

# Cerebral palsy research news

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#### **Interventions and Management**

#### 1. Action Observation Training to Improve Upper Limb Function in Infants with Unilateral Brain Lesion - a Feasibility Study

Miriam von Gunten, Angela Botros, Bernadette Tobler-Ammann, Tobias Nef, Sebastian Grunt

Dev Neurorehabil. 2023 Mar 22;1-10. doi: 10.1080/17518423.2023.2193630. Online ahead of print.

In this study, the feasibility of Action Observation Training (AOT) in combination with sensor-based measurements in infants at high risk of Unilateral Spastic Cerebral Palsy (UCP) were evaluated. Over a four-week period, eight infants at high risk of UCP performed AOT at home while wearing sensors with assistance of caregivers. Sensor data were compared to clinical assessments of upper limb function, the Hand Assessment for Infants (HAI) and the Mini-Assisting Hand Assessment (Mini-AHA). AOT training time and acceptance by the caregivers were considered as indicators for feasibility. The excellent training adherence and positive feedback of the caregivers showed that the AOT was feasible in this patient group and setting. Sensor measurements were accepted and displayed significant correlations with hand function. These preliminary results indicate the potential of wearable sensors to record upper limb function over the course of AOT for infants at high risk of UCP. Thus, AOT in combination with sensor measurements are proposed as a feasible training tool to complement usual care.

PMID: 36949659

#### 2. Development of Hand Use with and Without Intensive Training Among Children with Unilateral Cerebral Palsy in Scandinavia

Gunvor L Klevberg, Manuela Zucknick, Reidun Jahnsen, Ann-Christin Eliasson

Dev Neurorehabil. 2023 Mar 22;1-9. doi: 10.1080/17518423.2023.2193256. Online ahead of print.

Aim: To describe hand use development in children with unilateral cerebral palsy who did/did not participate in constraint-induced movement therapy (CIMT) before 7 years of age. Method: The study included 334 participants (18 months-12 years) who were assessed with 1,565 Assisting Hand Assessments (AHAs) and categorized into no intensive training (NIT), CIMT (18 months-7 years), and Baby-CIMT (<18 months) groups. Results: AHA performance at 18 months (AHA-18) was positively associated with development regardless of training. The CIMT group had lower AHA-18 performance than the NIT group (p = .028), but higher stable limit (p = .076). The age when 90% of development was reached was highest in the CIMT group (p = .014). Although non-significant, the Baby-CIMT group had higher mean curve than NIT and CIMT combined (AHA-18 p = .459, limit p = .477). Conclusion: The CIMT group improved more over time than the NIT group. Intensive training extended the window of development, and Baby-CIMT might promote early development.

## 3. Can the Shriners Hospital Upper Extremity Evaluation (SHUEE) Detect Change in Dynamic Position and Spontaneous Function of the Upper Limb in People With Hemiplegic Cerebral Palsy?

Nancy Lennon, Chris Church, Thomas Shields, John Kee, John D Henley, Jose J Salazar-Torres, Tim Niiler, M Wade Shrader, Jennifer M Ty

J Pediatr Orthop. 2023 Mar 22. doi: 10.1097/BPO.000000000002403. Online ahead of print.

Background: The Shriners Hospital Upper Extremity Evaluation (SHUEE) is a video-based measure designed to assess upper extremity function in people with cerebral palsy (CP). The SHUEE completes both dynamic positional analysis (DPA; position during functional activities) and spontaneous functional analysis (spontaneous use of the involved limb). Although the SHUEE has been suggested as a measure for planning upper limb interventions and evaluating outcomes, limited evidence of its ability to detect change exists. Thus, this study aimed to describe responsiveness of the SHUEE to detect change after orthopaedic surgery. Methods: In this Institutional Review Board-approved retrospective cohort study, we identified children with CP who were administered SHUEE on≥2 encounters. We formed pairs of initial and follow-up visits between temporally adjacent visits. Pairs were assigned to a surgery or non-surgery group based on intervening upper limb orthopaedic surgery. We compared differences in baseline SHUEE scores between groups and differences in temporally adjacent SHUEE scores within groups using Welch unequal variances t tests and paired t tests, respectively. Results: Nineteen people (7 female) with hemiplegic CP had≥2 SHUEE assessments; Manual Ability Classification System levels I (3), II (8), III (7), IV (1); Gross Motor Function Classification System levels I (10), II (7), IV (2); mean age at baseline 11.9 (5.1 to 19.1) years; and follow-up at 13.4 (5.5 to 19.7) years. Six people had≥2 visits leading to 14 surgical pairs and 10 non-surgical pairs. At baseline, DPA of the wrist and forearm were significantly lower in the surgical group (P<0.05). At follow-up, no significant difference between the groups existed in DPA measures (P>0.05). After surgical intervention, there was a significant change in overall and wrist DPA (P<0.05). Conclusions: The DPA measures demonstrated responsiveness to expected positional changes in the arm after orthopaedic surgery in people with CP. The SHUEE was useful in identifying abnormal segmental alignment pre-surgically and documenting changes in alignment postoperatively. As orthopaedic surgery does not address limb neglect or bimanual ability, spontaneous functional analysis scores were as expected-unchanged. Level of evidence: Level III, retrospective cohort study.

PMID: 36952245

#### 4. CNS adverse effects of intrathecal baclofen treatment for spasticity in children with cerebral palsy and epilepsy: a case report

Marta De Rinaldis, Ivana Gallo, Maria Carmela Oliva, Antonio Trabacca

Neurol Sci. 2023 Mar 18. doi: 10.1007/s10072-023-06757-1. Online ahead of print.

No abstract available

PMID: 36933098

#### **5.** Is baclofen the least worst option for spasticity management in children? Joline E Brandenburg

J Pediatr Rehabil Med. 2023 Mar 15. doi: 10.3233/PRM-230001. Online ahead of print.

