Cerebral palsy research news

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Interventions and Management

1. The effects of hand deformity on upper-limb function and health-related quality of life in children with spastic hemiplegic cerebral palsy
Hasan Bingol, Mintaze Kerem Gunel

Hand Surg Rehabil. 2021 Aug 25;S2468-1229(21)00247-4. doi: 10.1016/j.hansur.2021.08.009. Online ahead of print.

The aim of this study was to investigate the effect of hand deformity on upper-limb function and health-related quality of life (HRQOL) in children with hemiplegic cerebral palsy (CP). The study included 44 children with hemiplegic CP between the ages of 6 and 14 years (mean age, 10.04 years; SD, 3.1; 23 males, 21 females). The Manual Ability Classification System (MACS) and Gross Motor Function Classification System (GMFCS) were used, with the Zancolli classification to characterize hand deformities on the more affected side. Upper-limb function was assessed in terms of unilateral capacity (Quality of Upper Extremity Skills Test: QUEST) and bimanual performance (Children's Hand-use Experience Questionnaire: CHEQ), while HRQOL was evaluated on the KIDSCREEN-27 questionnaire. Comparison of bimanual performance and unilateral capacity in children with Zancolli level I and II hemiplegic CP found statistically significant differences ($p < 0.01$). There was also a significant difference on the HRQOL 'physical activities and health' subdomain, in favor of Zancolli level I deformity ($P = 0.003$), but not on the other HRQOL domains ($p > 0.05$). Upper-limb function and the HRQOL physical health domain were poorer with greater hand deformity in children with hemiplegic CP.

PMID: 34454162

2. Impact of Upper Extremity Impairment and Trunk Control on Self-Care Independence in Children With Upper Motor Neuron Lesions
Jeffrey W Keller, Annina Fahr, Jan Lieber, Julia Balzer, Hubertus J A van Hedel

Phys Ther. 2021 Aug 1;101(8):pzab112. doi: 10.1093/ptj/pzab112.

Objective: The purpose of this study was to evaluate the relative importance of different approaches to measure upper extremity selective voluntary motor control (SVMC), spasticity, strength, and trunk control for explaining self-care independence in children affected by upper motor neuron lesions. Methods: Thirty-one patients (mean [SD] age = 12.5 [3.2] years) with mild to moderate arm function impairments participated in this observational study. Self-care independence was evaluated with the Functional Independence Measure for children (WeeFIM). Upper extremity SVMC was quantified with the Selective Control of the Upper Extremity Scale (SCUES), a similarity index (SISCUES) calculated from simultaneously recorded surface electromyography muscle activity patterns, and an accuracy and involuntary movement score derived from an inertial-measurement-unit-based assessgame. The Trunk Control Measurement Scale was applied and upper extremity

spasticity (Modified Ashworth Scale) and strength (dynamometry) were assessed. To determine the relative importance of these factors for self-care independence, 3 regression models were created: 1 included only upper extremity SVMC measures, 1 included upper extremity and trunk SVMC measures (overall SVMC model), and 1 included all measures (final self-care model). Results: In the upper extremity SVMC model (total variance explained 52.5%), the assessgame (30.7%) and SCUES (16.5%) were more important than the SISCUES (4.5%). In the overall SVMC model (75.0%), trunk SVMC (39.0%) was followed by the assessgame (21.1%), SCUES (11.0%), and SISCUES (4.5%). In the final self-care model (82.1%), trunk control explained 43.2%, upper extremity SVMC explained 23.1%, spasticity explained 12.3%, and strength explained 2.3%. Conclusion: Although upper extremity SVMC explains a substantial portion of self-care independence, overall trunk control was even more important. Whether training trunk control and SVMC can translate to improved self-care independence should be the subject of future research. Impact: This study highlights the importance of trunk control and selective voluntary motor control for self-care independence in children with upper motor neuron lesions.

PMID: 34464449

3. Effectiveness of Extracorporeal Shock Wave Therapy on Postural Control and Balance in Children With Unilateral Cerebral Palsy: A Randomized Controlled Trial
Furkan Bilek, Fatih Tekin

Percept Mot Skills. 2021 Sep 2;315125211044057. doi: 10.1177/00315125211044057. Online ahead of print.

The aim of this study was to investigate the effects of Extracorporeal Shock Wave Therapy (ESWT) applied to paraspinal muscles on balance and postural control in children with unilateral Cerebral Palsy (CP). A total of 32 children with unilateral CP were included in the study. The children participated in a one-session control to evaluate their reactions to ESWT before randomly assigning them into experimental and control groups. We evaluated children twice: before and after the treatment. We used the Trunk Control Measurement Scale, Trunk Impairment Scale, Pediatric Balance Scale (PBS), and Timed Up and Go (TUG) test in these assessments. Following the pre-test session, we applied Neuro-Developmental Treatment (NDT) programs to both groups at the rate of twice per week for eight weeks; additionally, we applied ESWT only to the experimental group at the rate of twice a week for eight weeks. While there were trends toward increased trunk control skills, PBS scores and TUG test performances from pre- to post-testing in both groups, these improvements were only statistically significant in the experimental group ($p < 0.05$). We conclude that for children with unilateral CP, ESWT applied to paraspinal muscles has significant additive value when combined with NDT to improve postural control and balance.

PMID: 34474622

4. Surgical outcomes of cervical myelopathy in patients with athetoid cerebral palsy
Takahiro Harada, Toshio Nakamae, Naosuke Kamei, Yoshinori Fujimoto, Hideki Manabe, Nobuhiro Tanaka, Yasushi Fujiwara, Kiyotaka Yamada, Yuji Tsuchikawa, Taiki Morisako, Toshiaki Maruyama, Nobuo Adachi

Eur J Orthop Surg Traumatol. 2021 Aug 29. doi: 10.1007/s00590-021-03109-7. Online ahead of print.

