1. The construct and concurrent validity of brief standing sway assessments in children with and without cerebral palsy
James B Tracy, Drew A Petersen, Benjamin C Conner, Justus G Matteson, De'Shjuan G Triplett, Henry G Wright, Christopher M Modlesky, Freeman Miller, Curtis L Johnson, Jeremy R Crenshaw


Background: Standing postural sway is often quantified from center of pressure trajectories. During assessments of longer durations, children may fidget, thus limiting the feasibility and validity of sway recordings. Research question: Do postural sway sample durations less than 30 s maintain construct and concurrent validity? Methods: In this case-control, observational study, we measured postural sway in 41 children (age 5-12 years, 23 typically developing (TD); 18 with spastic cerebral palsy (CP), 13 diplegic and 5 hemiplegic, 11 GMFCS level I and 7 level II) for 30-second eyes-opened and eyes-closed conditions. From a single recording, 5-second incremental durations of 5-30 s were considered in this analysis. We quantified anteroposterior, mediolateral, and transverse-plane sway using seven time-domain variables: root-mean-square error, total excursion, mean frequency, mean distance, sway area, and 95 % confidence circle and ellipse areas. Variables were calculated in eyes-opened and eyes-closed conditions, as well as the ratio of the two. Construct validity was evaluated by the persistence of large effect sizes (Glass's Δ ≥ 0.80) between CP and TD participants at shorter durations than 30 s. Concurrent validity was evaluated by the correlations of shorter duration measures to the 30 s measure. Results: Seven sway measures had large between-group effects (Glass's Δ ≥ 1.02) for the 30 s measure that persisted (Glass's Δ ≥ 0.81) at shorter durations (5-25 s) and also maintained concurrent validity (r ≥ 0.83). Six of these seven measures were taken in the eyes-closed condition, and all seven measures were in the mediolateral direction or transverse plane. Significance: Our analysis suggests that sway durations less than 30 s can uphold construct and concurrent validity. These measures were primarily in the eyes-closed conditions and mediolateral direction. These results are a promising indicator that shorter-duration sway measures may be of utility when fidgeting prevents longer recordings.

PMID: 33421952

2. Commercial Exergaming in Home-Based Pediatric Constraint-Induced Therapy: A Randomized Trial
Tien-Ni Wang, Yu-Lun Chen, Jeng-Yi Shieh, Hao-Ling Chen


Constraint-induced therapy (CIT) is highly effective yet not accessible to many families. Integrating commercial exergaming in home-based CIT may support the availability and attainability of the intervention. The study compared the effects of supplementary use of Nintendo Wii in home-based CIT with dose-equivalent conventional CIT. Eighteen children with cerebral palsy were randomly assigned to 8 weeks of CIT (CIT) or 4 weeks of CIT, followed by 4 weeks of Wii-augmented
CIT (CIT-Wii). Outcome measures included the Bruininks-Oseretsky Test of Motor Proficiency (Manual Dexterity), the ABILHAND-Kids, the WeeFIM (Self-Care), the Test of Playfulness, the Engagement Questionnaire, and the Parenting Stress Index-Short Form. Both groups significantly improved motor outcomes and playfulness. The CIT group demonstrated greater improvement in self-care skills, whereas parental stress decreased only in the CIT-Wii group. CIT-Wii yields no significant difference in treatment effects from conventional CIT and may provide psychosocial benefits.

PMID: 33435831

3. Results after spica cast immobilization following hip reconstruction in 95 cases: is there a need for alternative techniques?
L Pisecky, G Großbötzl, M Gahleitner, C Haas, T Gotterbarm, M C Klotz


