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Interventions and Management

1. Expert consensus on the surgical evaluation and management of upper extremity spasticity in adults

Christopher S Crowe, Paula A Pino, Peter C Rhee And The Upper Extremity Spasticity Working Group

Review J Hand Surg Eur Vol. 2023 Sep 17;17531934231192843. doi: 10.1177/17531934231192843. Online ahead of print.

In the last decade there has been incredible interest and advancement in the surgical care of adult patients with upper motor neuron (UMN) injuries. Spasticity represents a prevalent and debilitating feature of UMN syndrome, which can result from cerebral palsy, spinal cord injury, cerebrovascular accident and traumatic or anoxic brain injury. While several diagnostic tools and management strategies have been described for upper limb spasticity, evidence-based practice guidelines do not currently exist due to low patient volume and a paucity of surgeons routinely performing surgeries in UMN syndrome patients. As such, expert consensus may help provide guidance for patients, therapists and clinicians alike. In this article an expert panel was assembled, and the Delphi method was utilized to present diagnostic considerations, define operative indications, discuss surgical treatment modalities and encourage a standard set of outcome measures for patients with upper extremity spasticity.

PMID: 37717178

2. Comparative Analysis of Physical Examination, CT Scan, and Three-Dimensional Gait Analysis in Evaluating Lower Extremity Torsion Deformities in Children with Cerebral Palsy

Sheng Jin, Chunxin Xu, Haiqing Cai, Cen Chen, Yangyang Lu, Zhigang Wang, Min Shen

Med Sci Monit. 2023 Sep 18;29:e940948. doi: 10.12659/MSM.940948.

BACKGROUND The aim of this study was to analyze the correlation and the accuracy of lower-extremity torsion deformities measured by physical examination, CT scan, and three-dimensional gait analysis in children with CP. MATERIAL AND METHODS The study group included 72 children with CP with lower-extremity torsion deformities. All subjects were assessed by: 1. physical examination: maximum internal rotation (MIR), maximum external rotation (MER) for hip joint torsion, and transmalleolar axis (TMA) for tibial torsion; 2. CT scanning: femoral anteversion (FAV) and tibial torsion (TT); 3. three-dimensional gait analysis kinematic parameters: single-support phase of femoral rotation, double-support phase of femoral rotation, swing phase of femoral rotation and single-support phase of tibial rotation, double-support phase of tibial rotation, and swing phase of tibial rotation. Statistical analysis was performed using the Pearson correlation test. A significance level of $P<0.05$ was set. RESULTS In femurs, MIR and MER were correlated with FAV, and the correlation of MER was higher, while physical examination and FAV were not correlated with any kinematic data in gait analysis. In tibias, there was no correlation between TMA and TT, but both TMA and TT were correlated with the gait analysis kinematic data, and the correlation of TT was higher. TMA was more correlated with tibial rotation during swing phase, while TT was more correlated with tibial rotation in single-support phase. CONCLUSIONS Three-dimensional gait analysis can analyze the tibial rotation of children with cerebral palsy, which is highly correlated with CT and physical examination. However, femoral rotation was not associated with CT and physical examination.

PMID: 37721931

3. Impact of lower muscle stiffness on ankle dorsiflexion restriction in children with cerebral palsy evaluated using ultrasound elastography

Shinya Nakamura, Minoru Kimoto, Kyoji Okada, Uki Kawanobe, Hitoshi Sakamoto

Clin Biomech (Bristol, Avon). 2023 Sep 15;109:106092. doi: 10.1016/j.clinbiomech.2023.106092. Online ahead of print.