Purpose: Surgical treatment for cervical myelopathy with athetoid cerebral palsy remains unestablished. Instrumented fusion is reported to have good clinical results; however, there are no data of decompression surgery for this pathology in recent years. This study aimed to assess the surgical outcomes of laminoplasty with or without posterior instrumented fusion for cervical myelopathy in patients with athetoid cerebral palsy. Methods: A multi-centre surgical series of patients with cervical myelopathy and athetoid cerebral palsy were enrolled in this study. All patients showed symptoms and signs suggestive of cervical myelopathy and underwent laminoplasty with or without instrumented fusion. The Japanese Orthopaedic Association (JOA) score, Barthel index (BI), and changes in the C2-C7 sagittal Cobb angle in the lateral plain radiograph were analysed. Results: There were 25 patients (16 men and 9 women; mean age, 54.4 ± 10.8 years) with cervical myelopathy and athetoid cerebral palsy who underwent surgical treatment. The mean follow-up period was 41.9 ± 35.6 months. Overall, the BI significantly improved after surgery, whereas the JOA score and C2-C7 angle did not improve postoperatively. The recovery rate of the JOA score in the laminoplasty group was significantly higher than that of the fusion group ($P = 0.02$). Conclusions: Cervical laminoplasty with or without instrumented fusion for treating cervical myelopathy due to athetoid cerebral palsy is effective in improving activities of daily living. Cervical laminoplasty may be an effective and less invasive surgical method for selective patients, especially for those with small involuntary movements and no remarkable cervical kyphosis nor

instability.

PMID: 34455477

5. Seated postural control measure: Italian translation and validation in children with cerebral palsy
Marco Tofani, Giulia Blasetti G, Luca Lucibello, Maurizio Sabbadini, Anna Berardi, Giovanni Galeoto, Debra Field, Enrico Castelli

Prosthet Orthot Int. 2021 Aug 27. doi: 10.1097/PXR.0000000000000033. Online ahead of print.

Background: Seating interventions are part of the postural management program recommended for children with Cerebral Palsy (CP) who are nonambulatory or have difficulty in walking. The lack of validated outcome measures for assessing seating interventions limits access to obtain valid and comparable data internationally. Objective: To translate the Seated Postural Control Measure (SPCM) into Italian and assess reliability and validity of the translated measure in a population of children with CP. Study design: Cross-sectional and repeated measures study. Methods: The original version of the SPCM was translated and culturally adapted by a multidisciplinary team according to international guidelines. Internal consistency was examined with Cronbach's alpha. Both inter- and intrarater reliabilities were estimated using intraclass correlation coefficients with 95% confidence intervals. Intrarater reliability was estimated over a 1-week period. Convergent construct validity was investigated by comparing SPCM values with the Level of Sitting Scale, the Gross Motor Function Classification System, and the Manual Ability Classification System using Pearson's correlation coefficients. Results: The Italian version of the SPCM (IT-SPCM) was evaluated on 72 children with CP aged 4-18 years. Cronbach's alpha was 0.83, 0.95, and 0.93 for IT-SPCM alignment subscale, IT-SPCM functioning subscale, and total score, respectively. Reliability for IT-SPCM total score was high for both inter- and intrarater reliabilities (0.95 and 0.93). The IT-SPCM total score was moderately correlated with the Level of Sitting Scale (0.37), Gross Motor Function Classification System (-0.41), and Manual Ability Classification System (-0.56). Conclusion: Preliminary evidence supports the reliability and validity of using IT-SPCM with a population of children with CP.

PMID: 34456320

6. Relationship between Trunk Position Sense and Trunk Control in Children with Spastic Cerebral Palsy: A Cross-Sectional Study
Shilpa Monica, Akshatha Nayak, Abraham M Joshua, Prasanna Mithra, Sampath Kumar Amaravadi, Zulkifli Misri, Bhaskaran Unnikrishnan

Rehabil Res Pract. 2021 Aug 19;2021:9758640. doi: 10.1155/2021/9758640. eCollection 2021.

Methods: In this study, 24 children with spastic CP aged between 8 and 15 years were recruited. They were classified based on their functional performance using Gross Motor Function Classification System (GMFCS). Trunk control and trunk position sense were assessed using the trunk control measurement scale (TCMS) and digital goniometer, respectively. The correlation between these variables was tested using Spearman's correlation coefficient. Results: Significant negative correlation was found between trunk position sense and TCMS score. Similarly, a significant moderate correlation was found between trunk position sense and GMFCS. A strong negative correlation was also found between GMFCS and TCMS. Conclusion: Children with spastic CP with better trunk position sense had better trunk control. Similarly, children with higher functional performance had better trunk control and lesser error in trunk position sense. The current findings imply the relevance of proprioceptive training of the trunk for enhancing trunk motor control in children with spastic CP.

PMID: 34462670

7. The change in sagittal plane gait patterns from childhood to maturity in bilateral cerebral palsy
Bidzina Kanashvili, Freeman Miller, Chris Church, Nancy Lennon, Jason J Howard, John D Henley, Timothy Niiler, Julieanne P Sees, Kenneth J Rogers, M Wade Shrader

Gait Posture. 2021 Aug 28;90:154-160. doi: 10.1016/j.gaitpost.2021.08.022. Online ahead of print.