Baclofen is often considered a first-line treatment option for spasticity management in children. However, adverse effects, administration, and dosing can be barriers to effectiveness. In my practice, other medications for spasticity management are often used prior to initiating baclofen. In this article, baclofen use for spasticity management in children is briefly reviewed along with discussion of approaches using other medications as first-line treatment options. I will present a rationale for medication selection for spasticity management and discuss the approaches I take in medication selection that incorporate spasticity severity, patient goals, and medication side effect profiles.

#### 6. Parental Caregiver Expectations and Satisfaction Following Hip Reconstruction and Spinal Fusion in Children With Cerebral Palsy

David Geyer, Judith A Vessey, Anna Chen, Rachel L DiFazio

Orthop Nurs. 2023 Mar-Apr;42(2):94-102. doi: 10.1097/NOR.0000000000000928.

Children with severe (Gross Motor Function Classification System [GMFCS] IV-V) cerebral palsy (CP) exhibit profound physical and developmental impairments and require assistance for all activities of daily living. No curative treatments exist although surgical procedures to correct underlying hip and spine deformities can improve their quality of life. Despite the efficacy of these surgeries, little is known regarding parental caregivers' expectations specific to surgical outcomes and their satisfaction with surgical outcomes. The purpose was to explore parental caregiver expectations and satisfaction with hip and spine surgeries that their children with GMFCS IV-V CP underwent. Variations among preoperative expectations, changes in expectations over time, and the relationship of expectations on caregiver satisfaction were examined. A qualitative descriptive approach with conventional content analysis was utilized. Three preoperative caregiver expectation themes were identified: increasing functionality, increasing comfort, and maintaining health and averting crisis. These themes were still present at 5-year follow-up; however, more caregivers refocused their expectations from improving function to providing palliation. A clear relationship between expectations and caregiver satisfaction, however, was not identified. A deeper understanding surrounding caregiver expectation and satisfaction following surgical procedures is needed.

PMID: 36944203

#### 7. The Effects of Trunk Impairment on Fatigue and Balance in Children with Cerebral Palsy Mehmet Duray, Aziz Dengiz, Erdogan Kavlak, Serife Tutar

Percept Mot Skills. 2023 Mar 21;315125231165548. doi: 10.1177/00315125231165548. Online ahead of print.

Cerebral palsy (CP) involves trunk impairment, leading to decreased postural control that is an important contributor to problems maintaining daily activities without undue fatigue. Our aim in this research was to determine the effects of trunk impairment on fatigue and balance in children with hemiplegic CP. We included 65 ambulatory children with CP in this cross-sectional study, and we grouped them into those with greater trunk impairment (Group 1, n = 33) and lesser trunk impairment (Group 2, n = 32) according to their scores on the Trunk Imparment Scale (TIS). We assessed their fatique level using the Pediatric Quality of Life Inventory<sup>TM</sup> Version 4.0 Multidimensional Fatigue Scale (PedsQL-MFS), and their balance ability using the Pediatric Balance Scale (PBS). We found that the PedsQL-MFS parameters, except the general and cognitive fatigue scores, were significantly better for participants in Group 2 than in Group 1; however Group 2 showed significantly higher scores for balance ability than did Group 1 (p < .001). All of the TIS parameters were significantly correlated with sleep/rest fatigue, and PBS scores and trunk coordination were also significantly correlated with general fatigue (p = .013) and cognitive fatigue (p = .003) which are subparameters of the PedsQL-MFS and PBS (p < .001). However static balance was highly and negatively correlated with cognitive fatigue (p < .037). Increased trunk impairment contributed to the exacerbation of fatigue and balance problems in children with hemiplegic CP. Inadequate trunk control was associated with poor sleep and poor resting quality rather than perceived general and cognitive fatigue.

PMID: 36945131

8. Improving trunk postural control facilitates walking in children with cerebral palsy: a pilot study Shijun Yan, Seoung Hoon Park, Renee Keefer, Weena Dee, Ana-Marie Rojas, William Zev Rymer, Ming Wu

Am J Phys Med Rehabil. 2023 Feb 8. doi: 10.1097/PHM.000000000002206. Online ahead of print.

Objective: The aim of this study was to determine the effects of bilateral trunk support during walking on trunk and leg kinematics and neuromuscular responses in children with cerebral palsy (CP). Design: Fourteen children with spastic CP (GMFCS level I to III) participated in this study. Children walked on a treadmill under 4 different conditions, i.e., without support (BASELINE), with bilateral support applied to the upper trunk (UTS), the lower trunk (LTS), and combined upper and

lower trunk (CTS). The trunk and leg kinematics and muscle activity were recorded. Results: Providing bilateral support to the trunk had a significant impact on the displacement of the pelvis and trunk (p < 0.003) during walking. Children's weaker leg showed greater step length (p = 0.032) and step height (p = 0.012) in CTS compared to BASELINE, and greater step length in UTS (p = 0.02) and CTS (p = 0.022) compared to LTS. Changes in soleus EMG activity during stance phase of gait mirrored the changes in step length across all conditions. Conclusion: Providing bilateral upper or combined upper and lower trunk support during walking may induce improvements in gait performance, which may be due to improved pelvis kinematics. Improving trunk postural control may facilitate walking in children with CP.

PMID: 36946368

#### 9. Rehabilitation of Postural Control and Gait in Children with Cerebral Palsy: the Beneficial Effects of Trunk-Focused Postural Activities

Jonathan Pierret, Christian Beyaert, Rajul Vasa, Emilie Rumilly, Jean Paysant, Sébastien Caudron

Dev Neurorehabil. 2023 Mar 23;1-13. doi: 10.1080/17518423.2023.2193269. Online ahead of print.

Purpose: In children with cerebral palsy (CP), with impaired trunk control and toe-walking, trunk-focused rehabilitation (TFR) based on postural activities was hypothesized to improve trunk postural control, early trunk deceleration, and ankle dorsiflexion braking during walking. Methods: Seventeen children with CP (5-12 years) walking autonomously were randomly assigned to TFR and then usual rehabilitation (TFR-UR) or vice versa (UR-TFR). Results: Only after TFR was significant improvements in (i) the Trunk Control Measurement Scale score, postural sway on an unstable sitting device and standing, and (ii) early sternal and sacral decelerations and coupled negative ankle power due to plantar flexors. Conclusion: TFR improves trunk dynamics and consequently improves coupled toe-walking.