Introduction: Developmental dysplasia of the hip (DDH), neurogenic dysplasia of the hip (NDH), and Perthes disease often require surgical treatment. Spica casting is a common postoperative immobilization. The purpose of this study was to evaluate the complications related to the immobilization. Materials and methods: In a retrospective analysis, we included 83 patients (95 hips), who underwent hip reconstructive surgery between 2008 and 2018. We had 43 female and 40 male patients. Age reached from 3 months to 19 years. All patients were treated with a spica cast postoperatively for a 6-week protocol. Complications were analyzed using the full medical documentation and classified according to Clavien-Dindo. Results: We had complications in 23 patients (27.7%). We counted superficial skin lesions in seven, deep skin lesions in three, spasticity of adductors in three, subluxation in two, infection of the plate in one, compliance problem in one, dislocations of the cast in two, reluxation in one, delayed bone healing in one and spasticity of knee flexors in one case. According to the classification of Clavien-Dindo, we were able to count ten type I, four type II, nine type III, zero type IV and zero type V adverse events. Conclusion: The usage of a spica cast after hip reconstructive surgery is still the most popular way of aftertreatment. It has a low complication rate, which may be lowered by well-applied casts and foam padding. Known complications such as spasticity in patients with cerebral palsy, skin lesions, and pressure sores should be observed and avoided. Shorter protocols for immobilization with the usage of foam padding and foam splints lead to less complications. Clinical relevance: Evidence level level IV, case series.

PMID: 33426605

4. Tendon release reduced joint stiffness with unaltered leg stiffness during gait in spastic diplegic cerebral palsy
Chien-Chung Kuo, Hsing-Po Huang, Ting-Ming Wang, Shih-Wun Hong, Li-Wei Hung, Ken N Kuo, Tung-Wu Lu


Biomechanical deviations at individual joints are often identified by gait analysis of patients with cerebral palsy (CP). Analysis of the control of joint and leg stiffness of the locomotor system during gait in children with spastic diplegic CP has been used to reveal their control strategy, but the differences between before and after surgery remain unknown. The current study aimed to bridge the gap by comparing the leg stiffness-both skeletal and muscular components-and associated joint stiffness during gait in 12 healthy controls and 12 children with spastic diplegic CP before and after tendon release surgery (TRS). Each subject walked at a self-selected pace on a 10-meter walkway while their kinematic and forceplate data were measured to calculate the stiffness-related variables during loading response, mid-stance, terminal stance, and pre-swing. The CP group altered the stiffness of the lower limb joints and decreased the demand on the muscular components while maintaining an unaltered leg stiffness during stance phase after the TRS. The TRS surgery improved the joint and leg stiffness control during gait, although residual deficits and associated deviations still remained. It is suggested that the stiffness-related variables be included in future clinical gait analysis for a more complete assessment of gait in children with CP.

PMID: 33449939

5. Dynamic stability in cerebral palsy during walking and running: Predictors and regulation strategies
Roman Rethwilm, Harald Böhm, Madeleine Haase, Dennis Perchthaler, Chakravarthy U Dussa, Peter Federolf
Background: The postural control in cerebral palsy (CP) is often deficient and manifests in a variety of impairments. Consequently, maintaining balance and controlling posture is impeded and results in an increased cost of locomotion and higher risk of falls. The margin of stability is an established measure to quantify dynamic stability during gait. It can be facilitated to analyze impaired control mechanisms, but it is unknown if and how people with CP manage to control the margin of stability during a more demanding motor task, such as running. Research question: How do people with cerebral palsy regulate dynamic stability during walking and running? Methods: Children and adolescents with bilateral cerebral palsy (N = 117; 50 female, 67 male; age 11.0 ± 3.2) were retrospectively included. All underwent instrumented 3D gait analysis, walking and running barefoot at a self-selected gait speed. People with CP were compared to a control group of N = 25 typically developed (TD). Repeated measures ANOVAs were computed to analyze group differences and multiple linear regressions to identify predictors for the medio-lateral margin of stability. Results: The medio-lateral margin of stability was significantly higher in the CP group and was statistically unchanged during running. Different adaptions when running were particularly observed in the lateral trunk lean and step width, which remained high in CP, whereas the TD increased the trunk lean and reduced their step width. Step width was the main predictor for the medio-lateral margin of stability in both gait conditions. Significance: Young people with cerebral palsy manage to maintain their medio-lateral margin of stability during walking and running, however, with significantly higher safety margins compared to typically developed. This conservative strategy may reflect an adaption to motor and postural control impairments.