Background: Plantar flexor muscles always contribute to limiting the range of motion of ankle dorsiflexion in children with spastic cerebral palsy, but the individual contributions of these muscles are not well defined. This study aimed to identify which muscles' stiffness impacts the dorsiflexion range of motion in children with cerebral palsy. Methods: Twenty-five children with cerebral palsy were included. The maximum passive dorsiflexion range of motion was measured in two positions: hip and knee joints in flexion, and both joints in full extension. Strain ratios indicating muscle stiffness were measured using strain elastography of the lateral and medial gastrocnemius, soleus, flexor hallucis longus, peroneus longus, peroneus brevis, and tibialis posterior muscles. To analyze which muscles impact the limitation of the dorsiflexion range, multiple regression analyses were conducted. The values of muscle stiffness were included as independent valuables, and the values of the dorsiflexion range were included as dependent valuables. A p-value <0.05 was considered statistically significant. Findings: In the analyses, the soleus and flexor hallucis longus muscle stiffness were significant independent factors for the dorsiflexion range of motion of hip and knee flexion (adjusted R2: 0.50). The lateral gastrocnemius muscle stiffness was a significant independent factor for the dorsiflexion range of motion with both joints in full extension (adjusted R2: 0.61). Interpretation: Flexor hallucis longus muscle stiffness, in addition to triceps surae muscle stiffness, was shown to impact dorsiflexion range; attention should be paid to muscle stiffness in children with cerebral palsy.

PMID: 37738919

4. Influence of Crouch Angle on Postural Stability in Quiet Stance and Functional Tasks Among Children with Cerebral Palsy

Triveni Shetty, Rajani Mullerpatan, Sailakshmi Ganesan

Dev Neurorehabil. 2023 Sep 21;1-4. doi: 10.1080/17518423.2023.2259979. Online ahead of print.

The aim of this study is to investigate postural stability and function (functional tasks) in younger (<12 yr) and older (>12 yr) children with lesser (<25°) and greater crouch (>25°) during stance. Postural stability and functional tasks were assessed in 53 ambulatory children with spastic cerebral palsy (CP). Younger and older children with greater crouch angle demonstrated higher displacement of center of pressure compared to children with lesser crouch angle during gait ($p < .01$). All (younger and older) children with severe crouch angle demonstrated strong association of postural control with stair climb ($r = 0.732$; $p < .05$) and timed-up-and-go test ($r = 0.84$; $p < .01$). Greater crouch angle demonstrates a moderate association with postural stability ($r = 0.528$; $p < .01$) in quiet stance and a strong association with functional tasks in children with CP ($r = 0.7$-0.84; $p < .05$).

PMID: 37732404

5. Protocol for a randomized controlled trial to evaluate a year-long (NICU-to-home) evidence-based, high dose physical therapy intervention in infants at risk of neuromotor delay

Weiyang Deng, Sofia Anastasopoulos, Raye-Ann deRegnier, Nicole Pouppirt, Ann K Barlow, Cheryl Patrick, Megan K O'Brien, Sarah Babula, Theresa Sukal-Moulton, Colleen Peyton, Catherine Morgan, John A Rogers, Richard L Lieber, Arun Jayaraman

PLoS One. 2023 Sep 19;18(9):e0291408. doi: 10.1371/journal.pone.0291408. eCollection 2023.

Introduction: Developmental disabilities and neuromotor delay adversely affect long-term neuromuscular function and quality of life. Current evidence suggests that early therapeutic intervention reduces the severity of motor delay by harnessing neuroplastic potential during infancy. To date, most early therapeutic intervention trials are of limited duration and do not begin soon after birth and thus do not take full advantage of early neuroplasticity. The Corbett Ryan-Northwestern-Shirley Ryan AbilityLab-Lurie Children's Infant Early Detection, Intervention and Prevention Project (Project Corbett Ryan) is a multi-site longitudinal randomized controlled trial to evaluate the efficacy of an evidence-based physical therapy intervention initiated in the neonatal intensive care unit (NICU) and continuing to 12 months of age (corrected when applicable). The study integrates five key principles: active learning, environmental enrichment, caregiver engagement, a strengths-based approach, and high dosage (ClinicalTrials.gov identifier NCT05568264). Methods: We will recruit 192 infants at risk for neuromotor delay who were admitted to the NICU. Infants will be randomized to either a standard-of-care group or an intervention group; infants in both groups will have access to standard-of-care services. The intervention is initiated in the NICU and continues in the infant's home until 12 months of age. Participants will receive twice-weekly physical therapy sessions and caregiver-guided daily

activities, assigned by the therapist, targeting collaboratively identified goals. We will use various standardized clinical assessments (General Movement Assessment; Bayley Scales of Infant and Toddler Development, 4th Edition (Bayley-4); Test of Infant Motor Performance; Pediatric Quality of Life Inventory Family Impact Module; Alberta Infant Motor Scale; Neurological, Sensory, Motor, Developmental Assessment; Hammersmith Infant Neurological Examination) as well as novel technology-based tools (wearable sensors, video-based pose estimation) to evaluate neuromotor status and development throughout the course of the study. The primary outcome is the Bayley-4 motor score at 12 months; we will compare scores in infants receiving the intervention vs. standard-of-care therapy.