PMID: 36959769

#### 10. Feasibility, validity, and reliability of lower limb tactile and body awareness assessments in children with upper motor neuron lesions

Petra Marsico, Lea Meier, Prof Marietta L van der Linden, Prof Tom H Mercer, Prof Hubertus J A van Hedel

Arch Phys Med Rehabil. 2023 Mar 17;S0003-9993(23)00152-1. doi: 10.1016/j.apmr.2023.02.017. Online ahead of print.

Objective: To investigate the feasibility, discriminative and convergent validity, and inter-rater reliability of a lower limb tactile function and two body awareness assessments in children with upper motor neuron (UMN) lesions. Design: Cross-sectional psychometric study SETTING: Pediatric rehabilitation center PARTICIPANTS: Forty individuals with UMN lesions (mean age 11.7 years, SD 3.4 years; 27 girls) and 40 neurotypically developing children of the same age participated. Interventions: Not applicable. Main outcome measures: We assessed the tactile threshold (TT) with monofilaments and body awareness with tactile localization tasks (TLT) for structural (TLTaction) and spatial (TLTperception) body representation at the foot sole. We compared the test outcomes between children with UMN lesions and neurotypically developing children with the Wilcoxon signed-rank test. Furthermore, we quantified the relationships between the three tests with Spearman correlations (rs) and the interrater reliability with quadratic weighted kappa (kQW). Results: About 80% of the children with UMN lesions perceived the tests easy to perform. The children with UMN lesions had significantly reduced somatosensory function compared to the neurotypically developing children. For the more affected leg, we found good relationships between the TT and the TLTaction (rs = 0.71; p < 0.001) and between the two TLTs (rs = 0.66; p < 0.001), and a fair relationship between the TT and the TLT perception (rs =0.31; p=0.06). The inter-rater reliability analyses for the sum scores showed almost perfect agreement for the TT (κOW more affected leg 0.86; less affected leg 0.81), substantial agreement for TLTaction (κOW more affected leg 0.76; less affected leg 0.63), and almost perfect agreement for TLTperception (kQW more affected leg 0.88; less affected leg 0.74). Conclusion: The three tests are feasible to assess lower limb somatosensory function in children with UMN lesions. Discriminative and convergent validity and reliability of the three tests were confirmed. Further studies should investigate responsiveness and association with motor function of these outcome measures.

#### 11. Quantifying the effects of achilles tendon lengthening surgery: An intraoperative approach Elena Brendecke, Stefanos Tsitlakidis, Marco Götze, Sébastien Hagmann, Filiz Ates

Front Physiol. 2023 Mar 6;14:1143292. doi: 10.3389/fphys.2023.1143292. eCollection 2023.

Achilles tendon lengthening (ATL) is frequently used in the treatment of foot deformities. However, there is currently no objective method to determine the optimal muscle length during surgery. We developed an intraoperative approach to evaluate the passive and active forces of the triceps surae muscle group before and after ATL and aimed to test the following hypotheses: 1) the ankle passive range of motion (ROM) increases, 2) passive muscle forces decrease post-ATL, and 3) forces measured from patients with non-neurological and neurological conditions demonstrate different characteristics. Passive forces at various ankle joint positions were measured in ten patients (11.3  $\pm$  3.0 years old) pre- and post-ATL using a force transducer attached to the Achilles tendon. In six patients, active isometric forces were measured by stimulating the triceps surae supramaximally. Passive forces decreased by 94.3% (p < 0.0001), and ROM increased by 89.4% (p < 0.0001) post-ATL. The pre-ATL passive forces were 70.8%  $\pm$  15.1% lower in patients with idiopathic foot deformities than in patients with neurological conditions (p < 0.001). The peak active force of 209.8  $\pm$  114.3 N was achieved at an ankle angle of 38.3°  $\pm$  16.0°, where the passive force was 6.3  $\pm$  6.7 N. The inter-individual variability was substantial in both groups. In conclusion, the hypotheses posed were supported. The present findings suggest that muscle passive and active force production as well as the inter-individual variability should be considered when planning further treatment.

PMID: 36950296

#### 12. Inter- and intra-rater reliability of new application software for computerised paediatric version of Wisconsin Gait Scale

Agnieszka Guzik, Andżelina Wolan-Nieroda, Mariusz Drużbicki

Sci Rep. 2023 Mar 23;13(1):4757. doi: 10.1038/s41598-023-31436-8.

The paediatric version of Wisconsin Gait Scale (WGS) is a reliable tool for gait assessment in children with spastic hemiplegic cerebral palsy (CP). We decided to develop a solution which will make it possible to objectify the descriptive paediatric version of the WGS, and which, consequently, will allow researchers/clinicians to more easily perform accurate assessment of gait patterns in patients. The aim of the study was to assess inter- and intra-rater reliability of new application software for computerised paediatric version of the WGS in children with hemiplegic CP. The study involved 31 children with hemiplegic CP. The app was designed using a model based on thematic categories of the paediatric WGS, and utilising auxiliary lines between specific points on the patient's body, and taking into account angular values, duration and length of the specific gait phases, in order to enable acquisition of quantitative data corresponding to the components of the WGS. The gait of the study participants was recorded, in series of videos. These provided material for three independent raters who reviewed the recordings twice and assessed the participants' gait using the app. After the evaluation was completed, the data were retrieved from the software. The new application software for the computerised paediatric WGS presents very good inter- and intra-rater reliability. Intra-class correlation coefficient (ICC) was very high in measurement 1 (ICC > 0.9) and 2 (ICC > 0.8), which reflects a very high degree of agreement between the three examiners; there was also high agreement for the specific examiners, between the two measurements (ICC > 0.9). The observational gait scale, objectified through the new software, and enabling computer-aided use of the paediatric WGS, presents practical advantages for examiners since it facilitates decisions taken in the process of WGS-based assessment in children with spastic hemiplegic CP.

