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# Cerebral palsy research news

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***Professor Nadia Badawi AM***
Macquarie Group Foundation Chair of Cerebral Palsy

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## Interventions and Management

### 1. Rehabilitation Approach for a Child with Cerebral Palsy and Upper Limb Deficiency

Hiroshi Mano, Emi Inakazu, Satoko Noguchi, Chika Nishizaka, Sayaka Fujiwara, Nobuhiko Haga

Case Reports Prog Rehabil Med. 2021 Mar 18;6:20210016. doi: 10.2490/prm.20210016. eCollection 2021.

Background: Congenital limb deficiency is a rare and intractable anomaly of the limbs; however, prostheses can partially complement the motor function and appearance of the missing limbs. The first prosthesis is usually prescribed for children with upper limb deficiencies at approximately 6-8 months of age. In affected children with additional problems associated with motor function, such as limb paralysis, the age for initiating prosthetic therapy and the benefit of prostheses in promoting and expanding their motor function and activities is unknown. Case: In this case presentation, we describe a 25-month-old boy with cerebral palsy and left unilateral congenital upper limb deficiency caused by congenital constriction band syndrome. The patient could stand with assistance and crawl on his hands and knees. However, he was unable to walk with assistance or to stand on his own. A forearm prosthesis with a passive hand was prescribed and issued, and rehabilitation therapy for wearing and using the prosthesis was performed. At 34 months of age, the patient was able to walk forward using a walker with the prosthesis. Without the prosthesis, he still could not walk using a walker. The upper limb prosthesis also improved other movements such as sitting, standing, and tasks performed on a desk or on the floor. Discussion: The prosthesis was apparently effective in improving motor function. Prosthesis prescription should be considered at an appropriate and early age considering individual developmental stages and needs, regardless of the existence of additional problems associated with motor function.

PMID: 33768185

### 2. Risk factors for hip dislocation in dyskinetic cerebral palsy

Kyoko Okuno, Yukihiro Kitai, Toru Shibata, Hiroshi Arai

J Orthop Surg (Hong Kong). Jan-Apr 2021;29(1):23094990211001196. doi: 10.1177/23094990211001196.

Purpose: To investigate the risk factors for hip displacement in patients with dyskinetic cerebral palsy (DCP). Methods: We evaluated 81 patients with DCP, 45 males and 36 females, aged 10-22 years, risk factors for hip displacement were evaluated using multivariate logistic regression analysis with primary brain lesions, Gross Motor Function Classification System (GMFCS) level, gestational age, birth weight, Cobb's angle, and complication of epilepsy as independent factors. Hip displacement was defined as migration percentage >30%. Primary brain lesions were classified into globus pallidus (GP), thalamus and putamen (TP), and others using brain magnetic resonance imaging (MRI). Perinatal and clinical features were compared between patients with GP lesions and those with TP lesions. Results: Hip displacement was observed in 53 patients (67%). Higher GMFCS levels ($p = 0.013$, odds ratio [OR] 2.6) and the presence of GP lesions ($p = 0.04$, OR 16.5) were independent risk factors for hip displacement. Patients with GP lesions showed significantly higher GMFCS levels, more frequent hip displacement, and lower gestational age and birth weight than those with TP lesions. Conclusion: Primary brain lesion location may be an important factor in predicting hip displacement among patients with DCP. Appropriate risk assessment using brain MRI may contribute to the early detection and intervention of hip displacement because brain lesion location can be assessed during infancy before GMFCS level is decided.

PMID: 33745368

### 3. Dystonia assessment in children with cerebral palsy and periventricular leukomalacia

Ioanna Papadimitriou, Zoi Dalivigka, Chrysa Outsika, Nikolaos Scarmeas, Roser Pons

Eur J Paediatr Neurol. 2021 Mar 11;32:8-15. doi: 10.1016/j.ejpn.2021.03.005. Online ahead of print.

Objective: To describe the frequency, motor phenotype, clinical patterns and functional consequences of dystonia in patients with cerebral palsy (CP) in the setting of periventricular leukomalacia. Methods: Retrospective analysis of a cohort of 31 patients with CP and periventricular leukomalacia. Gross Motor Function Classification System (GMFCS) and Manual Ability Classification System (MACS) were used to classify functional ability. Spasticity was rated using the Modified Ashworth Scale. Presence of dystonia was assessed by reviewing video recordings, and its severity by using the Burke-Fahn-Marsden Dystonia Rating Scale. Results: All patients showed evidence of dystonia involving upper and/or lower limbs, neck, trunk, mouth and eyes in order of frequency. In 29% of patients dystonia involved only the limbs and in 71% it was multifocal. Dystonia severity ranged from slight to severe. Severity and distribution of dystonia did not correlate with gender, age, weeks of gestation or duration of neonatal unit stay. GMFCS and MACS correlated with dystonia but not with spasticity. Conclusions: Severity of dystonia, but not spasticity is associated with the severity of motor functional disability in CP patients with periventricular leukomalacia and demonstrates the key role of dystonia in the motor function of these patients.

PMID: 33743389

### 4. Abducted Standing in Children With Cerebral Palsy: Effects on Hip Development After 7 Years

Caroline Martinsson, Kate Himmelmann

Pediatr Phys Ther. 2021 Apr 1;33(2):101-107. doi: 10.1097/PEP.0000000000000789.

Purpose: The effects of abducted standing on hip migration percentage (MP) and range of hip and knee motion in children with cerebral palsy were studied for more than 7 years in 2 case-control groups. One group had adductor-psoas tenotomy and the other did not. Methods: Case participants performed 15° to 30° of abduction per hip. Controls performed 0° to 10° of abduction and were matched on age, surgery, and follow-up time. Results: Median MP decreased 7.0% in the participants without surgery, increased 6.5% in their matched participant, and decreased 3.5% in the participants who had adductor-psoas tenotomy. Migration percentage decreased 18% in participants who after surgery performed abducted standing. Range of motion was greater after surgery in participants who performed standing abduction. Conclusions: Standing abduction at 15° to 30° for 10 h/wk reduces MP and preserves range of motion for up to 7 years. This is a new contribution to research for children with cerebral palsy.