PMID: 37725613

6. The Effectiveness of Aquatic Therapy Based on the Halliwick Concept in Children with Cerebral Palsy: A Systematic Review

Catalina Tapia, Javiera Constanzo, Valentina González, R Mauricio Barría

Review Dev Neurorehabil. 2023 Sep 20;1-6. doi: 10.1080/17518423.2023.2259986. Online ahead of print.

The aim of the review was to evaluate the evidence regarding the effectiveness of aquatic rehabilitation based on the Halliwick concept (HC) in psychomotor development, gross motor function and aquatic skills of children with cerebral palsy. We followed PRISMA recommendations, performing a systematic search in PubMed, Science Direct, LILACS, SciELO, and PEDro database. We identified 474 studies; five met the eligibility criteria and were included in the review. Four studies demonstrated a significant improvement in gross motor function and aquatic skills. Social interaction skills were also improved. However, the methodological quality of these studies was limited, and then, research that adopt controlled experimental designs are necessary.

PMID: 37728374

7. Is the Pediatric Sleep Questionnaire sensitive for sleep-disordered breathing in children with complex chronic disease?

Mine Kalyoncu, Nurtuğ Namlı, Cansu Yilmaz Yegit, Muruvvet Yanaz, Aynur Gulieva, Almala Pınar Ergenekon, Merve Selçuk, Emine Atağ, Nilay Baş İkizoğlu, Meltem Sabancı, Kadir Lale, Yasemin Gokdemir, Refika Ersu, Fazilet Karakoç, Bulent Karadag, Ela Erdem Eralp

Sleep Breath. 2023 Sep 21. doi: 10.1007/s11325-023-02915-z. Online ahead of print.

Purpose: Sleep-disordered breathing (SDB) is a disease defined by breathing or breathing irregularities while asleep. The current study examines the association between results of polysomnography (PSG) and the Pediatric Sleep Questionnaire (PSQ), and the specificity and sensitivity of the PSQ for obstructive sleep apnea (OSA) in patients with chronic illnesses. Methods: Demographic and clinical attributes, in addition to PSQ and PSG outcomes were examined retrospectively among patients who underwent polysomnography (PSG) at our facility between 2012 and 2021. Results: Of 745 patients included in the study, 462 (62%) were male. The median age was 81 months (34-151 months). 117 of the patients (15/8%) had chronic lung disease, and 80 (10.7%) had cerebral palsy. The most common indications for PSG were symptoms of OSA (n = 426; 57.1%). According to obstructive apnea-hypopnea index (AHI), 361 patients (48.5%) had normal PSG. The median PSQ score was 0.40 (0.22-0.57). The sensitivity and specificity of the PSQ were 71.8% and 40.4%, respectively, for individuals aged 2 to 18 years. Among the disease subgroups, the cerebral palsy group had the highest sensitivity of PSQ (88.8%) for diagnosis of OSA. Conclusion: Questionnaires for evaluating SDB are not sensitive or specific for identification of OSA in children with chronic conditions, and PSG remains the best method.

PMID: 37733254

8. Nutritional status and dietary intake of children and adolescents with cerebral palsy

María Elisabeth Cieri, María M Ruiz Brunner, Ana Laura Condinanzi, Johana Escobar, Eduardo Cuestas

Clin Nutr ESPEN. 2023 Oct;57:391-398. doi: 10.1016/j.clnesp.2023.07.080. Epub 2023 Jul 22.