Background: The longitudinal stability of sagittal gait patterns in diplegic cerebral palsy (CP), stratified using the Rodda classification, is currently unknown. Research question: What is the trajectory of sagittal plane gait deformities as defined by the Rodda classification in a large cohort treated with orthopedic surgery guided by gait analysis? Methods: A retrospective study utilized gait analysis to evaluate sagittal gait parameters before age 8 and after age 15 years. Individual limbs were categorized at each time point according to the Rodda classification based on mean sagittal plane knee and ankle angle during stance. Welch's t-tests compared gait variables from early childhood with maturity and examined changes associated with plantarflexor lengthening surgery. Results: 100 youth with CP were evaluated twice: at a mean age of 5.49 ± 1.18 and 19.09 ± 4.32 years, respectively. Gross Motor Function Classification System distribution at maturity was I (10.5 %), II (55.2 %), III (28.6 %), and IV (5.7 %). At the initial visit, most limbs were in either true equinus (30 %) or jump-knee gait (26.5 %). At maturity, crouch gait (52.5 %) was the most common classification, of which 47.6 % were mild (1-3 standard deviations from age-matched norm; 21°-30°) and 52.4 % moderate or severe. For the entire cohort, at initial and final visits, respectively, mean knee flexion in stance was 26.8°±14.8° and 25.9°±11.4° ($p = 0.320$), ankle dorsiflexion in stance increased from -0.3°±11.5° to 9.0°±6.0° ($p < 0.001$), and passive knee flexion contracture was -2.3°±7.0° and -3.9°±8.0° ($p = 0.043$). In children who started in true equinus, apparent equinus, and crouch, there was no difference in stance phase knee flexion at maturity between those who underwent plantarflexor lengthenings versus those who did not ($p > 0.18$). Significance: The trend in this cohort was toward crouch with increased stance phase ankle dorsiflexion from early childhood to maturity. Plantarflexor lengthenings were not a significant factor in the progression of stance phase knee flexion.

PMID: 34481266

8. Designing a robust controller for a lower limb exoskeleton to treat an individual with crouch gait pattern in the presence of actuator saturation
Maryam Khamar, Mehdi Edrisi, Saeed Forghany

ISA Trans. 2021 Aug 20;S0019-0578(21)00455-9. doi: 10.1016/j.isatra.2021.08.027. Online ahead of print.

Crouch gait is a gait anomaly observed in youngsters with cerebral palsy (CP). Rehabilitation robots are useful for treating individuals with crouch gait. Multiple factors have impact on crouch, including contracture, spasticity, weak motor control, and muscle feebleness, which make the designing and controlling of these exoskeletons for this population a challenging job. A harsh kinematic trajectory enforced by an exoskeleton control strategy may place individuals with spasticity at a high risk of muscle tissue injury. Therefore, in this article, a multi-input multi-output (MIMO) control method is proposed to reduce this risk and improve crouch gait pattern. A constrained control law is used in the model since high power demands may threaten the wearer. In addition, the controller needs to be robust enough against external disturbances and uncertainties. Thus, a nonlinear disturbance observer (NDO) is presented to compute the wearer-generated muscular torque and the uncertainties in the modeling. In addition, a robust constrained MIMO backstepping sliding controller (CMBSC) based on NDO is used to deal with the effect of actuator saturation and uncertainties. A simulation test was used to validate the proposed model and controller. The results of Simulation confirmed the efficiency of the proposed control method when applied to crouch gait with subject specific gait reference. Then, some experimental tests were undertaken to validate the efficiency of the proposed controller.

PMID: 34479722

9. Predictors of Treatment Response to Progressive Resistance Training for Adolescents with Cerebral Palsy
Nicola Theis, Marika Noorkoiv, Grace Lavelle, Jennifer Ryan

Phys Ther. 2021 Aug 27;pzab202. doi: 10.1093/ptj/pzab202. Online ahead of print.

Objective: The aim of the study was to examine the variability in plantar-flexor muscle strength changes after progressive resistance training for adolescents with cerebral palsy (CP) and to identify baseline variables associated with change in muscle strength. Methods: Thirty-three adolescents with CP were randomized to a 10-week progressive resistance training program as part of a randomized controlled trial (STAR trial). The associations between muscle strength at 10 weeks (n = 30 adolescents) and 22 weeks (n = 28 adolescents) and biomechanical and neuromuscular baseline characteristics, motor function, and fidelity to the program were examined with multivariable linear regression. Conclusion: Assessing levels of muscle activation may be able to identify responders to a progressive resistance training program for adolescents with CP. These findings are a first step toward developing tools that can inform decision making in the clinical setting. Impact: Due to the heterogenous nature of CP, it is challenging to assess the efficacy of strength training programs in individuals with CP and to understand the variability in outcomes among participants. This study provides a better understanding of the factors that predict response to an exercise program so that resistance training can be directed to those who will potentially benefit from it. Lay summary: There is wide variability in how well young people with CP respond to resistance training. If you are a young person with CP, your physical therapist can measure the amount of your gastrocnemius muscle activity to get an indication of how well you will respond.

PMID: 34473304

10. Physical therapy with hippotherapy compared to physical therapy alone in children with cerebral palsy: systematic review and meta-analysis
Gustavo Santos de Assis, Tatiane Schlichting, Beatriz Rodrigues Mateus, Alana Gomes Lemos, Adriana Neves Dos Santos

Review Dev Med Child Neurol. 2021 Aug 28. doi: 10.1111/dmcn.15042. Online ahead of print.

