causation between the changes to the microstructure and the overall muscle response is difficult to determine experimentally, since components of muscle cannot be manipulated individually; however, utilising a modelling approach allows greater control over each factor. We find that extracellular matrix volume fraction has the largest effect on whole muscle stiffness and mitigates effects from sarcomere length.

PMID: [35153814](#)

24. A core outcome set for multimorbidity risk in individuals with cerebral palsy

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Aim: To: (1) investigate the importance of outcome measurement instruments (OMIs) within a core outcome set (COS) for multimorbidity (at least two chronic health conditions) risk in individuals with cerebral palsy (CP); (2) investigate the feasibility of OMIs within the COS in international clinical research settings in adolescents and adults with CP; and (3) describe the associations between the COS data and Gross Motor Function Classification System (GMFCS) levels. **Method:** Eighty-three individuals with CP completed a survey on health outcomes: physical behaviour, nutrition, sleep, endurance, body composition, blood pressure, blood lipids, and glucose. A cross-sectional study assessed the feasibility of the COS in 67 adolescents and adults with CP (mean age 30y, SD 15y 1mo, min-max: 14-68y, 52.2% male) at four centres. Prevalence of multimorbidity risk and associations with GMFCS levels are described. **Results:** Most participants rated physical behaviour, nutrition, sleep, and endurance as very important. Body composition, blood pressure, nutrition, and sleep were highly feasible since data were collected in 88% or more participants who consented to having the assessments. Physical behaviour, cardiorespiratory endurance, and blood draws were collected in less than 60% of participants. Total time sedentary ($\rho=0.53$, $p<0.01$) and endurance ($\rho=-0.46$, $p<0.01$) were significantly associated with GMFCS level. **Interpretation:** The COS identified that most participants had poor sleep quality and endurance, did not have healthy diets, and showed increased sedentary behaviour. Individuals with CP valued these outcomes as most important, suggesting a need to assess these modifiable behaviours in this population. Objective measures of physical behaviour and cardiorespiratory endurance in the COS required additional personnel, time, and participant burden. We recommend that healthcare providers should perform a simpler first screen using questionnaire-based assessments and then focus the use of the remainder of the COS if required for the patient.

PMID: [35174883](https://pubmed.ncbi.nlm.nih.gov/35174883/)