Objective: To analyze the association between energy and nutrient intake, nutritional status and motor compromise in children and adolescents aged 2-19 years with cerebral palsy (CP) attending rehabilitation centers in the Province of Cordoba, Argentina. Methods: Cross sectional study. Data from 105 children and adolescents of both sexes aged 2-19 years with CP (67 boys [63.8% 53.84-72.95]) were collected. Motor compromise was assessed with GMFCS. 24 h records were collected and analyzed. The results were compared with the recommended by FAO/UNU/WHO for age and sex. Normal data were presented with mean and SD, while those of non-normal distribution were described as medians with their ranges. The relationship between variables was analyzed using Fisher, t, or Mann-Whitney tests, with a p value < 0.05. Results: The mean age was 11

years 6 months (SD 4 years 4 months). Fifteen [14.3% 8.23-22.48] children failed to meet at least 80% of the recommended energy. Children with GMFCS IV-V consume fewer daily calories and carbohydrate calories than their peers (I-III). The median protein intake of children GMFCS IV-V group was significantly lower than that of their peers (47.37 g vs. 71.56 g, p = 0.0057). Those who did not reach 80% of the recommended energy intake had lower intakes of macro and micronutrients. Conclusion: The greater the motor compromise in children with CP, the greater the compromise in the adequacy of nutrient intake. The intake of macro and micronutrients was different according to whether or not they were able to cover at least 80% of the recommended energy for their age.

PMID: 37739685

9. Physical Therapy Interventions in Children With Cerebral Palsy: A Systematic Review

Natalie A Gonzalez, Raghavendra R Sanivarapu, Usama Osman, Abishek Latha Kumar, Aishwarya Sadagopan, Anas Mahmoud, Maha Begg, Mawada Tarhuni, Monique N Fotso, Safeera Khan

Review Cureus. 2023 Aug 21;15(8):e43846. doi: 10.7759/cureus.43846. eCollection 2023 Aug.

Cerebral palsy is a group of disorders affecting individuals already from birth. It enormously impacts an individual's physical and emotional life and can bring many challenges to the individual, caregivers, and families. Therefore, it is crucial to investigate interventions that could improve various symptoms in children with cerebral palsy. Our systematic review intends to assess the effect of different exercise and physical therapy interventions in children with cerebral palsy. We used three databases for our article search: PubMed, Medical Literature Analysis and Retrieval System Online (MEDLINE), and PubMed Central (PMC). The combined number of papers found in all databases was 65,412. We then applied our inclusion and exclusion criteria, filters, key terms, and Medical Subheadings (MeSH). After applying our quality assessment tools, we included nine papers in our systematic review. The studies included in our review used various interventions to assess for improvement in symptoms in individuals with cerebral palsy. Interventions included stretching and resistance exercises, horse riding, biking, core stability exercises, slackline training, a home exercise program using an online tool, sit-to-stand exercise program, and functional training. Many studies have shown that interventions improved symptoms like balance, coordination, gait, and cardiovascular endurance in cerebral palsy. This review suggests that some of the included interventions have great potential to improve the symptoms of cerebral palsy and, therefore, can be a great addition to existing training and rehabilitation programs. Given that studies included a relatively small number of participants and were conducted over a short time, more research with a more significant number of participants over a longer time is necessary.

PMID: 37736433

10. Transcranial Focused Ultrasound to V5 Enhances Human Visual Motion Brain-Computer Interface by Modulating Feature-Based Attention

Joshua Kosnoff, Kai Yu, Chang Liu, Bin He

bioRxiv. 2023 Sep 5;2023.09.04.556252. doi: 10.1101/2023.09.04.556252. Preprint

Paralysis affects roughly 1 in 50 Americans. While there is no cure for the condition, brain-computer interfaces (BCI) can allow users to control a device with their mind, bypassing the paralyzed region. Non-invasive BCIs still have high error rates, which is hypothesized to be reduced with concurrent targeted neuromodulation. This study examines whether transcranial focused ultrasound (tFUS) modulation can improve BCI outcomes, and what the underlying mechanism of action might be through high-density electroencephalography (EEG)-based source imaging (ESI) analyses. V5-targeted tFUS significantly reduced the error for the BCI speller task. ESI analyses showed significantly increased theta activity in the tFUS condition at both V5 and downstream the dorsal visual processing pathway. Correlation analysis indicates that the dorsal processing pathway connection was preserved during tFUS stimulation, whereas extraneous connections were severed. These results suggest that V5-targeted tFUS' mechanism of action is to raise the brain's feature-based attention to visual motion.

PMID: 37732253

11. Reliability and validity of the Japanese version of the Visual Function Classification System for children with cerebral palsy

Hisato Nishibu, Yousuke Ikeda, Takahito Inoue, Nobuaki Himuro

Child Care Health Dev. 2023 Sep 18. doi: 10.1111/cch.13175. Online ahead of print.