Aim: To compare the effects of physical therapy with hippotherapy versus physical therapy alone on the gross motor function of children and adolescents with cerebral palsy (CP). Method: Electronic searches were conducted in January 2021. We included controlled trials of the gross motor function of children and adolescents with CP, aged 2 to 18 years. We extracted means, standard deviations, and changes from the baseline to the end of the intervention. We used the Cochrane Collaboration's tool modified by Effective Practice and Organization of Care (EPOC) to assess the methodological quality and the Grading of Recommendations Assessment, Development and Evaluation (GRADE) method to verify evidence synthesis. We conducted the meta-analysis using Revman 5.3. Results: A total of 315 individuals from six studies were included. Both groups received physical therapy including strength, aerobic, stretch, and mobility exercises, and neurodevelopmental treatment. Studies presented high risk of bias. Both therapies presented similar effects for Gross Motor Function Measure scores, cadence, stride length, and speed during gait. The level of evidence was very low. The change was greater for the physical therapy with hippotherapy group, but inferior to the smallest real difference or the minimal detectable change. Interpretation: Physical therapy with hippotherapy presented similar effects to physical therapy alone on the gross motor function of children and adolescents with CP. Future studies should include larger sample sizes and studies with low risk of bias.

PMID: 34453750

11. The PURPLE N study: objective and perceived nutritional status in children and adolescents with cerebral palsy
Andras Fogarasi, Elisa Fazzi, Ana R P Smorenburg, Maria Mazurkiewicz-Beldzinska, Argirios Dinopoulos, Alena Pobiecka, Dea Schröder-van den Nieuwendijk, Josef Kraus, Hasan Tekgül, PURPLE N study group

Disabil Rehabil. 2021 Sep 2;1-8. doi: 10.1080/09638288.2021.1970255. Online ahead of print.

Purpose: To obtain information on characteristics, management, current objective nutritional status and perception of nutritional status of children with cerebral palsy (CP) from healthcare professionals (HCPs) and caregivers. Materials and methods: A detailed survey of several items on eight main topics (general characteristics, motor function, comorbidities, therapies, anthropometry, feeding mode and problems and perceived nutritional status) was developed and tested for the study. Correlation between nutritional status and Gross Motor Function Classification System (GMFCS) levels was assessed using continuous variables (Z-scores for weight-for-age, height-for-age, weight-for-height, and body mass index-for-age), and categorical variables (being malnourished, stunted, or wasted). HCP and caregiver perceptions of the child's nutritional status as well as agreement between perceived and objective nutritional status and agreement between perceived nutritional status and concerns about the nutritional status were analyzed. Results: Data were available for 497 participants from eight European countries. Poorer nutritional status was associated with higher (more severe) GMFCS levels. There was minimal agreement

between perceived and objective nutritional status, both for HCPs and caregivers. Agreement between HCP and caregiver perceptions of the child's nutritional status was weak (weighted kappa 0.56). However, the concerns about the nutritional status of the child were in line with the perceived nutritional status. Conclusions: The risk of poor nutritional status is associated with more severe disability in children and adolescents with CP. There is a mismatch between HCP and caregiver perceptions of participants' nutritional status as well as between subjective and objective nutritional status. Our data warrant the use of a simple and objective screening tool in daily practice to determine nutritional status in children and adolescents with CP. Clinical trial registration: ClinicalTrials.gov Identifier: NCT03499288 (https://clinicaltrials.gov/ct2/show/NCT03499288). IMPLICATIONS FOR REHABILITATION: Use of the ESPGHAN recommendations and simple screening tools in daily practice is needed to improve nutritional care for individuals with CP. Attention should be paid to the differences in the perception of nutritional status of individuals with CP between professionals and caregivers to improve appropriate referral for nutritional support. Objective measures rather than the professional's perception need to be used to define the nutritional status of individuals with CP.

PMID: 34473588

12. Taking a closer look into Crohn's disease and cerebral palsy
Minh Hoang Nam Nguyen, Eric Martin Sieloff, Joseph Fakhoury, Nicholas Helmstetter

Clin J Gastroenterol. 2021 Aug 31. doi: 10.1007/s12328-021-01510-7. Online ahead of print.

Inflammatory bowel disease (IBD), which includes Crohn's disease (CD), is a chronic, immune-mediated disease involving the gastrointestinal tract. Cerebral palsy (CP) has not been associated with IBD aside from a single previously published case series. In this study, we describe two cases of CD in patients with CP. Both patients had global developmental delay. They were also underweight and exclusively gastrostomy tube dependent for nutrition. By detailing their clinical courses, we illustrate the importance of becoming familiar with the extraintestinal manifestations of IBD. In the setting of developmental delay and its associated communication barriers, the evaluation of IBD might shift from assessing subjective symptoms to recognizing the subtle presentations of the disease, including anemia, weight loss, malnutrition, and failure to thrive. We also hypothesize that malnutrition and a change in the gastrointestinal microbiota associated exclusively with enteral nutrition may be linked to the development of CD in patients with CP. Overall, this study provides information for clinicians caring for patients with CP or developmental delay, and how to recognize extraintestinal manifestations of IBD.

PMID: 34462888

13. The medium to long-term effects of two-duct ligation for excessive drooling in neurodisabilities, a cross-sectional study
Stijn Bekkers, Sanne de Bock, Karen van Hulst, Saskia E Kok, Arthur R T Scheffer, Frank J A van den Hoogen

Int J Pediatr Otorhinolaryngol. 2021 Aug 27;150:110894. doi: 10.1016/j.ijporl.2021.110894. Online ahead of print.

Objectives: This study aims to evaluate the medium to long-term (1-12 years) effectiveness of two-duct ligation (2-DL) in patients with nonprogressive neurodisabilities. Methods: Main outcomes included a Visual Analogue Scale (VAS), Drooling Severity (DS) and Drooling Frequency (DF), collected at baseline, 32 weeks postintervention and 1-12 years postintervention. Secondary outcomes were adverse events (AEs), and satisfaction. Results: Forty-two patients were analyzed (mean age 17 years, mean baseline VAS 82). VAS decreased significantly from baseline to long-term (n = 30. Mean difference -36.5, CI -47.0 to -26.0, $p \leq 0.001$; -26.1, CI -36.2 to -15.9, $p \leq 0.001$). However, long-term VAS significantly increased compared to VAS at 32 weeks (+10.4, CI 1.0-19.8, $p = 0.031$). Out of 42 patients, 64% would recommend 2-DL to peers. Conclusion: There is a significant subjective 2-DL effect on drooling severity in the medium to long-term as reported by patients and caregivers, but there is also a certain degree of recurrence in this time span, and 33% of the patients required subsequent treatment. However, the majority of patients and/or caregivers would recommend 2-DL to peers.