PMID: 33770798

### 5. Commentary on "Abducted Standing in Children With Cerebral Palsy: Effects on Hip Development After 7 Years"

Christiaan Gmelig Meyling, Liesbeth van Vulpen

Pediatr Phys Ther. 2021 Apr 1;33(2):108. doi: 10.1097/PEP.0000000000000788.

PMID: 33770799

### 6. Progression of Postural Asymmetry in Young Adults With Cerebral Palsy Who Are Not Walking: An Exploratory

### Study

Carlee Holmes, Kim Brock, Prue Morgan

Pediatr Phys Ther. 2021 Apr 1;33(2):E94-E98. doi: 10.1097/PEP.0000000000000787.

Purpose: Young adults with cerebral palsy (CP) who are not walking are at risk of developing or increasing musculoskeletal asymmetries affecting the rib cage, spine, pelvis, and hips. This longitudinal study aimed to explore postural change using the Goldsmith Indices of Body Symmetry (GIofBS) over an 18-month period in adults with CP who are not walking. Methods: Demographic and medical data were accessed from participant's history. Posture was recorded using the GIofBS to collect data during an 18-month period following skeletal maturity. Results: All participants had postural asymmetry at study onset with evidence of minimal change in some GIofBS outcome measures and fluctuations in other outcomes over 18 months. Conclusions: Physical therapists may use the GIofBS across the lifespan to screen for deterioration in musculoskeletal status or in assessing longer-term outcomes of interventions impacting posture in this complex population. Video abstract: For more insights from the authors, see Supplemental Digital Content 1, available at: http://links.lww.com/PPT/A314.

PMID: 33770801

### 7. Systematic Evaluation of the Effect of Rehabilitation of Lower Limb Function in Children with Cerebral Palsy Based on Virtual Reality Technology

Chao Gao, Yongli Wu, Junting Liu, Runhan Zhang, Manting Zhao

J Healthc Eng. 2021 Mar 8;2021:6625604. doi: 10.1155/2021/6625604. eCollection 2021.

As times go by, people's standard of living is constantly improving, but the medical pressure exerted by children with cerebral palsy is also increasing and the recovery of lower limb function in children with cerebral palsy after treatment has also gets people's attention. Of course, the relevant medical equipment is constantly improving, especially the appearance of virtual reality technology, which has played an extremely important role in restoring the lower extremities of children with cerebral palsy. In order to study the role that virtual reality technology can play in children with cerebral palsy, this article collects relevant information, builds a case template, and uses comprehensive quantification by investigating patients, examining relevant documents, and interviewing professionals. With qualitative analysis, a damage assessment matrix is created. Experimental results prove that the use of virtual reality technology can improve the treatment efficiency of children with cerebral palsy by more than 30%. In terms of the rehabilitation effect of children's lower limb function, virtual reality technology is more targeted for the treatment of children with far-reaching effects. It is much higher than traditional treatment methods, and the mobility of the children's lower limbs is restored by more than 80%. This shows that virtual reality technology can play an important role in the rehabilitation of the lower limbs of children with cerebral palsy.

PMID: 33763193

### 8. Dynamic muscle-tendon length following zone 2 calf lengthening surgery in two populations with equinus gait: Idiopathic Toe Walkers and Cerebral Palsy

Alexis Brierty, Henry Patrick John Walsh, Paula Jeffries, David Graham, Sean Horan, Chris Carty

Clin Biomech (Bristol, Avon). 2021 Mar 15;84:105323. doi: 10.1016/j.clinbiomech.2021.105323. Online ahead of print.

Background: Two populations commonly presenting with equinus gait are Idiopathic Toe-Walkers and children with Cerebral Palsy. Surgical intervention to treat equinus is defined by three zones. Zone three surgery, performed at the Achilles tendon, is most commonly used clinically. There is however, evidence from simulation studies that zone two surgery, performed at the muscle belly, might provide better functional outcomes. The purpose of this study was to investigate the effect of zone two calf -lengthening on post-operative gait in these populations. Methods: A retrospective audit of the Queensland Children's Motion Analysis Service database identified 17 toe-walkers (mean age 10.13 (SD 2.625)) and 11 Cerebral Palsy (mean age 9.72 (SD 4.04)) participants that received calf-lengthening surgery for plantarflexion contracture and had pre- and post-surgery 3D gait analysis. Inverse kinematics, dynamics, and muscle analysis were performed in OpenSim (v3.3) using a modified gait2392 model. Pre to post-surgery comparisons were performed in MATLAB using statistical parametric mapping. Dependent variables included ankle kinematics, powers and muscle-tendon length estimates. Findings: The primary outcome of this study was that ankle dorsiflexion increased in both Idiopathic Toe Walking and Cerebral Palsy groups post-calf lengthening across 90% and 85% of the gait cycle respectively. There was an increase in modelled muscle-tendon lengths, specifically in the medial gastrocnemius, of 78% (toe-walkers), and 100% (Cerebral Palsy) of the gait cycle. Power generation during push-off was not affected. Interpretation: Overall, the results appear to support the efficacy of zone 2 calf-lengthening for children with Cerebral Palsy and Idiopathic Toe Walking.

PMID: 33770533

### 9. Perceived Limitations of Walking in Individuals With Cerebral Palsy

Jennifer A Nelson, Elizabeth R Boyer

Phys Ther. 2021 Mar 24;pzab102. doi: 10.1093/ptj/pzab102. Online ahead of print.