Background: Children with cerebral palsy (CP) often experience visual dysfunction that affects motor function and activities of daily living, but no 'gold standard' classification of visual function has been established. In recent years, however, a valid and reliable Visual Function Classification System (VFCS) for children with CP has been developed. Aims: To examine the

reliability and validity of the Japanese version of the VFCS in individuals with CP. Methods: The translation of the VFCS was performed according to international standards for the translation of measurements. We conducted questionnaires of professionals (three physicians, eight physical therapists, five occupational therapists, six speech-language-hearing therapists and a certified orthoptist) regarding the content validity of the Japanese version of the VFCS. For reliability and concurrent validity, 148 individuals with CP were classified twice by professionals using the Japanese version of the VFCS, Gross Motor Function Classification System (GMFCS), Manual Ability Classification System (MACS), Communication Function Classification System (CFCS) and Eating and Drinking Ability Classification System (EDACS), with several weeks between each evaluation. Results: The content validity of the Japanese version of the VFCS almost met the criteria set. The percentage of positive and neutral opinions given by the professionals with regard to the four items ranged from 74% to 92%. The intra-rater reliability was 0.86 (95% CI 0.75-0.96) by Cohen's kappa and 0.93 (95% CI 0.88-0.96) by intraclass correlation coefficient. The inter-rater reliability was 0.67 (95% CI 0.56-0.78) by Cohen's kappa and 0.79 (95% CI 0.69-0.86) by intraclass correlation coefficient. The Spearman correlation coefficients between the VFCS and the GMFCS, MACS, CFCS and EDACS were 0.783, 0.764, 0.738, 0.738 and 0.830, respectively. The concurrent validity was confirmed by the correlations observed with other classification systems. Conclusions: The results indicated good reliability and validity for the Japanese version of the VFCS.

PMID: 37723844

12. Season of Conception and Risk of Cerebral Palsy

Haoran Zhuo, Beate Ritz, Joshua L Warren, Zeyan Liew

JAMA Netw Open. 2023 Sep 5;6(9):e2335164. doi: 10.1001/jamanetworkopen.2023.35164.

Importance: Cerebral palsy (CP) is the most prevalent neuromotor disability in childhood, but for most cases the etiology remains unexplained. Seasonal variation in the conception of CP may provide clues for their potential etiological risk factors that vary across seasons. Objective: To evaluate whether the month or season of conception is associated with CP occurrence. Design, setting, and participants: This statewide cohort study examined more than 4 million live births that were registered in the California birth records during 2007 to 2015 and were linked to CP diagnostic records (up to year 2021). Statistical analyses were conducted between March 2022 and January 2023. Exposures: The month and season of conception were estimated based on the child's date of birth and the length of gestation recorded in the California birth records. Main outcomes and measures: CP status was ascertained from the diagnostic records obtained from the Department of Developmental Services in California. Poisson regression was used to estimate the relative risk (RR) and 95% CI for CP according to the month or the season of conception, adjusting for maternal- and neighborhood-level factors. Stratified analyses were conducted by child's sex and neighborhood social vulnerability measures, and the mediating role of preterm birth was evaluated. Results: Records of 4 468 109 children (51.2% male; maternal age: 28.3% aged 19 to 25 years, 27.5% aged 26 to 30 years; maternal race and ethnicity: 5.6% African American or Black, 13.5% Asian, 49.8% Hispanic or Latinx of any race, and 28.3% non-Hispanic White) and 4697 with CP (55.1% male; maternal age: 28.3% aged 19 to 25 years, 26.0% aged 26 to 30 years; maternal race and ethnicity: 8.3% African American or Black, 8.6% Asian, 54.3% Hispanic or Latinx of any race, and 25.8% non-Hispanic White) were analyzed. Children conceived in winter (January to March) or spring (April to June) were associated with a 9% to 10% increased risk of CP (winter: RR, 1.09 [95% CI, 1.01-1.19]; spring: RR, 1.10 [95% CI, 1.02-1.20]) compared with summer (July to September) conceptions. Analyses for specific months showed similar results with children conceived in January, February, and May being at higher risk of CP. The associations were slightly stronger for mothers who lived in neighborhoods with a high social vulnerability index, but no child sex differences were observed. Only a small portion of the estimated association was mediated through preterm birth. Conclusions and relevance: In this cohort study in California, children conceived in winter and spring had a small increase in CP risk. These findings suggest that seasonally varying environmental factors should be considered in the etiological research of CP.