PMID: 36959308

## 13. Case report: The gait deviation index may predict neurotherapeutic effects of FES-assisted gait training in children with cerebral palsy

Ahad Behboodi, Aswhini Sansare, Nicole Zahradka, Samuel C K Lee

Case Reports Front Rehabil Sci. 2023 Mar 3;4:1002222. doi: 10.3389/fresc.2023.1002222. eCollection 2023.

Background: Children with cerebral palsy (CP) show progressive loss of ambulatory function characterized by kinematic deviations at the hip, knee, and ankle. Functional electrical stimulation (FES) can lead to more typical lower limb kinematics

during walking by eliciting appropriately timed muscle contractions. FES-assisted walking interventions have shown mixed to positive results in improving lower limb kinematics through immediate correction of gait during the application of FES, or long -term, persisting effects of non-FES-assisted gait improvements following multi-week FES-assisted gait training, at the absence of stimulation, i.e., neurotherapeutic effects. It is unknown, however, if children with CP will demonstrate a neurotherapeutic response following FES-assisted gait training because of the CP population's heterogeneity in gait deviations and responses to FES. Identifying the neurotherapeutic responders is, therefore, important to optimize the training interventions to those that have higher probability of benefiting from the intervention. Objective: The purpose of this case study was to investigate the relationship between immediate and neurotherapeutic effects of FES-assisted walking to identify responders to a FES-assisted gait training protocol. Methods: The primary outcome was Gait Deviation Index (GDI) and secondary outcome was root mean squared error (RMSE) of the lower extremity joint angles in the sagittal plane between participants with CP and a typically developing (TD) dataset. Potential indicators were defined as immediate improvements from baseline during FES-assisted walking followed by neurotherapeutic improvements at the end of training. Case description: Gait analysis of two adolescent female participants with spastic diplegia (Gross Motor Function Classification System level II and III) was conducted at the start and end of a 12-week FES-assisted treadmill training protocol. Participant 1 had scissoring crouch gait, while participant 2 had jump gait. Outcomes: The GDI showed both immediate (presence of FES) and neurotherapeutic (absence of FES after training period) improvements from baseline in our two participants. Joint angle RMSE showed mixed trends between immediate and neurotherapeutic changes from baseline. The GDI warrants investigation in a larger sample to determine if it can be used to identify responders to FES-assisted gait training.

PMID: 36937105

#### 14. Does use of ankle foot orthoses affect the dynamic motor control index during walking in cerebral palsy and idiopathic toe walking populations?

Prabhav Saraswat, Lisa T Carson, Emily R Shull, Lauren C Hyer, David E Westberry

Gait Posture. 2023 Mar 11;102:100-105. doi: 10.1016/j.gaitpost.2023.03.007. Online ahead of print.

Background: The dynamic motor control (walk-DMC) index during walking is a measure of the complexity of muscle activation pattern. Ankle Foot Orthoses (AFO) are frequently used to improve the gait of children with Cerebral Palsy (CP) and Idiopathic Toe Walking (ITW). The purpose of this study was to assess the change in walk-DMC index secondary to AFO use. Research question: Does the change in walk-DMC reflect the change in walking kinematics with the use of AFO. Methods: Individuals with diagnosis of CP or ITW with gait analysis data available for barefoot and AFO condition were retrospectively identified. For each individual, the walk-DMC index, Gait Deviation Index (GDI) and Gait Variable Scores (GVS) of knee and ankle kinematics were computed for BF and AFO conditions. Paired t-tests were used to compare key variables between BF and AFO conditions. Multi-variate stepwise regression analysis was performed to identify variables that may predict the increase in walk-DMC between BF and AFO condition. Results: 253 individuals were included in the study. For CP individuals (n = 208), statistically significant but quantitatively minimal improvement was observed in walk-DMC (1  $\pm$  9), GDI (2  $\pm$  9) and ankle GVS (2  $\pm$  7). For ITW individuals (n = 45), larger improvements were observed in walk-DMC (11  $\pm$  13), GDI (9  $\pm$  11) and ankle GVS (6 ± 7). Diagnosis of ITW, use of Solid-AFO and Posterior Leaf Spring-AFO were the significant predictor of increase in walk-DMC with AFO. Higher ankle GVS at BF condition (larger deviation from TD) led to larger increase in walk-DMC. Higher knee GVS (larger deviation from TD) led to smaller increase in walk-DMC. Significance: Use of AFO can lead to improvement in walking kinematics that is reflected in increase in walk-DMC with AFO compared to BF for ITW individuals. The change in kinematics and walk-DMC with use of AFO was minimal for CP individuals.

PMID: 36958157

#### 15. Predictors of Physical Activity Levels in Ambulant/Semiambulant Children and Adolescents With Cerebral Palsy: A Clinical Cohort Study

Christina Esmann Fonvig, Jens Troelsen, Jan Christian Brønd, Sören Möller, Anders Holsgaard-Larsen

Adapt Phys Activ Q. 2023 Mar 24;1-23. doi: 10.1123/apaq.2022-0169. Online ahead of print.

Children and adolescents with cerebral palsy (CP) show a reduced physical activity (PA) level compared with their typically developed peers. This study aimed to identify potential predictors of objectively evaluated habitual PA using data from a national clinical registry-the Cerebral Palsy Follow-Up Program (CPFP)-and proxy-reported questionnaires. Data from the CPFP database showed that female sex was associated with a lower PA level. Additional questionnaire data revealed age,

popliteal angle limitation, Pediatric Outcomes Data Collection Instrument-Sports and Physical Functioning Scale score, screen time, Functional Mobility Scale score at 50 m, and fatigue as predictors. The proposed models can be used for the prediction and early detection of the PA level and consequently for the potential improvement among ambulant/semiambulant individuals with CP. Further research should investigate the predictive impact of personal, social, and environmental factors on the PA level and the gap in PA levels between girls and boys.