PMID: 34474251

14. A pilot feasibility study of gabapentin for managing pain in children with dystonic cerebral palsy
Adrienne Harvey, Mary-Clare Waugh, James Rice, Giuliana Antolovich, Lisa Copeland, Francesca Orsini, Adam Scheinberg, Clare McKinnon, Megan Thorley, Felicity Baker, George Chalkiadis, Kirsty Stewart

BMC Pediatr. 2021 Aug 28;21(1):368. doi: 10.1186/s12887-021-02847-1.

Background: Gabapentin is often used to manage pain in children with dystonic cerebral palsy, however the evidence for its effectiveness in this population is limited. The primary objective of this feasibility pilot study was to assess the factors which might impact on a future randomised controlled trial including the ability to recruit and retain participants, assess adherence/ compliance to the prescribed intervention, and ability to complete all outcome assessments. The secondary objective was to gather preliminary evidence for the effectiveness of gabapentin at reducing pain, improving comfort and reducing dystonia in children with dystonic cerebral palsy. Methods: This open label pilot study recruited children aged 5-18 years with dystonic cerebral palsy and accompanying pain affecting daily activities from four centres around Australia. Children were prescribed gabapentin for 12 weeks and were assessed at baseline, 6 weeks and 12 weeks. The primary outcome was feasibility of the protocol. Secondary outcomes were pain behaviour, pain intensity, care and comfort, individualised goal setting and dystonia severity. Results: Thirteen children (mean age 10.4 years (SD 2.4yrs), 9 females) were recruited from 71 screened over 15 months. Two children withdrew while eight children experienced side effects. There were issues with adherence to medication dosage regimens and data collection. Improvements were seen in pain behaviour, comfort and pain related goals at 12 weeks. Dystonia was not significantly changed. Conclusions: Whilst gabapentin has potential to improve pain and comfort in children with dystonic CP, the feasibility of implementing a definitive randomised controlled trial is low. Alternative trials designs are required to further examine the effectiveness of gabapentin in this heterogeneous population. Trial registration: The trial was registered with the Australian Clinical Trial Registry (ACTRN12616000366459) on 22/03/2016 and the Therapeutic Goods Administration (CT-2016-CTN-00500-1) on 22/06/2016.

PMID: 34454442

15. The applicability of magnetic resonance imaging classification system (MRICS) for cerebral palsy and its association with perinatal factors and related disabilities in a Croatian population-based sample
Sanja Lovrić Kojundžić, Danijela Budimir Mršić, Ivana Jelovina, Benjamin Benzon, Maja Tomasović

Croat Med J. 2021 Aug 31;62(4):367-375.

Aim: To investigate the association of cerebral palsy motor disorders, perinatal factors, and related disabilities with brain magnetic resonance imaging classification score (MRICS)-based groups in a population-based sample. Methods: The study enrolled children with cerebral palsy born from 2003 to 2015 treated at Split University Hospital who underwent brain MRI scanning. Perinatal data (plurality, birth weight, gestational age, and Apgar score) were collected from hospital records. Motor disorders of cerebral palsy (gross and fine motor function) and the related disabilities (intellectual status, speech and eating ability, epilepsy, vision and hearing status) were evaluated with neurological status assessment. Neuroimaging findings were presented as MRICS-based groups. Results: Of 115 enrolled children, an abnormal finding on brain MRI was confirmed in 95%, including white matter injury (66%), maldevelopments (13.9%), gray matter injury (9.6%), and miscellaneous findings (6.1%). Gross and fine motor function were not significantly associated with MRICS-based group. All related disabilities and perinatal factors, except Apgar score, were significantly associated with MRICS-based group. Conclusion: Brain MRICS-based groups were associated with perinatal risk factors and related disabilities of cerebral palsy, but not with common motor disorders. MRI classification score is a reliable diagnostic tool, which strongly correlates with perinatal factors and related disabilities of cerebral palsy.

PMID: 34472740

16. A chasm: Consequences of poor collaboration between health and education in paediatric cerebral palsy care in Johannesburg
Martha Lydall, Berna Gerber

S Afr J Commun Disord. 2021 Aug 19;68(1):e1-e8. doi: 10.4102/sajcd.v68i1.817.

Background: Nearly 20 years since the establishment of the National Rehabilitation Policy, strides have been made within the health and education sectors to improve accessibility to rehabilitation services as well as the quality of life of children with cerebral palsy (CP). Shortfalls, however, still exist in implementing the policy. An in-depth study into the implementation of policy would be beneficial in identifying and understanding the shortfalls of the rehabilitation process. Objectives: To investigate the perceptions of Speech-Language Therapists (SLTs) working in the Gauteng Department of Health (GDH) and Gauteng Department of Education (GDE), in Johannesburg Region A, about systemic strengths and weaknesses surrounding the service delivery for children with CP, from birth to 6 years. Method: A qualitative study was conducted. Thirty-one (31) SLTs working in public hospitals, clinics and schools for Learners with Special Educational Needs participated in eight focus group interviews. Interviews were audio-recorded for transcription and subsequent thematic analysis. Results: The participants reported a lack of resources and knowledge that contributed to a perceived chasm between the GDH and GDE, resulting in fragmented and uncoordinated service delivery for children with CP leaving the health system and entering the education system. Conclusion: The results suggest that a cohesive plan should be formulated to bridge the perceived chasm between GDH and GDE in the referral process of children with CP from the health setting, into the school environment. This may facilitate communication, collaboration, education, as well as resource-sharing between the departments. Rehabilitation professionals should actively participate in such planning processes.