Objective: Impairments associated with cerebral palsy (CP) can affect gait quality and limit activity and participation. The purpose of this study was to quantify (1) which of 6 factors (pain, weakness, endurance, mental ability, safety concerns, balance) were perceived to limit walking ability the most in individuals with CP and (2) whether age or Gross Motor Function Classification System (GMFCS) level is related to that perception. Methods: This cross-sectional study queried data from a gait laboratory database. Perceived walking limitation was quantified using a 5-point Likert scale ("never" to "all of the time"). Included were 1566 children and adults (mean age = 10.9 y [SD = 6.8]; range = 3.0-72.1 y) with CP (GMFCS level I: 502; II: 564; III: 433; IV: 67). Results: Patients or caregivers perceived balance to limit walking ability to the greatest extent, followed by endurance, weakness, safety, mental ability, and pain. Balance was perceived to always limit walking ability in 8%, 22%, 30%, and 34% of individuals in GMFCS levels I through IV, respectively. Endurance was perceived to always limit walking ability in 5%, 13%, 13%, and 27% of individuals in GMFCS levels I through IV, respectively. There were minor differences in the perceived extent of limitation caused by the factors by GMFCS level. Only weak associations between age and pain, mental ability, safety, and balance were observed (Spearman rho = -0.13 to 0.24). Conclusion: Patients or caregivers perceived decreased balance and endurance to most strongly limit walking ability. Efforts should be made to clinically track how both perceived and objective measures of these limiting factors change with age and intervention. Impact: Following a patient-and family-centered care model, therapy that places greater emphasis on balance and cardiovascular endurance may have the greatest effect on walking ability for individuals with CP. Future research should quantify which therapeutic, surgical, and pharmacologic interventions minimize these impairments and optimize activity and participation. Lay summary: Balance and endurance are perceived to be the greatest factors limiting walking in people with CP. If you have CP, your physical therapist might emphasize balance and cardiovascular endurance to improve your walking ability.

PMID: 33764475

### 10. The effect of combined transcranial pulsed current stimulation and transcutaneous electrical nerve stimulation on lower limb spasticity in children with spastic cerebral palsy: a randomized and controlled clinical study

Zhenhuan Liu, Shangsheng Dong, Sandra Zhong, Fang Huang, Chuntao Zhang, Yuan Zhou, Haorong Deng

BMC Pediatr. 2021 Mar 24;21(1):141. doi: 10.1186/s12887-021-02615-1.

Background: In the current study, we applied a combination of non-invasive neuromodulation modalities concurrently with multiple stimulating electrodes. Specifically, we used transcranial pulsed current stimulation (tPCS) and transcutaneous electrical nerve stimulation (TENS) as a novel strategy for improving lower limb spasticity in children with spastic cerebral palsy (SCP) categorized on levels III-V of the Gross Motor Function Classification System (GMFCS) with minimal side effects. Methods: Sixty-three SCP children aged 2-12 years, who were classified on levels III-V of the GMFCS were randomly assigned to one of two groups, resulting in 32 children in the experimental group and 31 children in the control group. The experimental group underwent a combination therapy of tPCS (400 Hz, 1 mA cerebello-cerebral stimulation) and TENS (400 Hz, max 10 mA) for 30 min, followed by 30 min of physiotherapy five times per week for 12 weeks. The control group underwent physiotherapy only 30 mins per day five times per week for 12 weeks. In total, all groups underwent 60 treatment sessions. The primary outcome measures were the Modified Ashworth Scale (MAS) and Modified Tardieu Scale (MTS). Evaluations were performed 3 days before and after treatment. Results: We found a significant improvement in MAS and MTS scores of the lower limbs in the experimental group compared to the control group in the hip adductors (Left: $p = 0.002$; Right: $p = 0.002$), hamstrings (Left: $p = 0.001$; Right: $p < 0.001$, and gastrocnemius (Left: $p = 0.001$; Right: $p = 0.000$). Moreover, MTS scores of R1, R2 and R2-R1 in left and right hip adduction, knee joint, and ankle joint all showed significant improvements ($p \leq 0.05$). Analysis of MAS and MTS scores compared to baseline scores showed significant improvements in the experimental group but declines in the control group. Conclusion: These results are among the first to demonstrate that a combination of tPCS and TENS can significantly improve lower limb spasticity in SCP children classified on GMFCS levels III-V with minimal side effects, presenting a novel strategy for addressing spasticity challenges in children with severe SCP. Trial registration: ChiCTR.org, ChiCTR1800020283, Registration: 22 December 2018 (URL: http://www.chictr.org.cn/showproj.aspx?proj=33953 ).

PMID: 33761932

**11. Measurement of physical activity in children and adolescents with cerebral palsy: a scoping review protocol**
Mette Johansen, Helle M Rasmussen, Katarina Lauruschkus, Britt Laugesen

JBI Evid Synth. 2021 Mar 23. doi: 10.11124/JBIES-20-00283. Online ahead of print.

Objective: The objective of the scoping review outlined in this protocol is to identify and map subjective instruments that have been developed for children and adolescents with cerebral palsy from birth to 18 years to measure physical activity and sedentary behavior. Introduction: Children and adolescents with cerebral palsy often do not meet recommended levels of physical activity, although regular physical activity is an important determinant of good health. Considering the importance of physical activity, there is a need for systematic collection of data on daily activity and sedentary behavior of children and adolescents with cerebral palsy. Subjective instruments with clinical utility for use in quality registers are needed for evaluation of interventions and to investigate the relationship between activity and health outcomes. Inclusion criteria: This review will consider studies that include children and adolescents from birth to 18 years with cerebral palsy across levels I to V of the Gross Motor Function Classification System. Specifically, this scoping review will report on subjective instruments used to measure physical activity and sedentary behavior and that distinguish between physical activity performance and physical activity capability. Methods: This scoping review will be conducted in accordance with the JBI methodology for scoping reviews and will search the following databases: PubMed, CINAHL, Web of Science, Cochrane Database of Systematic Reviews, JBI Evidence Synthesis, Embase, and PEDro, as well as specific journals relating to physical activity assessment.