PMID: 36963406

#### 16. Changes in the Status of Spastic Diplegic Children in Terms of Gross Motor Function Classification System and Functional Mobility Scale Following Surgical Intervention: A Single Centre Experience

Laxmish R, Vikas Gupta, Nitu Mishra, Shubhangi Gupta, Prateek Behera

Cureus. 2023 Feb 17;15(2):e35105. doi: 10.7759/cureus.35105. eCollection 2023 Feb.

Introduction Most centers in low- to mid-income countries (LMICs) lack facilities for a comprehensive instrumented gait analysis (IGA) which is often considered the preferred method for assessment of the functional results of surgery in children with spastic diplegia. We aimed to study if there were any changes in the Gross Motor Function Classification System (GMFCS) levels and Functional Mobility Scale (FMS) scores after surgery and whether they can be used as an indirect indicator of change in the functional status of a child. Methods This prospective study was conducted at the Pediatric Orthopedic unit of a teaching hospital on spastic diplegic children requiring surgical intervention. GMFCS levels and FMS scores were recorded before the surgery and at each follow-up visit, with the latest record being two years post-surgery. The change in the scores was indicated as an improvement, deterioration, or no change from the baseline and compared to the score of the preceding visit. In addition, it was examined whether the age at surgery had any effect on the temporal change in the scores. Results A total of 25 children were included for analysis after excluding those who failed to fulfill the predefined inclusion and exclusion criteria. Both the GMFCS levels and FMS scores improved from the third month to one-year postsurgery, after which a few patients had a worsening of their scores at the two years follow-up visit. The age at which surgery was performed had no significant effect on the pattern of change in the scores. Most children sought consultations with the physical therapy department only when they visited the surgical team for follow-up. Conclusion This study shows that surgical interventions do improve the functional outcomes in children with spastic CP when assessed using FMS scores while maintaining an undeteriorated GMFCS level in most children. While a peak improvement can be expected one year after surgery in most patients, possible of worsening from baseline scores do exist, and the parents must be informed of the same. Any decision for surgery must involve the parents, and the usefulness of postoperative physical therapy must be impressed upon them before the surgery and during each follow-up visit too.

PMID: 36945285

#### 17. The significance of MUAC z-scores in diagnosing pediatric malnutrition: A scoping review with special emphasis on neurologically disabled children

Kürşad Aydın, Buket Dalgıç, Aydan Kansu, Hasan Özen, Mukadder Ayşe Selimoğlu, Hasan Tekgül, Bülent Ünay, Aysel Yüce

Review Front Pediatr. 2023 Mar 6;11:1081139. doi: 10.3389/fped.2023.1081139. eCollection 2023.

This review by a panel of pediatric gastroenterology-hepatology-nutrition and pediatric neurology experts aimed to address the significance of mid-upper arm circumference (MUAC) assessment in diagnosis of pediatric malnutrition. Specifically, the potential utility of recently developed MUAC z-score tape in clinical practice for larger patient populations was addressed including the neurologically disabled children. In accordance with the evidence-based data, four statements were identified by the participating experts on the utility of MUAC z-score tape, including (1) MUAC z-scores correlate with body mass index (BMI) and weight for height/length (WFH/l) z-scores in diagnosing malnutrition; (2) MUAC z-score tape offers a higher sensitivity to diagnose the mild and moderate malnutrition and better ability to track the changes in nutritional status over time than the other single datapoint measurements; (3) Using single-step MUAC z-score tape in children with cerebral palsy (CP) seems to provide more reliable data on anthropometry; and (4) The clinical value of the tool in classifying secondary malnutrition in CP should be investigated in large-scale populations. In conclusion, enabling single-step estimation of nutritional status in a large-scale pediatric population regardless of age and within a wide range of weight, without formal training or the need for ancillary reference charts and calculators, MUAC z-tape offers a favorable tool for easier and earlier diagnosis of pediatric malnutrition. Nonetheless, further implementation of MUAC z-score screening in larger-scale and/or special populations is necessary to justify its utility in relation to other primary anthropometric indicators in diagnosis of

malnutrition as well as in treatment monitoring in the community and hospital setting.

PMID: 36950173

18. Hematological and biochemical profiles in children with cerebral palsy: A cross-sectional study Rohit Khandelwal, Vivek Vankalapati Manjunath, Leeni Mehta, Shivalingappa Bussenahalli Mangajjera

J Pediatr Rehabil Med. 2023 Mar 13. doi: 10.3233/PRM-201514. Online ahead of print.

Purpose: Cerebral palsy is a common pediatric neurological problem that has multiple comorbidities, including nutritional issues. Hematological and biochemical parameters significantly affect the health status of patients with cerebral palsy, and till now very few studies have analyzed these parameters. This study aimed to describe the hematological and biochemical parameters of children diagnosed with cerebral palsy. Methods: This four-year observational study included children with cerebral palsy who were admitted to the Department of Pediatrics of a medical college teaching hospital. Hemoglobin, platelet count, white blood count (WBC), red blood cells (RBCs), packed cell volume, RBC indices, and biochemical parameters (urea, creatinine, total bilirubin, total protein, albumin, globulin, aspartate transaminase, alanine transaminase, electrolytes, calcium and alkaline phosphatase) were noted and values (mean, standard deviation, and interquartile range) presented. Age and gender -based analyses were performed. Results: The average hemoglobin level was 11.48 mg/dl, platelet count was 301.24×109/L, and WBC count was 11.13 109/L. Anemia was observed more commonly in males younger than nine years of age. Of 282 patients, 14 (4.96%) had a platelet count of less than 150×109/L. Abnormal alkaline phosphatase levels were observed more commonly in patients who were younger than nine years of age and in females more than males, and the difference was statistically significant. Protein levels and calcium levels were similar between both age groups. Conclusion: Anemia was more common in younger patients and males with cerebral palsy. Abnormal bone turnover markers (alkaline phosphatase) were more common in young patients and females with cerebral palsy. Understanding the differential effect of age and gender on various investigational parameters will help improve care of children with cerebral palsy by initiating appropriate and timely clinical interventions, thereby providing a better quality of life.