PMID: 37738049

13. Assessing the reliability and validity of a health-related quality of life instrument, CPADULT, in a Dutch sample of adults with severe disabilities who are non-ambulatory

Trees A L Zalmstra, Heleen A Reinders-Messelink, Agnes Elema, Willemijn van Gils, Corry K van der Sluis, Annette A J van der Putten

J Appl Res Intellect Disabil. 2023 Sep 22. doi: 10.1111/jar.13160. Online ahead of print.

Background: A measure to provide insight regarding health-related quality of life of adults with severe motor and intellectual disabilities was lacking. For this reason, the CPADULT was developed. This measure includes domains relating to an individual's physical, mental, and social functioning. The purpose of this study was to assess the psychometric characteristics of the CPADULT. Method: Caregivers (n = 47; 77% female, 23% male) of individuals with severe disabilities who are non-ambulatory completed the questionnaire. Internal consistency, test-retest reliability and construct validity were analysed. Results: Internal consistency was adequate with Cronbach's alpha values from 0.75 to 0.95. Test-retest reliability was good, as intraclass correlation coefficient of the total score was 0.84 (domains: 0.61-0.89). Construct validity was confirmed with

significant differences between subgroups of motor or intellectual abilities. Conclusion: The CPADULT has sufficient reliability and validity as a proxy measure of health-related quality of life for adults with severe disabilities who are non-ambulatory.

PMID: 37737053

14. F-words and intervention content in early intervention for non-ambulatory children with cerebral palsy: a comprehensive literature review [F-Wörter und Interventionsinhalte in der Frühförderung nicht gehfähiger Kinder mit Cerebralparese: eine umfangreiche Literaturübersicht] [Article & Abstract in German]

Review Dev Med Child Neurol. 2023 Sep 23. doi: 10.1111/dmcn.15756. Online ahead of print.

Ana Carolina De Campos, Álvaro Hidalgo-Robles, Egmar Longo, Claire Shrader, Ginny Paleg

OBJECTIVE: To examine the content of early intervention interventions for young children with cerebral palsy (CP) with Gross Motor Function Classification System (GMFCS) levels IV and V and to identify “F-words” addressed by the interventions. Method: Search in four electronic databases. Inclusion criteria: original experimental studies that met the following PCC components: Population: Young children (aged 0-5 years, at least 30% of the sample) with CP and significant motor impairment (GMFCS levels IV or V, at least 30% of the sample); Concept: non-surgical and non-pharmacological early intervention services that measure outcomes from one of the areas of the International Classification of Functioning, Disability and Health (ICF); and context: studies published between 2001 and 2021, in all constellations and not limited to a specific geographical location. Results: 87 studies were included in this literature review, with qualitative (n = 3), mixed methods (n = 4), quantitative descriptive (n = 22), quantitative non-randomized (n = 39) and quantitative randomized (n = 19).) designs. Most experimental studies focused on fitness (n = 59), family (n = 46), and function (n = 33), while there were few studies on fun (n = 6), friends (n = 5), and future (n = 14). Various environmental factors (n = 55) were also significant, e.g. B. the range of services, professional training, therapy dosage and environmental adaptations. Interpretation: Many studies support parent training and the use of assistive technology to promote various F-words. A “menu” of early intervention content was identified, with suggestions for further research to implement this into clinical practice with families.

PMID: 37740649

15. Managing mothers' and fathers' uncertainty during their journey through early neurodevelopmental follow-up for their high-risk infants-A qualitative account

Alice Fortune, Elizabeth Perkins, Fauzia Paize, Balamurugan Palanisami, Melissa Gladstone

Child Care Health Dev. 2023 Sep 22. doi: 10.1111/cch.13168. Online ahead of print.