PMID: 33769334

**12. Describing the Delivery of Evidence-Based Physical Therapy Intervention to Individuals With Cerebral Palsy**
Amy F Bailes, Kelly Greve, Jason Long, Brad G Kurowski, Jilda Vargus-Adams, Bruce Aronow, Alexis Mitelpunkt

Pediatr Phys Ther. 2021 Apr 1;33(2):65-72. doi: 10.1097/PEP.0000000000000783.

Purpose: To characterize by evidence grades and examine variation in type of physical therapy intervention delivered in routine clinical care in individuals with cerebral palsy (CP). Methods: Retrospective data collection from the electronic record over 1 year at a tertiary care pediatric outpatient therapy division. Results: Four hundred sixty-five individuals with CP received 28 344 interventions during 4335 treatment visits. Sixty-six percent of interventions were evidence-based interventions (EBIs). Significant variation was demonstrated across Gross Motor Function Classification System levels, with children classified as level V receiving the least and level III the most. The most frequent EBIs delivered were caregiver education, motor control, functional strengthening, ankle-foot orthoses, treadmill training, and fit of adaptive equipment. Conclusions: Further work is needed to determine whether amount of EBI is related to better outcomes. Combining this information with other aspects of dose (intensity, time, and frequency) may elucidate the contribution of each with outcomes.

PMID: 33770793

**13. Commentary on "Describing the Delivery of Evidence-Based Physical Therapy Intervention to Individuals With Cerebral Palsy"**
Jennifer B Christy, J Adam Knott

Pediatr Phys Ther. 2021 Apr 1;33(2):73. doi: 10.1097/PEP.0000000000000792.

PMID: 33770794

### 14. Parent-delivered interventions used at home to improve eating, drinking and swallowing in children with neurodisability: the FEEDS mixed-methods study

Jeremy Parr, Lindsay Pennington, Helen Taylor, Dawn Craig, Christopher Morris, Helen McConachie, Jill Cadwgan, Diane Sellers, Morag Andrew, Johanna Smith, Deborah Garland, Elaine McColl, Charlotte Buswell, Julian Thomas, Allan Colver

Health Technol Assess. 2021 Mar;25(22):1-208. doi: 10.3310/hta25220.

Background: Eating, drinking and swallowing difficulties are common in young children with neurodisability. These difficulties may lead to inadequate calorie intake, which affects a child's nutrition, growth and general physical health. Objective: To examine which interventions are available that can be delivered at home by parents to improve eating, drinking and swallowing in young children with neurodisability and are suitable for investigation in pragmatic trials. Design: This was a mixed-methods study that included focus groups, surveys, an update of published systematic reviews of interventions, a systematic review of measurement properties of existing tools, evidence mapping, evidence synthesis, a Delphi survey and stakeholder workshops. Setting: The study was carried out in NHS hospitals, community services, family homes and schools. Participants: Parents of children who had neurodisability and eating, drinking and swallowing difficulties. Professionals from health and education. Young people with eating, drinking and swallowing difficulties or young people who had previously experienced eating, drinking and swallowing difficulties. Data sources: Literature reviews; national surveys of parents and professionals; focus groups with parents, young people and professionals; and stakeholder consultation workshops. Review methods: An update of published systematic reviews of interventions (searched July-August 2017), a mapping review (searched October 2017) and a systematic review of measurement properties using COnsensus-based Standards for the Selection of health status Measurement INstruments (COSMIN) methodology (searched May 2018). Results: Significant limitations of the available research evidence regarding interventions and tools to measure outcomes were identified. A total of 947 people participated: 400 parents, 475 health professionals, 62 education professionals and 10 young people. The survey showed the wide range of interventions recommended by NHS health professionals, with parents and professionals reporting variability in the provision of these interventions. Parents and professionals considered 19 interventions as relevant because they modified eating, drinking and swallowing difficulties. Parents and professionals considered 10 outcomes as important to measure (including Nutrition, Growth and Health/safety); young people agreed that these were important outcomes. Stakeholder consultation workshops identified that project conclusions and recommendations made sense, were meaningful and were valued by parents and professionals. Parents and health professionals were positive about a proposed Focus on Early Eating, Drinking and Swallowing (FEEDS) toolkit of interventions that, through shared decision-making, could be recommended by health professionals and delivered by families. Limitations: The national surveys included large numbers of parents and professionals but, as expected, these were not representative of the UK population of parents of children with eating, drinking and swallowing difficulties. Owing to the limitations of research evidence, pragmatic decisions were made about interventions that might be included in future research and outcomes that might be measured. For instance, the reviews of research found only weak or poor evidence to support the effectiveness of interventions. The review of outcome measures found only limited low-level evidence about their psychometric properties. Conclusions: Opportunities and challenges for conducting clinical trials of the effectiveness of the FEEDS toolkit of interventions are described. Parents and professionals thought that implementation of the toolkit as part of usual NHS practice was appropriate. However, this would first require the toolkit to be operationalised through development as a complex intervention, taking account of constituent interventions, delivery strategies, implementation and manualisation. Subsequently, an evaluation of its clinical effectiveness and cost-effectiveness could be undertaken using appropriate research methods. Future work: Initial steps include FEEDS toolkit development and evaluation of its use in clinical practice, and identification of the most robust methods to measure valued outcomes, such as Nutrition and Growth. Trial registration: Current Controlled Trials ISRCTN10454425. Funding: This project was funded by the National Institute for Health Research (NIHR) Health Technology Assessment programme and will be published in full in Health Technology Assessment; Vol. 25, No. 22. See the NIHR Journals Library website for further project information.