PMID: 36938743

19. Understanding and managing respiratory infections in children and young adults with neurological impairment Marijke Proesmans, François Vermeulen, Mieke Boon

Expert Rev Respir Med. 2023 Mar 21;1-9. doi: 10.1080/17476348.2023.2192483. Online ahead of print.

Introduction: Patients with neurocognitive impairment (NI) have multiple medical needs, with respiratory problems leading to an important reduction in quality of life and life expectancy. We aimed to explain that the origin of chronic respiratory symptoms in patients with NI is multifactorial. Areas covered: In people with NI there is a high prevalence of swallowing dysfunction and hypersalivation inducing aspiration; cough efficacy is decreased resulting in chronic lung infection; sleep-disordered breathing is frequent and muscle mass is abnormal due to malnutrition. Technical investigations are not always specific and sensitive enough to better diagnose the causes of the respiratory symptoms; moreover, they can sometimes be difficult to perform in this vulnerable patient population. We provide a clinical pathway to adopt to identify, prevent, and treat respiratory complications in children and young adults with NI. A holistic approach in discussion with all care providers and the parents is highly recommended. Expert opinion: The care for people with NI and chronic respiratory problems is challenging. The interplay between several causative factors may be difficult to entangle. Well-performed clinical research in this field is largely missing and should be encouraged. Only then, evidence-based clinical care will become possible for this vulnerable patient group.

#### 20. The Prevalence of Bladder and Bowel Dysfunction in Children with Cerebral Palsy and its Association with Motor, Cognitive, and Autonomic Function

Moriah Baram, Luba Zuk, Tohar Stattler, Michal Katz-Leurer

Dev Neurorehabil. 2023 Mar 21;1-8. doi: 10.1080/17518423.2023.2193268. Online ahead of print.

Purpose: To describe the prevalence of bladder and bowel dysfunction (BBD) in 8-10-year-old children with cerebral palsy and its association with motor, cognitive, and autonomic dysfunction. Methods: A cross-sectional, random sample study of parents of 8-10-year-old children with cerebral palsy. Tools: The Enuresis/Urinary Incontinence Parental Questionnaire, the Functional Independence Measure children's version, the autonomic signs questionnaire, and the Gross Motor Function Classification System. Results: 39 out of 59 parents consented to participate, whereas 25.64% reported complete continence. Of the 29 children with BBD, 21 (72.4%) had lower urinary tract symptoms and bowel problems. Only two of the children received conservative and noninvasive treatments. Lastly, motor, cognitive and autonomic impairments were associated with incontinence. Conclusions: BBD is common in 8-10-year-old children with cerebral palsy at all levels of functioning. Most having both lower urinary tract symptoms and bowel problems.

PMID: 36943141

#### 21. Concurrent Validity of Measures of Upper Extremity Function Derived from Videogame-Based Motion Capture for Children with Hemiplegia

Richard J Adams, Christopher D Lunsford, Richard D Stevenson, Allison L Ellington, Matthew D Lichter, James T Patrie

Games Health J. 2023 Mar 21. doi: 10.1089/g4h.2022.0160. Online ahead of print.

Objective: Pediatric hemiplegia is associated with wide-ranging deficits in arm and hand motor function, negatively impacting participation in daily occupations and quality of life. This study investigated whether performance measures generated during therapy videogame play by children with hemiplegia can be valid indicators of upper extremity motor function. Materials and Methods: Ten children with hemiplegia used a custom therapy game system alternatively using their affected and non-affected hand to provide motion capture data that spans a wide range of motor function status. The children also completed a series of standardized outcome measure assessments with each hand, including the Quality of Upper Extremity Skills Test, the Jebsen Taylor Hand Function Test, and the Wolf Motor Function Test. Results: Statistical analysis using the nonparametric Spearman rank correlation revealed high and significant correlation between videogame-derived motion capture measures, characterizing the speed and smoothness of movements, and the standardized outcome measure assessments. Conclusion: The results suggest that a low-cost motion capture system can be used to monitor a child's motor function status and progress during a therapy program.

PMID: 36944143

## 22. Towards automated video-based assessment of dystonia in dyskinetic cerebral palsy: A novel approach using markerless motion tracking and machine learning

Helga Haberfehlner, Shankara S van de Ven, Sven A van der Burg, Florian Huber, Sonja Georgievska, Ignazio Aleo, Jaap Harlaar, Laura A Bonouvrié, Marjolein M van der Krogt, Annemieke I Buizer

Front Robot AI. 2023 Mar 2;10:1108114. doi: 10.3389/frobt.2023.1108114. eCollection 2023.

Introduction: Video-based clinical rating plays an important role in assessing dystonia and monitoring the effect of treatment in dyskinetic cerebral palsy (CP). However, evaluation by clinicians is time-consuming, and the quality of rating is dependent on experience. The aim of the current study is to provide a proof-of-concept for a machine learning approach to automatically assess scoring of dystonia using 2D stick figures extracted from videos. Model performance was compared to human performance. Methods: A total of 187 video sequences of 34 individuals with dyskinetic CP (8-23 years, all non-ambulatory) were filmed at rest during lying and supported sitting. Videos were scored by three raters according to the Dyskinesia Impairment Scale (DIS) for arm and leg dystonia (normalized scores ranging from 0-1). Coordinates in pixels of the left and right wrist, elbow, shoulder, hip, knee and ankle were extracted using DeepLabCut, an open source toolbox that builds on a pose estimation algorithm. Within a subset, tracking accuracy was assessed for a pretrained human model and for models

trained with an increasing number of manually labeled frames. The mean absolute error (MAE) between DeepLabCut's prediction of the position of body points and manual labels was calculated. Subsequently, movement and position features were calculated from extracted body point coordinates. These features were fed into a Random Forest Regressor to train a model to predict the clinical scores. The model performance trained with data from one rater evaluated by MAEs (model-rater) was compared to inter-rater accuracy. Results: A tracking accuracy of 4.5 pixels (approximately 1.5 cm) could be achieved by adding 15-20 manually labeled frames per video. The MAEs for the trained models ranged from  $0.21 \pm 0.15$  for arm dystonia to  $0.14 \pm 0.10$  for leg dystonia (normalized DIS scores). The inter-rater MAEs were  $0.21 \pm 0.22$  and  $0.16 \pm 0.20$ , respectively. Conclusion: This proof-of-concept study shows the potential of using stick figures extracted from common videos in a machine learning approach to automatically assess dystonia. Sufficient tracking accuracy can be reached by manually adding labels within 15-20 frames per video. With a relatively small data set, it is possible to train a model that can automatically assess dystonia with a performance comparable to human scoring.