PMID: 34476955

17. Epidemiology of scoliosis in cerebral palsy: A population-based study at skeletal maturity
Kate L Willoughby, Soon Ghee Ang, Pam Thomason, Erich Rutz, Benjamin Shore, Aaron J Buckland, Michael B Johnson, H Kerr Graham

J Paediatr Child Health. 2021 Aug 28. doi: 10.1111/jpc.15707. Online ahead of print.

Aim: This study investigated the prevalence of scoliosis in a large, population-based cohort of individuals with cerebral palsy (CP) at skeletal maturity to identify associated risk factors that may inform scoliosis surveillance. Methods: Young people with CP born between 1990 and 1992 were reviewed through routine orthopaedic review or a transition clinic. Classification of CP was recorded by movement disorder, distribution, gross and fine motor function. Clinical examination was undertaken and those with clinical evidence of scoliosis or risk factors had radiographs of the spine. Scoliosis severity was measured and categorised by Cobb angle. Results: Two hundred and ninety-two individuals were evaluated (78% of the birth cohort) at a mean age of 21 years, 4 months (range 16-29 years). Scoliosis (Cobb angle >10°) was found in 41%, with strong associations to the Gross Motor Function Classification System (GMFCS), Manual Abilities Classification System (MACS) and dystonic/mixed movement disorders. Those at GMFCS V were 23.4 times (95%CI 9.9-55.6) more likely to develop scoliosis than those at GMFCS I. Severe curves (Cobb >40°, 13% of the cohort) were found almost exclusively in those functioning at GMFCS IV and V, and were 18.2 times (95%CI 6.9-48.5) more likely to occur in those with dystonia than those with spasticity. Conclusions: Scoliosis was very common in young people with CP, with prevalence and severity strongly associated with GMFCS and MACS level and dystonic movement disorder. Severe curves were almost exclusively found in non-ambulant children. Clinical screening for scoliosis should occur for all children with CP, with radiographic surveillance focusing on those functioning at GMFCS IV and V.

PMID: 34453468

18. Epidemiology of eye diseases among children with disability in rural Bangladesh: a population-based cohort study
Mohammad Muhit, Tasneem Karim, Israt Jahan, Mahmudul Hassan Al Imam, Manik Chandra Das, Gulam Khandaker

Dev Med Child Neurol. 2021 Sep 1. doi: 10.1111/dmcn.15041. Online ahead of print.

Aim: To describe the epidemiology of eye diseases among children with disability in rural Bangladesh. Method: We established a population-based cohort of children with disability using the key informant method. Children younger than 18 years with disability (i.e. physical, visual, hearing, speech, epilepsy) were included. We used detailed ophthalmological assessments following World Health Organization (WHO) protocols by a multidisciplinary team including an ophthalmologist, optometrist, physician, and physiotherapist. Visual impairment, blindness, and severe visual impairment (SVI) were defined by following WHO categories. Results: Between October 2017 and February 2018, 1274 children were assessed (43.6% female;

median [interquartile range] age 9y 10mo [6y -13y 7mo]). Overall, 6.5% (n=83) had blindness/SVI, and 5.6% (n=71) had visual impairment. In the group with blindness/SVI, 47% (n=39) had cortical blindness; of those, 79.5% (n=31) had cerebral palsy (CP). The other main anatomical sites of abnormalities in this group included lens (13.3%, n=11), cornea (10.8%, n=9), and optic nerve (9.6%, n=8). In the group with visual impairment, 90.1% (n=64) had refractive error. Overall, 83.1% (n=69) and 78.8% (n=56) of those with blindness/SVI and visual impairment had avoidable causes. Most children with blindness/SVI and visual impairment lacked access to education. Interpretation: The burden of blindness/SVI/visual impairment is high among children with disability in rural Bangladesh, mostly due to avoidable causes. Overrepresentation of CP and cortical blindness in the group with blindness/SVI and refractive error in the group with visual impairment highlights the need for integration of ophthalmology assessment, eye care, and refraction services in comprehensive health care for children with disability including CP in rural Bangladesh.

PMID: 34468025

19. Endogenous erythropoietin at birth is associated with neurodevelopmental morbidity in early childhood
Elina J Rancken, Marjo P H Metsäranta, Mika Gissler, Leena K Rahkonen, Leena M Haataja

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Background: New biomarkers that predict later neurodevelopmental morbidity are needed. This study evaluated the associations between umbilical cord serum erythropoietin (us-EPO) and neurodevelopmental morbidity by the age of 2-6.5 years in a Finnish cohort. Methods: This study included 878 non-anomalous children born alive in 2012 to 2016 in Helsinki University Hospitals and whose us-EPO concentration was determined at birth. Data of these children were linked to data from the Finnish Medical Birth Register and the Finnish Hospital Discharge Register. Neurodevelopmental morbidity included cerebral palsy, epilepsy, intellectual disability, autism spectrum disorder, sensorineural defects, and minor neurodevelopmental disorders. Results: In the cohort including both term and preterm children, us-EPO levels correlated with gestational age (r = 0.526) and were lower in premature children. High us-EPO levels (>100 IU/l) were associated with an increased risk of severe neurodevelopmental morbidity (OR: 4.87; 95% CI: 1.05-22.58) when adjusted for the gestational age. The distribution of us-EPO levels did not differ in children with or without the later neurodevelopmental diagnosis. Conclusions: Although high us-EPO concentration at birth was associated with an increased risk of neurodevelopmental morbidity in early childhood, the role of us-EPO determination in clinical use appears to be minor. Impact: We determined whether endogenous umbilical cord serum erythropoietin would be a new useful biomarker to predict the risk of neurodevelopmental morbidity. This study evaluated the role of endogenous erythropoietin at birth in neurodevelopmental morbidity with a study population of good size and specific diagnoses based on data from high-quality registers. Although high umbilical cord serum erythropoietin concentration at birth was associated with an increased risk of neurodevelopmental morbidity in early childhood, the clinical value of erythropoietin determination appears to be minor.