PMID: 33769272

### 15. Boy with cerebral palsy and severe malnutrition: Do not miss the mealtime!

Chiara Udina, Elisabetta Cattaruzzi, Egidio Barbi

J Paediatr Child Health. 2021 Mar 24. doi: 10.1111/jpc.15449. Online ahead of print.

PMID: 33760327

### 16. Development and Psychometric Properties of the Arabic Parent Nutritional Assessment Scale (A-PNAS) for Children with Developmental Disabilities

Nihad A Almasri, Carl J Dunst, Maysoun Saleh, Reema Tayyem

Phys Occup Ther Pediatr. 2021 Mar 24;1-16. doi: 10.1080/01942638.2021.1900490. Online ahead of print.

Aims: To describe the development of the Arabic-Parent Nutritional Assessment Scale (A-PNAS), and to examine construct and known-group discriminant validity, internal consistency, and test-retest reliability of the A-PNAS.Methods: A cross-sectional design was used. Participants were 130-children with CP (mean age = 4.26 ± 3.29 years) who were registered in the national CP registry of Jordan with a matching group of 130-children with typical development (mean age = 4.65 ± 3.54 years). Parents completed the developed A-PNAS through a structured phone interview. Parents of children with CP confirmed their child's level of gross motor function classification system.Results: Exploratory factor analysis identified three subscales in the A-PNAS: Food Intake Problems, Health Problems, and Behavioral Problems which explained 31.6% of the variance in nutritional problems of children with CP. Cronbach's alpha indicated acceptable internal consistency for Food Intake ($\alpha = 0.61$) and Health Problems ($\alpha = 0.67$)subscales. Parents of children with CP reported that their children had more food intake, health, and behavioral problems compared to children with typical development ($p<.001$). Test-retest reliability was excellent for the subscales of the A-PNAS (ICCs = 0.96, 0.98, 0.96).Conclusions: The findings provide support for the face validity, construct validity, internal consistency, Known-Groups discriminant validity, and test-retest reliability of the A-PNAS.

PMID: 33761817

### 17. Importance of plasma ghrelin levels with special reference to nutritional metabolism and energy expenditure in pediatric patients with severe motor and intellectual disabilities

Masahiro Zenitani, Hiroshi Hosoda, Satoko Nose, Kenji Kangawa, Hisayoshi Kawahara, Takaharu Oue

Clin Nutr ESPEN. 2021 Apr;42:180-187. doi: 10.1016/j.clnesp.2021.01.043. Epub 2021 Feb 10.

Background & aims: Nutritional metabolism is complex in pediatric patients with severe motor and intellectual disability (SMID), and therefore, appropriate estimation of the energy requirements is difficult. Focusing on ghrelin's role in energy metabolism regulation, we investigated plasma ghrelin levels in pediatric SMID patients and analyzed its nutritional significance as a regulatory marker of energy reserve. Methods: Fasting plasma total, acyl, and des-acyl ghrelin levels in 40 patients with SMID, including cerebral palsy (CP) ($n = 20$) and muscular disease (MD) ($n = 8$), and healthy controls ($n = 13$) were investigated. The correlations of plasma ghrelin levels with anthropometry, blood nutritional markers, energy intake, and resting energy expenditure (REE) measured with indirect calorimetry were analyzed. A p value $< 0.05$ was considered significant. Results: SMID patients had significantly higher acyl ghrelin, and lower body mass index (BMI), z-scores of body weight (BW), body height and BMI, and albumin than controls. CP patients had significantly higher total and acyl ghrelin, z-score of the mid-upper arm circumference (MUAC), retinol-binding protein, transthyretin, creatinine, and glucose than MD patients. Total and acyl ghrelin in CP patients and des-acyl ghrelin in MD patients had significant negative correlations with MUAC and upper arm fat area. In CP patients, total and acyl ghrelin had significant positive correlations with REE/BW (kcal/kg), and total ghrelin was predictive of REE/BW ($r2 = 0.625$, $p < 0.0001$). Conclusions: An increase in acyl ghrelin observed in SMID patients possibly indicates energy reserve deficiency. In CP patients, total and acyl ghrelin inversely reflected total body fat mass, resulting in strongly positive correlations with REE/BW. The measurement of plasma ghrelin may be useful to assess nutritional metabolism and energy reserve in pediatric SMID patients, such as CP and MD patients.

PMID: 33745575

### 18. The relationship between gross motor function impairment in cerebral palsy and sleeping issues of children and caregivers

Andrea Obrecht, Marcela Fischer de Almeida, Luciana Maltauro, Weslei Douglas Leite da Silva, Marise Bueno Zonta, Ana Chrystina de Souza Crippa

Sleep Med. 2021 Mar 4;81:261-267. doi: 10.1016/j.sleep.2021.02.055. Online ahead of print.

Aim: To investigate, among children and adolescents with cerebral palsy (CP), the relationship between impairment of the gross motor function and: (i) child sleep disorders; (ii) the need for nocturnal support; and (iii) the quality of sleep of their caregivers. Methods: For children, we considered their scores on the gross motor function measure (GMFM-88) and on the sleep disturbance scale for children (SDSC), besides analyzing qualitative features about their sleep. For caregivers, we considered their scores in the Pittsburgh sleep quality index (PSQI). Results: Our sample was comprised of 87 participants with mean age of 11.4 years old (±3.4). We observed correlations between GMFM-88 and disorders of initiating and maintaining sleep (DIMS) (r = -0.22; p = 0.039), sleep-wake transition disorders (SWTD) (r = 0.26; p = 0.017) and disorders of arousal (DA) (r = 0.23; p = 0.033). Children receiving nocturnal support presented lower scores in the GMFM-88 (p = 0.001) and higher scores in the SDSC (p = 0.029). For the caregivers, we found no correlation between GMFM-88 and PSQI. Nonetheless, their PSQI scores correlated with the SDSC scores (r = 0.24; p = 0.027). Conclusion: Impairment of the gross motor function correlated with DIMS and the need for nocturnal support but might not have an impact on the caregivers' sleep, which in turn correlated with child sleep disorders.