PMID: 33445142

Ramona Clark, Emmah Baque, Cherie Wells, Andrea Bialocerkowski


Objectives: Accurate, clinically meaningful outcome measures that are responsive to change are essential for selecting interventions and assessing their effects. Little guidance exists on the selection and administration of neurological impairment tests in children with a neurological condition. Clinicians, therefore, frequently modify adult assessments for use in children, yet the literature is inconsistent. This study aims to establish consensus on neurological conditions most likely to require neurological impairment test in pediatrics and the barriers, enablers and modifications perceived to enhance test reliability. Methods: Over a 2-round modified Delphi study, a panel of experts (n = 24) identified neurological conditions perceived to typically require pediatric neurological testing and the modifications to address barriers/enablers to testing. Experts comprised of physical therapists with evidence of advanced training or research in pediatrics. Using a 6-point Likert scale, (6 = strongly agree, 5 = agree, 4 = somewhat agree, 3 = somewhat disagree, 2 = disagree, 1 = strongly disagree), experts rated statements from existing literature. Thematic analyses were conducted on responses to open-ended questions. A-priori consensus was pre-set at 65% agreement/disagreement. Median, mode and interquartile ranges estimated perceived importance. Cessation was pre-determined by non-consensus items < 10% and panel fatigue. Results: Experts reached consensus on 107/112 (96%) items, including identifying 25/26 (96%) neurological conditions they perceived to require routine neurological testing. Experts strongly agreed with high importance that appropriately trained, experienced therapists are less variable when testing children. Communication modifications were perceived most important. Conclusions: High levels of consensus support the use of lower limb neurological testing in a range of pediatric neurological conditions. Trained clinicians should document modifications such as visual aid use. Using recommended modifications could encourage consistency amongst clinicians. Impact: This is the first study to identify the barriers and enablers to pediatric neurological testing. Barriers and enablers were partially addressed through suggested modifications. Further rigorous examination of these modifications is required to support their use.

PMID: 33439245

7. Malnutrition in hospitalized adults with cerebral palsy
Ché Matthew Harris, Scott M Wright


Background: Malnutrition among hospitalized adults with cerebral palsy has not been extensively explored. We sought to
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identify the impact of malnutrition on clinical and resource outcomes among hospitalized adults with cerebral palsy. Methods: This retrospective cohort study surveyed years 2016 and 2017 from the National Inpatient Sample database examining hospitalized adults with cerebral palsy. Regression model analyses were used to evaluate mortality and resource utilization. Results: 154,219 adults with cerebral palsy were hospitalized. Among them, 21,064 (13.5%) had a secondary diagnosis for malnutrition. Patients with and without malnutrition were similar in age (mean age ± SEM: 45.1±0.30 vs. 45.2±0.18 years, p 0.70). Patients with malnutrition were more likely male (57.8% vs 54.8%, p <0.01), and had higher comorbidity scores (Charlson comorbidity score ≥ 3: 11.7% vs 10.8%, p = 0.01). Mortality rates were higher among patients with malnutrition (4.3% vs 2.1%; p<0.01), and they had higher odds for mortality (adjusted Odds Ratio {aOR} = 2.1, [Confidence Interval (CI) 1.7-2.5]; p <0.01). Those with malnutrition were less likely discharged home following hospitalization, aOR = 0.52, [CI 0.48-0.56]; p<0.01). Hospital charges were higher (adjusted Mean Difference {aMD} = + $42,540 CI [$36,934-$48,146]; p<0.01) and length of stay longer (aMD = + 4.3 days CI [3.9-4.7]; p<0.01) among patients with malnutrition. Conclusion: Malnutrition in hospitalized patients with cerebral palsy is associated with increased mortality and hospital resource utilization. Flagging these patients as being 'high risk' when they are hospitalized may result in heightened attentiveness about clinical outcomes in this vulnerable population.

PMID: 33438229

8. Complications of intrathecal baclofen pump treatment in children with spastic cerebral palsy. A comparative analysis of patients weighing over or under 20 kg at the time of implantation
Anne Tabard-Fougère, Federico Canavese, Christina N Steiger, Geraldo De Coulon