PMID: 34465877

20. Ischemic hypoxic encephalopathy: The role of MRI of neonatal injury and medico-legal implication
Federico Midiri, Corinne La Spina, Alberto Alongi, Federica Vernuccio, Marcello Longo, Antonina Argo, Massimo Midiri

Forensic Sci Int. 2021 Aug 20;327:110968. doi: 10.1016/j.forsciint.2021.110968. Online ahead of print.

Hypoxic ischemic encephalopathy is one of the major causes of neonatal death and neurological disability in the child, and represents the most common birth injury claim. Intrapartum asphyxia often leads to several long-term sequalae, such as cerebral palsy and/or developmental delay, epilepsy. Through the neuroimaging it's possible to identify and define the different lesioned pictures and provide useful elements to establish the moment in which the damage occurred; indeed, timing of injury is a key element in the legal arena. Magnetic resonance imaging (MRI) is emerging as one of the most important tools in identifying the etiologic of neonatal encephalopathy as well as in predicting long-term outcomes. The aim of this study is to evaluate all MRI tests performed in a group of infants and young patients with possible neonatal encephalopathy, in order to determine the role of MRI in perinatal hypoxic-ischemic damage and the specific patterns that can point towards a diagnosis of the time of the damage's onset. Another goal is to assess the role of MRI in cases subject to legal-medical ligation. Since the advent of hypothermic neuroprotection, new malpractice allegations have arisen, including the failure to initiate cooling in a timely manner. In all cases, documentation of the status of the baby at birth, including a thorough neurologic exam, can be

extremely helpful to the later defence of a malpractice claim, which might occur years later.

PMID: 34455399

21. Predictive validity of a qualitative and quantitative Prechtl's General Movements Assessment at term age: Comparison between preterm infants and term infants with HIE
Helen Robinson, Denise Hart, Brigitte Vollmer

Early Hum Dev. 2021 Aug 17;161:105449. doi: 10.1016/j.earlhumdev.2021.105449. Online ahead of print.

Aim: To determine (1) if the General Movement Optimality Score (GMOS) at term age enhances prediction of motor impairment at 12 and 24 months of age in high-risk infants, when compared to a global General Movement Assessment (GMA), and (2) compare predictive validity for two high-risk populations: infants born preterm and infants born at term with hypoxic ischaemic encephalopathy who have received therapeutic hypothermia. Methods: Fifty-nine extremely preterm or term age infants with hypoxic ischaemic encephalopathy underwent term age GMA. A GMA score of normal or abnormal, and a comparative numerical General Movement Optimality Score (GMOS, total values 5-42) were assigned. Neurology and motor assessment were carried out at age 12 and 24 months using standardised assessments; Alberta Infant Motor Scale, Bayley Scales of Infant and Toddler Development or Ages and Stages Questionnaire. Outcomes were recorded as normal, motor delayed or cerebral palsy. Motor outcome prediction at 12 and 24 months of age was calculated using the GMA and, using ROC analysis, GMOS cut-off scores were determined. Results: At 12 and 24 months global GMA sensitivity for preterms was 80% and 100%, and for Term HIE was 100% at both ages. Specificity values for preterm infants at 12 and 24 months were 68.8% and 60% versus 28.8% and 21.4% for term HIE. Median GMOS scores were lower in the term HIE group than the preterm group in the normal and poor repertoire categories. Optimality cut off scores enhanced specificity, but values remained low. Interpretation: At term age, specificity for identification of infants with later normal motor outcome is low. The GMOS may assist identification of infants with the highest probability of motor impairment, enabling targeted intervention during critical periods for neuroplasticity.

PMID: 34481188

22. Motor outcomes of children born extremely preterm; from early childhood to adolescence
Kate L Cameron, Tara L FitzGerald, Jennifer L McGinley, Kim Allison, Jeanie Ly Cheong, Alicia J Spittle

Semin Perinatol. 2021 Aug 21;151481. doi: 10.1016/j.semperi.2021.151481. Online ahead of print.

Children and adolescents born extremely preterm (EP; <28 weeks' gestation) are at greater risk of motor impairment, including cerebral palsy and developmental coordination disorder, than their term born peers. Importantly, motor impairment has implications beyond performing motor skills; it negatively affects outcomes as diverse as school success, emotional wellbeing, physical health, and physical activity (PA) participation. This review will outline what is known about PA participation across childhood and adolescence for children born EP and term, recognising that PA may improve physical, social, and mental health outcomes. Critically, PA participation occurs in the context of children's and adolescents' daily lives, and is influenced by the family, social and physical environment, as well as by the child's personal factors, such as motor impairment. Further research is needed to better understand PA participation levels and correlates for children and adolescents born preterm, to better inform effective and sustainable interventions.

PMID: 34454740

23. Stronger proprioceptive BOLD-responses in the somatosensory cortices reflect worse sensorimotor function in adolescents with and without cerebral palsy
Timo Nurmi, Julia Jaatela, Jaakko Vallinoja, Helena Mäenpää, Harri Piitulainen

Neuroimage Clin. 2021 Aug 21;32:102795. doi: 10.1016/j.nicl.2021.102795. Online ahead of print.