PMID: 36936408

#### 23. Effect of Moringa Oleifera fortified porridge consumption on protein and vitamin A status of children with cerebral palsy in Nairobi, Kenya: A randomized controlled trial

Janet Kajuju Malla, Sophie Ochola, Irene Ogada, Ann Munyaka

PLOS Glob Public Health. 2022 Nov 4;2(11):e0001206. doi: 10.1371/journal.pgph.0001206. eCollection 2022.

Background: Malnutrition due to inadequate dietary intake is commonly reported in children with Cerebral palsy (CP). Poor dietary intakes are majorly caused by feeding dysfunctions secondary to oro-motor impairment characteristic of the condition. Strategies that improve nutrient densities in foods can help enhance nutrient intakes by these children. Objective: This study investigated the effect of consumption of fermented finger millet porridge fortified with Moringa oleifera leaf powder (MoLP) on the protein and vitamin A status of children with CP. Design: A randomized controlled trial was conducted among 113 children aged 5-11 years with CP. The study had two arms (intervention [N = 57] and control [N = 56]). The intervention group received a daily serving of fortified finger millet porridge for 3 months while the control group received non-fortified finger millet porridge servings. All children received the same amounts of porridge servings. The levels of serum albumin and retinol between the groups were compared at both baseline and end line. The BMI-for-age Z-scores (BMIAZ) and morbidity prevalence of the children were also assessed. Results: At baseline, the two study groups were similar in all demographic and socio-economic characteristics, nutrient intakes, serum levels of albumin and retinol, weight status and morbidity. At end line, the children from the intervention group had significantly higher intakes of vitamin A at  $717.12\pm432.7 \,\mu\text{g/d}$  (p = 0.038) and protein at 44.367±17.2 g/d (p = 0.031) respectively. The serum nutrients levels increased significantly from baseline by 0.456±0.12 g/dL (p<0.001) for albumin and by 0.243±0.10 μmol/L (p<0.001) for retinol among children in the intervention group. Among the children in the control group, the changes in the levels of both serum albumin 0.012±0.07 g/dL (p = 0.868) and serum retinol [0.0021±0.02 µmol/L (p = 0.890)] were not significant. At endline, the BMI-for-age Z-scores results showed that 10.52% and 34.0% of children from intervention and control group respectively were undernourished [ $\chi 2 = 30.985$ ; p = 0.037]. Among the children in the intervention, group there was a significant change in the weight status between baseline and endline (p = 0.036). The weight status among children in the control group was not significantly different between baseline and endline (p = 0.109). Significant difference in morbidity prevalence between the two groups was also observed at endline (p = 0.003) with the prevalence being 24.6% and 51.8% among children in the intervention and control group respectively. Conclusion: Consumption of M. oleifera fortified porridge significantly improved the children's serum albumin and retinol levels, as well as BMIAZ. Registration number and name of trial registry: The trial is registered at Pan African Clinical Trials Registry, number PACTR202107669905145 URL link: https://pactr.samrc.ac.za/.

PMID: 36962678

## 24. Barriers to providing healthcare to children living with cerebral palsy in Ghana: A qualitative study of healthcare provider perspectives

Habibat A Oguntade, Thamanna Nishath, Prince G Owusu, Christina Papadimitriou, Kwame S Sakyi

PLOS Glob Public Health. 2022 Dec 9;2(12):e0001331. doi: 10.1371/journal.pgph.0001331. eCollection 2022.

Children with neurodevelopmental disabilities in low- and middle-income countries (LMICs) experience profound health and social inequities. While challenges faced by children living with disabilities and their caregivers have been widely documented, little is known about barriers faced by healthcare providers (HCPs) who serve these children. This study seeks to understand

the barriers to testing, diagnosing, referral, and treatment of children living with cerebral palsy (CLWCP) from the perspectives of HCPs in Ghana. This qualitative study was conducted in the Greater Accra region of Ghana. A snowball sampling strategy was used to recruit HCPs from major hospitals, education centers, and health facilities. Data were collected through 11 semi-structured in-depth interviews (IDIs) with HCPs. Using an adapted version of the Sweat & Denison socio-ecological framework (SDSF), barriers to providing healthcare to CLWCPs were organized into superstructural, structural, environmental, relational, individual, and technological levels. We found that barriers to providing healthcare to CLWCPs exist at all levels of the adapted framework. The most salient barriers were identified at the superstructural, structural, and environmental levels. All HCPs expressed frustration with Ghana's health insurance policies and inadequacies of the health systems infrastructures, such as patient assessment rooms, health information systems, and pharmaceutical products for CP care. HCPs also reported that disability-related stigma often discourages providers in training from specializing in the area of developmental disabilities. HCPs emphasized critical challenges related to local perceptions of disability, gender norms and ideologies, and health system policies and infrastructure. Findings highlight the importance of identifying multi-level factors that can influence testing, diagnosing, referral, treatment, and provision of care for CLWCPs in Ghana. Addressing identified challenges from each level of influence may improve CLWCP's experiences throughout the care continuum.

PMID: 36962893

#### 25. JPRM cerebral palsy special issue 2023

Heakyung Kim, Rachel Byrne, Michael Green

Editorial J Pediatr Rehabil Med. 2023 Mar 15. doi: 10.3233/PRM-239002. Online ahead of print.