Objective: The purpose of this study was to investigate the incidence of complications in pediatric spastic cerebral palsy (CP) patients weighing less than 20 kg at the time of intrathecal baclofen (ITB) pump implantation and to compare it with spastic CP patients heavier than 20 kg. Material and methods: Twenty-seven patients with spastic CP (14 males) treated with ITB implantation at our institution between January 2002 and January 2018 were retrospectively reviewed. Eight of the 27 patients had a bodyweight below 20 kg (group A) and 19 had weight above 20 kg (group B). Results: Group A had a significantly more important proportion of patients with the Gross Motor Function Classification System V compared to group B (88 vs. 42%). The median follow-up was respectively 2.5 (1.8-4.6) and 4.6 (1.9-10.0) years in groups A and B. Median age at the time of ITB implantation was 7.4 (2.8-12.8) and 13.7 (6.5-16.8) years in groups A and B (P = 0.002). The proportion of patients with complications or reoperation was NS between groups A and B (P > 0.05). No postoperative infections were recorded in any of the groups. During follow-up, five patients died (63%) in group A and three (16%) in group B (P = 0.049) within 3.8 years on average after ITB implantation. Conclusions: ITB therapy in spastic CP patients weighing less than 20 kg seems to be as well tolerated and effective as it is in heavier (>20 kg) pediatric patients.

PMID: 33448748

9. Auditory cues reveal intended movement information in middle frontal gyrus neuronal ensemble activity of a person with tetraplegia
Tommy Hosman, Jacqueline B Hynes, Jad Saab, Kaitlin G Wilcoxen, Bradley R Buchbinder, Nicholas Schmansky, Sydney S Cash, Emad N Eskandar, John D Simeral, Brian Franco, Jessica Kelemen, Carlos E Vargas-Irwin, Leigh R Hochberg


Intracortical brain-computer interfaces (iBCIs) allow people with paralysis to directly control assistive devices using neural activity associated with the intent to move. Realizing the full potential of iBCIs critically depends on continued progress in understanding how different cortical areas contribute to movement control. Here we present the first comparison between neuronal ensemble recordings from the left middle frontal gyrus (MFG) and precentral gyrus (PCG) of a person with tetraplegia using an iBCI. As expected, PCG was more engaged in selecting and generating intended movements than in earlier perceptual stages of action planning. By contrast, MFG displayed movement-related information during the sensorimotor processing steps preceding the appearance of the action plan in PCG, but only when the actions were instructed using auditory cues. These results describe a previously unreported function for neurons in the human left MFG in auditory processing contributing to motor control.
10. Population-based study on the prevalence and clinical profile of adults with cerebral palsy in Northern Ireland
Karen McConnell, Emma Livingstone, Oliver Perra, C Kerr


Objectives: This study aimed to report the prevalence and clinical characteristics of adults with cerebral palsy (CP) in a geographically defined region of the UK. Design and setting: Cross-sectional study using the Northern Ireland Cerebral Palsy Register (NICPR). Participants: All validated cases known to the NICPR, born 1981-2001 and alive and resident in Northern Ireland at age 19 years were included. Results: The study included 1218 persons with CP aged 19-39 years, 46 of whom died in adulthood. The prevalence of CP was 2.38 per 1000. The majority of cases had spastic CP (n=1132/1218, 93%) and could walk (n=949/1218, 78%). Those that died in adulthood typically had bilateral spastic CP (n=39/46) and used a wheelchair (n=40/46). Conclusion: The prevalence of CP in adults is similar to other common neurological conditions such as multiple sclerosis and Parkinson's disease. The needs of adults with CP vary widely with almost half having two or more associated impairments that may require multiprofessional and multiagency coordination. Results from this study can be used to inform transformation of health and care services for adults with CP.

PMID: 33419918

11. Congenital anomalies in children with postneonatally acquired cerebral palsy: an international data linkage study
Shona Goldsmith, Sarah McIntyre, Heather Scott, Kate Himmelmann, Hayley Smithers-Sheedy, Guro L Andersen, Eve Blair, Nadia Badawi, Ester Garne, Comprehensive CA-CP Study Group