PMID: 33743473

### 19. Perceived Usefulness of a Social Robot Augmented Telehealth Platform by Therapists in the United States

Michael J Sobrepera, Vera G Lee, Suveer Garg, Rochelle Mendonca, Michelle J Johnson

IEEE Robot Autom Lett. 2021 Apr;6(2):2946-2953. doi: 10.1109/lra.2021.3062349. Epub 2021 Feb 25.

With the shortage of rehabilitation clinicians in rural areas and the ongoing COVID-19 pandemic, remote rehabilitation (telerehab) fills an important gap in access to rehabilitation, especially for the treatment of adults and children experiencing upper arm disability due to stroke and cerebral palsy. We propose the use of a socially assistive robot with arms, a torso, and a face to play games with and guide patients, coupled with a telepresence platform, to maintain the patient-clinician interaction, and a computer vision system, to aid in automated objective assessments, as a tool for achieving more effective telerehab. In this paper, we outline the design of such a system, Lil'Flo, and present a uniquely large perceived usefulness evaluation of the Lil'Flo platform with 351 practicing therapists in the United States. We analyzed responses to the question of general interest and 5 questions on Lil'Flo's perceived usefulness. Therapists believe that Lil'Flo would significantly improve communication, motivation, and compliance during telerehab interactions when compared to traditional telepresence. 27% of therapists reported that they were interested in using Lil'Flo. Therapists interested in using Lil'Flo perceived it as having significantly higher usefulness across all measured dimensions than those who were not interested in using it.

PMID: 33748417

### 20. A hinge-free, non-restrictive, lightweight tethered exosuit for knee extension assistance during walking

Evelyn J Park, Tunc Akbas, Asa Eckert-Erdheim, Lizeth H Sloot, Richard W Nuckols, Dorothy Orzel, Lexine Schumm, Terry D Ellis, Louis N Awad, Conor J Walsh

IEEE Trans Med Robot Bionics. 2020;2(2):165-175. doi: 10.1109/tmrb.2020.2989321. Epub 2020 Apr 21.

In individuals with motor impairments such as those post-stroke or with cerebral palsy, the function of the knee extensors may be affected during walking, resulting in decreased mobility. We have designed a lightweight, hinge-free wearable robot combining soft textile exosuit components with integrated rigid components, which assists knee extension when needed but is otherwise highly transparent to the wearer. The exosuit can apply a wide range of assistance profiles using a flexible multi-point reference trajectory generator. Additionally, we implemented a controller safety limit to address the risk of hyperextension stemming from the hinge-free design. The exosuit was evaluated on six healthy participants walking uphill and downhill on a treadmill at a 10° slope with a set of joint power-inspired assistance profiles. A comparison of sagittal plane joint angles between no exosuit and exosuit unpowered conditions validated the device transparency. With positive power assistance, we observed reduction in average positive knee biological power during uphill walking (left: 17.5 ± 3.21%, p = 0.005; right: 23.2 ± 3.54%, p = 0.008). These initial findings show promise for the assistive capability of the device and its potential to improve the quality of gait and increase mobility in clinical populations.

PMID: 33748694

### 21. Reliability and sources of variability of 3D kinematics and electromyography measurements to assess newly-acquired gait in toddlers with typical development and unilateral cerebral palsy

Anca Irina Grigoriu, Sylvain Brochard, Morgan Sangeux, Liliana Padure, Mathieu Lempereur

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The aim was to 1) determine intersession and intertrial reliability and 2) assess three sources of variability (intersubject, intersession and intertrial) of lower limb kinematic and electromyographic (EMG) variables during gait in toddlers with typical development (TD) and unilateral cerebral palsy (UCP) (age <3 years, independent walking experience ≤6 months). Gait kinematics and surface EMG were recorded in 30 toddlers (19 TD and 11 UCP), during two, 3D-motion capture sessions. Standard error of measurement (SEM) between trials (gait cycles) of the same session and between sessions was calculated to assess reliability. Standard deviations (SD) between subjects, sessions and trials were calculated to estimate sources of variability. Sixty-four percent of kinematic SEM-values were acceptable (2°-5°). Frontal plane measurements were most reliable (SEM 2°-4.6°). In toddlers with UCP, EMG variables were most reliable for affected side, distal muscles. Intrinsic (intertrial and intersubject) variability was high, reflecting both motor immaturity and the high variability of toddler gait patterns. In toddlers with UCP, variability was amplified by motor impairment and delayed motor development. 3D gait analysis and surface EMG are partially reliable tools to study individual gait patterns in toddlers in clinical practice and research, although some variables must be interpreted with caution.

PMID: 33761385

### 22. The Journey of Humanizing Care for People With Disabilities

Lauren Clark, Cherisse Watts

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The Max Bramer keynote address at the annual Special Care Dentistry Association (SCDA) conference provided an opportunity for reflection on our journey as health care professionals to humanize care for all people, and particularly those who are disabled. It is important to review the history of disability and discrimination to understand the progress made to date, and then consider the array of steps possible in further refining the humanistic dimensions of dental care. The typologies of tourist, traveler, adventurer, and scout were formulated to consider varieties of humanized care found within articles published in Special Care in Dentistry. Considering exemplars for each of these four typologies leads to reflections about how dentistry can embrace understanding, improve training of new dental professionals, tailor care for people with disabilities, and advocate for equity.

PMID: 33745377

### 23. Exploring participation in family and recreational activities among children with cerebral palsy during early childhood: how does it relate to motor function and parental empowerment?