No abstract available

PMID: 36938746

#### 26. [Epigenetic influences and brain development][Article in Russian]

P L Sokolov, N V Chebanenko, D M Mednaya

Zh Nevrol Psikhiatr Im S S Korsakova. 2023;123(3):12-19. doi: 10.17116/jnevro202312303112.

In recent years, the amount of scientific data on the involvement of epigenetic processes in the regulation of brain development in postnatal ontogenesis has been rapidly growing. The article provides an overview of scientific research on the mechanisms of epigenetic influences on brain development. Information was searched in the Scopus, Web of Science, MedLine, The Cochrane Library, PubMed, Pedro, Scholar, eLibrary, CyberLeninka and RSCI databases for the period 1940-2022 by keywords: brain development, epigenetics, neuroontogenesis, methylation, histone modifications, chromatin remodeling, noncoding RNAs. Today, the mechanisms of epigenetic influence on the genome include DNA and RNA methylation, covalent modification of histones, chromatin remodeling, and the influence of non-coding RNAs. Epigenetic modifications are often reversible and provide the necessary plasticity for the response of progenitor cells to environmental signals. The influence of each of these factors on the neurodevelopment is considered. The possibility of transsynaptic transmission of hereditary material by means of circular RNA is indicated. The main ways of microRNA influence on brain development are presented and their universality as an «overgenic» regulator of organism adaptation to external conditions is indicated. Data on the relationship of long non-coding RNAs with the regulation of the functional activity of oligodendroglia are presented. Also, the data presented indicate the paths to the pathogenetically determined prevention of congenital brain pathology.

PMID: 36946391

#### 27. Abnormal fetal heart rate patterns caused by pathophysiologic processes other than fetal acidemia Anthony M Vintzileos, John C Smulian

Review Am J Obstet Gynecol. 2023 Mar 17;S0002-9378(22)00346-5. doi: 10.1016/j.ajog.2022.05.002. Online ahead of print.

Fetal acidemia is a common final pathway to fetal death, and in many cases, to fetal central nervous system injury. However, certain fetal pathophysiological processes are associated with significant category II or category III fetal heart rate changes before the development of or in the absence of fetal acidemia. The most frequent of these processes include fetal infection and/ or inflammation, anemia, fetal congenital heart disease, and fetal central nervous system injury. In the presence of significant category II or category III fetal heart rate patterns, clinicians should consider the possibility of the aforementioned fetal processes depending on the clinical circumstances. The common characteristic of these pathophysiological processes is that their associated fetal heart rate patterns are linked to increased adverse neonatal outcomes despite the absence of acidemia at birth. Therefore, in these cases, the fetal heart rate patterns may provide more insight about the fetal condition and pathophysiology than the acid-base status at birth. In addition, as successful timing of intrapartum interventions on the basis of evolution of fetal heart rate patterns aims to prevent fetal acidemia, it may not be logical to continue to use the fetal acid-base status at birth as the gold standard outcome to determine the predictive ability of category II or III fetal heart rate patterns. A more reasonable approach may be to use the umbilical cord blood acid-base status at birth as the gold standard for determining the appropriateness of the timing of our interventions.

PMID: 36964003

#### 28. Children Newly Diagnosed with Fetal and Neonatal Alloimmune Thrombocytopenia: Neurodevelopmental Outcome at School Age

Thijs W de Vos, Maud van Zagten, Masja de Haas, Dick Oepkes, Ratna N G B Tan, C Ellen van der Schoot, Sylke J Steggerda, Linda S de Vries, Enrico Lopriore, Jeanine M M van Klink

J Pediatr. 2023 Mar 16;S0022-3476(23)00184-1. doi: 10.1016/j.jpeds.2023.02.031. Online ahead of print.

Objective: To evaluate the neurodevelopmental outcome at school age in children newly diagnosed with fetal neonatal alloimmune thrombocytopenia (FNAIT). Study design: This observational cohort study included children diagnosed with FNAIT between 2002 and 2014. Children were invited for cognitive and neurological testing. Behavioral questionnaires and school performance results were obtained. A composite outcome of neurodevelopmental impairment (NDI) was used, defined and subdivided into mild-to-moderate and severe NDI. Primary outcome was severe NDI, defined as IQ < 70, cerebral palsy with Gross Motor Functioning Classification Scale (GMFCS) level ≥ III, or severe visual/hearing impairment. Mild-tomoderate NDI was defined as IQ 70-85, minor neurological dysfunction or cerebral palsy GMFCS level ≤ II or mild visual/ hearing impairment. Results: In total, 44 children were included at a median age of 12 years (range 6-17 years). Neuroimaging at diagnosis was available in 82% (36/44) of children. High-grade intracranial hemorrhage (ICH) was detected in 14% (5/36). Severe NDI was detected in 7% (3/44); two children had high-grade ICH and one low-grade ICH and perinatal asphyxia. Mildto- moderate NDI was detected in 25% (11/44); one child had high-grade ICH and eight were without ICH, yet for two children neuroimaging was not performed. Adverse outcome (perinatal death or NDI) was 39% (19/49). Four children (9%) attended special needs education, three of whom had severe NDI and one with mild-to-moderate NDI. Total behavioral problems within the clinical range were reported in 12%, which is comparable with 10% in the general Dutch population. Conclusion: Children who are newly diagnosed with FNAIT are at increased risk for long- term neurodevelopmental problems, even those without ICH.

PMID: 36933767

#### 29. Cerebellar Deep Brain Stimulation in Cerebral Palsy: Promising early results and a look forward to a larger clinical trial

Iahn Cajigas, Melanie A Morrison, Marta San Luciano, Philip Starr

World Neurosurg. 2023 Mar 18;S1878-8750(23)00391-1. doi: 10.1016/j.wneu.2023.03.069. Online ahead of print.

No abstract available

## 30. Editorial: Aging with neurodevelopmental disorders (NDD) Zheng Wang, Matthew W $\operatorname{Mosconi}$

Editorial Front Integr Neurosci. 2023 Mar 2;17:1167014. doi: 10.3389/fnint.2023.1167014. eCollection 2023.

No abstract available