Aim: To describe the major congenital anomalies present in children with postneonatally acquired cerebral palsy (CP), and to compare clinical outcomes and cause of postneonatally acquired CP between children with and without anomalies. Method: Data were linked between total population CP and congenital anomaly registers in five European and three Australian regions for children born 1991 to 2009 (n=468 children with postneonatally acquired CP; 255 males, 213 females). Data were pooled and children classified into mutually exclusive categories based on type of congenital anomaly. The proportion of children with congenital anomalies was calculated. Clinical outcomes and cause of postneonatally acquired CP were compared between children with and without anomalies. Results: Major congenital anomalies were reported in 25.6% (95% confidence interval [CI] 21.7-29.9) of children with postneonatally acquired CP. Cardiac anomalies, often severe, were common and present in 14.5% of children with postneonatally acquired CP. Clinical outcomes were not more severe in children with congenital anomalies than those without anomalies. Cause of postneonatally acquired CP differed with the presence of congenital anomalies, with cerebrovascular accidents predominating in the anomaly group. Congenital anomalies were likely associated with cause of postneonatally acquired CP in 77% of children with anomalies. Interpretation: In this large, international study of children with postneonatally acquired CP, congenital anomalies (particularly cardiac anomalies) were common. Future research should determine specific causal pathways to postneonatally acquired CP that include congenital anomalies to identify opportunities for prevention.

PMID: 33432582

12. The General Movements Assessment in Neonates With Hypoxic Ischemic Encephalopathy
Nicole R Pouppirt, Valerie Martin, Linda Pagnotto-Hammitt, Alicia J Spittle, John Flibotte, Sara B DeMauro


Background: Clinical measures after birth and studies such as electroencephalogram (EEG) and brain imaging do not fully predict neurodevelopmental outcomes of infants with hypoxic-ischemic encephalopathy. Early detection of adverse neurologic outcomes, and cerebral palsy in particular, in high-risk infants is essential for ensuring timely management. The General
Movements Assessment is a tool that can be used in the early detection of cerebral palsy in infants with brain injury. The majority of studies on the General Movements Assessment in the late preterm and term population were performed prior to the introduction of therapeutic hypothermia. Aims: To apply the General Movements Assessment in late preterm and term infants with hypoxic-ischemic encephalopathy (including those who received therapeutic hypothermia), to determine if clinical markers of hypoxic-ischemic encephalopathy predict abnormal General Movements Assessment findings, and to evaluate interrater reliability of the General Movements Assessment in this population. Study design: Pilot prospective cohort study Subjects: We assessed 29 late preterm and full-term infants with mild, moderate, and severe hypoxic-ischemic encephalopathy in Philadelphia, PA. Results: Most infants’ general movements normalized by the fidgety age. Only infants with moderate or severe hypoxic-ischemic encephalopathy had abnormal general movements in both the writhing and the fidgety ages (n = 6). Seizure at any point during the initial hospitalization was the clinical sign most predictive of abnormal general movements in the fidgety age (sensitivity 100%, specificity 55%, positive predictive value 40%, negative predictive value 100%). Interrater reliability was greatest during the fidgety age (κ = 0.67). Conclusions: Seizures were the clinical predictor most closely associated with abnormal findings on the General Movements Assessment. However, clinical markers of hypoxic-ischemic encephalopathy are not fully predictive of abnormal General Movements Assessment findings. Larger future studies are needed to evaluate the associations between the General Movements Assessment and childhood neurologic outcomes in patients with hypoxic-ischemic encephalopathy who received therapeutic hypothermia.

PMID: 33439066

13. Automated movement recognition to predict motor impairment in high-risk infants: a systematic review of diagnostic test accuracy and meta-analysis
Kamini Raghuram, Silvia Orlandi, Paige Church, Tom Chau, Elizabeth Uleryk, Petros Pechlivanoglou, Vibhuti Shah


Aim: To assess the sensitivity and specificity of automated movement recognition in predicting motor impairment in high-risk infants. Method: We searched MEDLINE, Embase, PsycINFO, CINAHL, Web of Science, and Scopus databases and identified additional studies from the references of relevant studies. We included studies that evaluated automated movement recognition in high-risk infants to predict motor impairment, including cerebral palsy (CP) and non-CP motor impairments. Two authors independently assessed studies for inclusion, extracted data, and assessed methodological quality using the Quality Assessment of Diagnostic Accuracy Studies-2. Meta-analyses were performed using hierarchical summary receiver operating characteristic models. Results: Of 6536 articles, 13 articles assessing 59 movement variables in 1248 infants under 5 months corrected age were included. Of these, 143 infants had CP. The overall sensitivity and specificity for motor impairment were 0.73 (95% confidence interval [CI] 0.68-0.77) and 0.70 (95% CI 0.65-0.75) respectively. Comparatively, clinical General Movements Assessment (GMA) was found to have sensitivity and specificity of 98% (95% CI 74-100) and 91% (95% CI 83-93) respectively. Sensor-based technologies had higher specificity (0.88, 95% CI 0.80-0.93). Interpretation: Automated movement recognition technology remains inferior to clinical GMA. The strength of this study is its meta-analysis to summarize performance, although generalizability of these results is limited by study heterogeneity.