Runa Kalleson, Reidun Jahnsen, Sigrid Østensjø

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Purpose: To explore participation in real-life activities during early childhood, compare children's participation based on motor function and investigate relationships between participation and parental empowerment. Methods: Data derived from the Cerebral Palsy Follow-up Program (CPOP) in Norway and the research registry Habilitation Trajectories, Interventions, and Services for Young Children with CP (CPHAB). Fifty-six children (12-56 months, GMFCS levels I-IV, MACS levels I-V) and their families were included. Frequency and enjoyment of participation were assessed by the Child Engagement in Daily Life Questionnaire and parental empowerment in family and service situations by the Family Empowerment Scale at least twice during the preschool years. Differences between groups based on motor function were explored by the Kruskal-Wallis tests. A linear mixed model was conducted to explore relationships between child participation and parental empowerment. Results: Similarities and differences in participation between children at different motor function levels varied between the activities explored. Fluctuations in frequency and stable enjoyment scores over time were most common. A statistically significant relationship was revealed between child participation and parental empowerment in family situations, but not in service situations. Conclusions: Child participation appears as context-dependent and complexly influenced by both motor function and parental empowerment. This supports a focus on transactional processes when exploring and promoting child participation.Implications for RehabilitationFamily and recreational activities represent real-life contexts providing opportunities for interactions and experiences supporting development and learning.Children with CP appreciate a wide range of activities in the home and community, which emphasizes the importance of providing opportunities for such participation in order to fulfill their desires and interests.Child participation appears as complexly influenced by the unique activity setting, motor function and characteristics of the family environment, requiring attention to transactional processes when aiming to explore and promote participation.

PMID: 33749471

### 24. Effects of equine-assisted activities on attention and quality of life in children with cerebral palsy in a randomized trial: examining the comorbidity with attention-deficit/hyperactivity disorder

Bomi Ahn, Yoo-Sook Joung, Jeong-Yi Kwon, Dong Ik Lee, Soohwan Oh, Byoung-Uk Kim, Jung Yoon Cha, Ji-Hae Kim, Ji Young Lee, Hye Yeon Shin, Yun Sik Seo

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Background: Attention problems and decreased quality of life are frequently accompanied in Cerebral Palsy (CP), which can negatively affect rehabilitation of physical disability. However, the majority of affected children remain untreated in the aspects of attention or psychosocial factors. Equine-Assisted Activities and Therapies (EAAT) use horse as a therapeutic modality including grooming as well as mounted riding activities in which patients exercise and experience mounted stimulation. It is known to help improve attention in children with ADHD, so that it can be an exercise therapy that is expected to improvement of attention as well as rehabilitating effects in CP patients. EAA may be a promising strategy to address the unmet need for CP patients. This study aims to investigate the efficacy of EAA for children with CP, those with both CP and ADHD and confirm the comorbidity between CP and ADHD. Methods: Forty-six children with cerebral palsy participated in this study. For the exercise group, they participated in a 40-min session twice a week for a 16-week period, while the control group engaged in daily life without any special treatments. Each children individually were assessed on attention and psychological wellbeing at baseline and post-treatment. Comorbidity were identified based on the Diagnostic and Statistical Manual of Mental Disorder 5th edition (DSM-5) and confirmed by Korean Kiddie-Schedule for Affective Disorders and Schizophrenia Present and Lifetime Version (K-SADS-PL). Results: Perseveration rated using the Conner's Performance Test (CPT) showed a significant decrease only in the exercise group ($p < .024$). However, no significant improvement in children's quality of life was observed after EAA program compared with control group. Among the total participants, fifteen children (31.91%) were diagnosed with ADHD. When conducting an additional analysis with the subsample of CP patients diagnosed with ADHD, the d', commission error and perseveration showed a significant decrease only in the exercise group. Children with CP and ADHD reported an improvement in quality of life both in exercise and control group, but only in the exercise group social functioning exhibited a significant difference. Conclusion: The positive effects of the EAA on attention and quality of life were confirmed. Children with CP in the exercise group were more capable to sustain their attention longer. Those with CP and ADHD showed an increase in attention and perceived to have better social skills after receiving 16 weeks of EAA compared to those in the control group. Considering high comorbidity of CP and ADHD, it seems that the EAA program could be the better alternative treatment for CP with attentional problem. The results of this study will contribute to growing evidence for the efficacy of EAA in children especially with CP and ADHD. Trial registration: This trial was registered on ClinicalTrials.gov ( NCT03870893 ). Registered 26 July 2017.

PMID: 33740922

### 25. Electronic health record and patterns of care for children with cerebral palsy

Brad G Kurowski, Kelly Greve, Amy F Bailes, Janet Zahner, Jilda Vargus-Adams, Mary A Mcmahon, Bruce J Aronow, Alexis Mitelpunkt

Dev Med Child Neurol. 2021 Mar 25. doi: 10.1111/dmcn.14867. Online ahead of print.

Aim: To characterize the patterns of care of children with cerebral palsy (CP) in a tertiary healthcare system. Method: Electronic health record data from 2009 to 2019 were extracted for children with CP. Machine learning hierarchical clustering was used to identify clusters of care. The ratio of in-person to care coordination visits was calculated for each specialty. Results: The sample included 6369 children with CP (55.7% males, 44.3% females, 76.2% white, 94.7% non-Hispanic; with a mean age of 8y 2mo [SD 5y 10mo; range 0-21y; median 7y 1mo]) at the time of diagnosis. A total of 3.7 million in-person visits and care coordination notes were identified across 34 specialties. The duration of care averaged 5 years 5 months with five specialty interactions and 21.8 in-person visits per year per child. Seven clusters of care were identified, including: musculoskeletal and function; neurological; high-frequency/urgent care services; procedures; comorbid diagnoses; development and behavioral; and primary care. Network analysis showed shared membership among several clusters. Interpretation: Coordination of care is a central element for children with CP. Medical informatics, machine learning, and big data approaches provide unique insights into care delivery to inform approaches to improve outcomes for children with CP.