Cerebral palsy (CP) is a motor disorder where the motor defects are partly due to impaired proprioception. We studied cortical proprioceptive responses and sensorimotor performance in adolescents with CP and their typically-developed (TD) peers. Passive joint movements were used to stimulate proprioceptors during functional magnetic resonance imaging (fMRI) session to quantify the proprioceptive responses whose associations to behavioral sensorimotor performance were also examined. Twenty-three TD (15 females, age: mean ± standard deviation 14.2 ± 2.4 years) and 18 CP (12 females, age: mean ± standard deviation, 13.8 ± 2.3 years; 12 hemiplegic, 6 diplegic) participants were included in this study. Participants' index fingers and ankles were separately stimulated at 3 Hz and 1 Hz respectively with pneumatic movement actuators. Regions-of-interest were used to quantify BOLD-responses from the primary sensorimotor (SM1) and secondary (SII) somatosensory cortices and were compared across the groups. Associations between responses strengths and sensorimotor performance measures were also examined. Proprioceptive responses were stronger for the individuals with CP compared to their TD peers in SM1 ($p < 0.001$) and SII ($p < 0.05$) cortices contralateral to their more affected index finger. The ankle responses yielded no significant differences between the groups. The CP group had worse sensorimotor performance for hands and feet ($p < 0.001$). Stronger responses to finger stimulation in the dominant SM1 ($p < 0.001$) and both dominant and non-dominant SII ($p < 0.01$, $p < 0.001$) cortices were associated with the worse hand sensorimotor performance across all participants. Worse hand function was associated with stronger cortical activation to the proprioceptive stimulation. This association was evident both in adolescents with CP and their typically-developed controls, thus it likely reflects both clinical factors and normal variation in the sensorimotor function. The specific mechanisms need to be clarified in future studies.

PMID: 34474316

24. [Comprehensive approach to children with cerebral palsy] [Article in Spanish]
María José Peláez Cantero, Esther Eugenia Moreno Medinilla, Ana Cordón Martínez, Silvia Gallego Gutiérrez

An Pediatr (Barc). 2021 Aug 27;S1695-4033(21)00249-6. doi: 10.1016/j.anpedi.2021.07.011. Online ahead of print.

Infantile cerebral palsy is one of the most prevalent diseases and the most frequent cause of disability in paediatrics. Children with cerebral palsy have complex health care needs and often require the care of a multidisciplinary team. However, in many cases there is no paediatrician with overall responsibility for coordinating follow-up. We have produced a support document intended for paediatricians coordinating the care of children with cerebral palsy. Our aim is to provide an ordered compilation of the main issues these patients may develop, to know how to identify and address them if necessary, and to establish criteria for referring these patients to other specialists.

PMID: 34462228

25. Editorial: Cerebral Palsy: New Developments
Antigone Papavasiliou, Hilla Ben-Pazi, Sotiria Mastroyianni, Els Ortibus

Editorial Front Neurol. 2021 Aug 11;12:738921. doi: 10.3389/fneur.2021.738921. eCollection 2021.

PMID: 34456856

26. Cerebral palsy and epilepsy: a health informatics approach
Richard F Chin

Dev Med Child Neurol. 2021 Sep 2. doi: 10.1111/dmcn.15038. Online ahead of print.

PMID: 34472638

27. Comparative assessment of health-related quality of life with and without anticonvulsant therapy in patients with childhood epilepsy with centrotemporal spikes
Olga An, Lidia Mayumi Nagae, Altyn Aringazina, Steven Parrish Winesett

J Int Med Res. 2021 Aug;49(8):3000605211039805. doi: 10.1177/03000605211039805.

Objective: To estimate anti-seizure medication (ASM) treatment burden and its effects on health-related quality of life (HRQOL) in new-onset childhood epilepsy with centrotemporal spikes (CECTS) using different treatment approaches in Kazakhstan. Methods: Forty-three patients were followed prospectively during 2015 to 2020 for at least 2 years. Patients were divided into three groups: (1) history of ≤3 seizures (n = 32); (2) ≥4 seizures (n = 6); (3) cerebral palsy coexisting with CECTS (n = 5). The first group was subdivided into treated (n = 8) and observed (n = 24) subgroups. The shortened Quality of Life in Childhood Epilepsy Questionnaire (QOLCE-55) was completed by parents after 6 months of follow-up. Results: At the end of the study, all children had a sustained remission from seizures for at least 2 years. Differences were identified in emotional, social, and physical subscales between patients in the low seizure frequency group. Signs of low self-esteem, anxiety, depression, limited social interaction owing to pharmacotherapy, painful medical procedures, and stigma were reasons for decreased HRQOL in the treated subgroup. Overall HRQOL in treated (89.2 ± 5.2) patients was significantly decreased compared with observed children with low seizure frequency (98.0 ± 3.0). Conclusion: ASM therapy does not necessarily improve and may decrease HRQOL in children with low seizure frequency CECTS.

PMID: 34459274

28. Mesenchymal Stromal Cells Perspective: New Potential Therapeutic for the Treatment of Neurological Diseases
Takeo Mukai, Kenshi Sei, Tokiko Nagamura-Inoue

Review Pharmaceutics. 2021 Jul 27;13(8):1159. doi: 10.3390/pharmaceutics13081159.

Several studies have shown that mesenchymal stromal/stem cells (MSCs) exert their neuroprotective and neurorestorative efficacy via the secretion of neurotrophic factors. Based on these studies, many clinical trials using MSCs for the treatment of neurological disorders have been conducted, and results regarding their feasibility and efficacy have been reported. The present review aims to highlight the characteristics and basic research regarding the role of MSCs in neurological disease and to discuss the recent progress in clinical trials using MSCs to treat various neurological disorders.

PMID: 34452120