PMID: 33421120

14. Spontaneous movements in the newborns: a tool of quantitative video analysis of preterm babies
Chiara Tacchino, Martina Impagliazzo, Erika Maggi, Marta Bertamino, Isa Blanchi, Francesca Campone, Paola Durand, Marco Fato, Psiche Giannoni, Riccardo Iandolo, Massimiliano Izzo, Pietro Morasso, Paolo Moretti, Luca Ramenghi, Keisuke Shima, Koji Shimatani, Toshio Tsuji, Sara Uccella, Nicolò Zanardi, Maura Casadio


Background and objectives: The number of preterm babies is steadily growing world-wide and these neonates are at risk of neuro-motor-cognitive deficits. The observation of spontaneous movements in the first three months of age is known to predict such risk. However, the analysis by specifically trained physiotherapists is not suited for the clinical routine, motivating the development of simple computerized video analysis systems, integrated with a well-structured Biobank to make available for preterm babies a growing service with diagnostic, prognostic and epidemiological purposes. Methods: MIMAS (Markerless Infant Movement Analysis System) is a simple, low-cost system of video analysis of spontaneous movements of newborns in their natural environment, based on a single standard RGB camera, without markers attached to the body. The original videos are transformed into binarized sequences highlighting the silhouette of the baby, in order to minimize the illumination effects
and increase the robustness of the analysis; such sequences are then coded by a large set of parameters (39) related to the spatial and spectral changes of the silhouette. The parameter vectors of each baby were stored in the Biobank together with related clinical information. Results: The preliminary test of the system was carried out at the Gaslini Pediatric Hospital in Genoa, where 46 preterm (PT) and 21 full-term (FT) babies (as controls) were recorded at birth (T0) and 8-12 weeks thereafter (T1). A simple statistical analysis of the data showed that the coded parameters are sensitive to the degree of maturation of the newborns (comparing T0 with T1, for both PT and FT babies), and to the conditions at birth (PT vs. FT at T0), whereas this difference tends to vanish at T1. Moreover, the coding method seems also able to detect the few ‘abnormal’ preterm babies in the PT populations that were analyzed as specific case studies. Conclusions: Preliminary results motivate the adoption of this tool in clinical practice allowing for a systematic accumulation of cases in the Biobank, thus for improving the accuracy of data analysis performed by MIMAS and ultimately allowing the adoption of data mining techniques.

PMID: 33421664

15. A matter of timing - At what age should multilevel surgery be performed in cerebral palsy patients?
Sophia Julia Häfner


A rather eventful decade draws to a close, but before the year concludes, we learn in this issue of the Biomedical Journal about the correlation of age with the improvement of motor functions in cerebral palsy patients after myofascial release surgery, and the impact on health and life quality of excessive weight during pregnancy. Moreover, we learn about differences in dexterity test norms between populations, and gain some insight into the latest improvements to the challenging medical study program in Taiwan. Finally, we attend an interesting discussion between experts of the field regarding the use of melatonin to protect the brains of preterm infants.

PMID: 33422266

16. Muscle-tendon unit in children with cerebral palsy
Richard L Lieber, Tim Theologis


Muscle-tendon unit surgery for correction of deformities and movement dysfunction in children with cerebral palsy (CP) is fairly complicated. An understanding of basic muscle-tendon unit properties and their adaptation to both CP and surgery are important to develop advances in this field. In this review, we provide information to therapists, surgeons, and scientists regarding the short- and long-term adaptations of the muscle-tendon unit. Surgical releases, lengthening, and transpositions are discussed, as are some of the tissue, cellular, and molecular adaptations.

PMID: 33426691

17. Mutations disrupting neuritogenesis genes confer risk for cerebral palsy.

18. Author Correction: Mutations disrupting neuritogenesis genes confer risk for cerebral palsy


PMID: 33432185

19. The evolution of our understanding of the conceptualization and genetics of cerebral palsy: Implications for genetic testing
Michael Shevell


PMID: 33423928