PMID: 33768551

### 26. Cost Comparison of Botulinum Toxin Injections Versus Surgical Treatment in Pediatric Patients With Cerebral Palsy: A Markov Model

Gregory S Kazarian, Ann E Van Heest, Charles A Goldfarb, Lindley B Wall

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Purpose: The purpose of this study was to compare the cost-effectiveness of surgical release to botulinum toxin injections in the treatment of upper-extremity (UE) cerebral palsy (CP). Methods: A Markov transition-state model was developed to assess the direct and indirect costs as well as accumulated quality-adjusted life-years associated with surgery (surgery group) and continuous botulinum toxin injections (botulinum group) for the treatment of UE CP in children aged 7 to 12 years. Direct medical costs were obtained from institutional billing departments. The number of parental missed workdays associated with each treatment was estimated and previously published regressions were used to calculate indirect costs associated with missed work. Total costs, cost-effectiveness, and incremental cost-effectiveness ratios were calculated. Incremental cost-effectiveness ratios and willingness to pay thresholds were used to make decisions regarding society's willingness to pay for the incremental cost of each treatment given the incremental benefit. Results: The surgery group demonstrated lower direct, indirect, and total costs compared with the botulinum group. Direct costs were $29,250.50 for the surgery group and $50,596.00 for the botulinum group. Indirect costs were $9,467.46 for the surgery group and $44,428.60 for the botulinum group. Total costs were $38,717.96 for the surgery group and $95,024.60 for the botulinum group, a difference of $56,306.64. The incremental cost-effectiveness ratio was -$42,019.88, indicating that surgery is a less costly and more effective treatment and that botulinum injections fall outside the societal willingness to pay threshold. Excluding indirect costs associated with parental missed work during home occupational therapy did not have a significant impact on the model. Conclusions: Surgery is associated with lower direct, indirect, and total costs, as well as a greater number of accumulated quality-adjusted life-years. Surgery provides a greater benefit at a lower cost, which suggests that botulinum injections should be used sparingly in this population. Treatment with surgery could represent savings of $5.6 to $11.3 billion annually in the United States. Type of study/level of evidence: Economic/Decision Analysis II.

PMID: 33745764

### 27. Neurodevelopmental Outcomes of Infants with Bronchopulmonary Dysplasia

Sara B DeMauro

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Preterm infants with bronchopulmonary dysplasia (BPD), and particularly those who develop the most severe forms of chronic lung disease during the neonatal period, are at high risk for poor developmental outcomes throughout childhood. Infants who require mechanical ventilation at 36 weeks post-menstrual age have significantly increased odds for cerebral palsy, developmental delay at 2 years, and poor academic achievement and low IQ in adolescence. Over the past several decades, many therapies and care strategies, including steroids, CPAP, surfactant, and other medications have been introduced into clinical practice. These approaches have changed the epidemiology of BPD in very preterm infants. However, BPD remains common and strongly associated with poor development throughout childhood. Only caffeine has been proven to reduce BPD and improve childhood developmental outcomes. In future research, it will be essential to better understand the developmental sequelae of BPD beyond school age and to test interventions to improve developmental trajectories in this population. As new management strategies for BPD are developed, it will be essential to rigorously evaluate both short-term and long-term effects before they are introduced into routine neonatal practice. This article is protected by copyright. All rights reserved.

PMID: 33749169

### 28. Neurodevelopmental outcome of preterm infants enrolled in myo-inositol randomized controlled trial

Ira Adams-Chapman, Kristi L Watterberg, Tracy L Nolen, Shawn Hirsch, Carol A Cole, C Michael Cotten, William Oh, Brenda B Poindexter, Kristin M Zaterka-Baxter, Abhik Das, Conra Backstrom Lacy, Ann Marie Scorsone, Andrea F Duncan, Sara B DeMauro, Ricki F Goldstein, Tarah T Colaizy, Deanne E Wilson-Costello, Isabell B Purdy, Susan R Hintz, Roy J Heyne, Gary J Myers, Janell Fuller, Stephanie Merhar, Heidi M Harmon, Myriam Peralta-Carcelen, Howard W Kilbride, Nathalie L Maitre, Betty R Vohr, Girija Natarajan, Helen Mintz-Hittner, Graham E Quinn, David K Wallace, Richard J Olson, Faruk H Orge, Irena Tsui, Michael Gaynon, Amy K Hutchinson, Yu-Guang He, Timothy W Winter, Michael B Yang, Kathryn M Haider, Martin S Cogen, Denise Hug, Don L Bremer, John P Donahue, William R Lucas, Dale L Phelps, Rosemary D Higgins, Eunice Kennedy Shriver National Institute of Child Health and Human Development Neonatal Research Network

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Objective: This study evaluates the 24-month follow-up for the NICHD Neonatal Research Network (NRN) Inositol for Retinopathy Trial. Study design: Bayley Scales of Infants Development-III and a standardized neurosensory examination were performed in infants enrolled in the main trial. Moderate/severe NDI was defined as BSID-III Cognitive or Motor composite score <85, moderate or severe cerebral palsy, blindness, or hearing loss that prevents communication despite amplification were assessed. Results: Primary outcome was determined for 605/638 (95%). The mean gestational age was 25.8 ± 1.3 weeks and mean birthweight was 805 ± 192 g. Treatment group did not affect the risk for the composite outcome of death or survival with moderate/severe NDI (60% vs 56%, $p = 0.40$). Conclusions: Treatment group did not affect the risk of death or survival with moderate/severe NDI. Despite early termination, this study represents the largest RCT of extremely preterm infants treated with myo-inositol with neurodevelopmental outcome data.

PMID: 33758387