Background: Early diagnosis of cerebral palsy is possible by 5 months corrected age for 'at-risk' infants, using diagnostic tools such as the Hammersmith Infant Neurological Examination (HINE), Prechtl's General Movements Assessment (GMA) and magnetic resonance imaging (MRI). This is an uncertain and stressful time for parents where provision of appropriate information and support is essential. Aim: To explore parents' views and experiences in relation to the new early neurodevelopmental follow-up of 'at-risk' infants. Methods: Thirteen in-depth one-to-one qualitative interviews were conducted by the primary researcher, with eight parents (six mothers and two fathers) of 'at-risk' infants eligible for a follow-up clinic where the GMA and HINE were performed at 12-week corrected age. Interviews used a pre-piloted topic guide and took place before and after the clinic. Interviews were audio-recorded, transcribed verbatim and analysed using inductive coding and thematic analysis using the framework approach. Findings: Seven themes were identified: (1) attempting to manage uncertainty, (2) taking priority, (3) trusting professionals, (4) independence in the parent role, (5) feeling understood, (6) patterns of care and (7) individuality. Parents reported experiencing uncertainty about their current situation and future. Adequate preparation for and timing of information are vital. When uncertainty is poorly managed, parents' wellbeing suffers. Individual parents' perspectives and infants' developmental trajectories differ, and information should be tailored specifically for this. Conclusion: A parent's understanding of the journey through neurodevelopmental care for their high risk infants is initially very limited. Implementing a counselling service for parents to access psychological support and digital reminder system for clinic appointments, as well as providing more tailored information through trusted professionals, could all improve future parents' experiences.

PMID: 37737651

16. Tissue Oxygenation Changes After Transfusion and Outcomes in Preterm Infants: A Secondary Near-Infrared Spectroscopy Study of the Transfusion of Prematures Randomized Clinical Trial (TOP NIRS)

Valerie Y Chock, Haresh Kirpalani, Edward F Bell, Sylvia Tan, Susan R Hintz, M Bethany Ball, Emily Smith, Abhik Das, Yvonne C Loggins, Beena G Sood, Lina F Chalak, Myra H Wyckoff, Stephen D Kicklighter, Kathleen A Kennedy, Ravi M Patel, Waldemar A Carlo, Karen J Johnson, Kristi L Watterberg, Pablo J Sánchez, Abbot R Laptook, Ruth B Seabrook, C

Michael Cotten, Toni Mancini, Gregory M Sokol, Robin K Ohls, Anna Maria Hibbs, Brenda B Poindexter, Anne Marie Reynolds, Sara B DeMauro, Sanjay Chawla, Mariana Baserga, Michele C Walsh, Rosemary D Higgins, Krisa P Van Meurs; Eunice Kennedy Shriver National Institute of Child Health and Human Development Neonatal Research Network

Randomized Controlled Trial JAMA Netw Open. 2023 Sep 5;6(9):e2334889. doi: 10.1001/jamanetworkopen.2023.34889.

Importance: Preterm infants with varying degrees of anemia have different tissue oxygen saturation responses to red blood cell (RBC) transfusion, and low cerebral saturation may be associated with adverse outcomes. Objective: To determine whether RBC transfusion in preterm infants is associated with increases in cerebral and mesenteric tissue saturation (Csat and Msat, respectively) or decreases in cerebral and mesenteric fractional tissue oxygen extraction (cFTOE and mFTOE, respectively) and whether associations vary based on degree of anemia, and to investigate the association of Csat with death or neurodevelopmental impairment (NDI) at 22 to 26 months corrected age. Design, setting, and participants: This was a prospective observational secondary study conducted among a subset of infants between August 2015 and April 2017 in the Transfusion of Prematures (TOP) multicenter randomized clinical trial at 16 neonatal intensive care units of the Eunice Kennedy Shriver National Institute of Child Health and Human Development Neonatal Research Network. Preterm neonates with gestational age 22 to 28 weeks and birth weight 1000 g or less were randomized to higher or lower hemoglobin thresholds for transfusion. Data were analyzed between October 2020 and May 2022. Interventions: Near-infrared spectroscopy monitoring of Csat and Msat. Main outcomes and measures: Primary outcomes were changes in Csat, Msat, cFTOE, and mFTOE after transfusion between hemoglobin threshold groups, adjusting for age at transfusion, gestational age, birth weight stratum, and center. Secondary outcome at 22 to 26 months was death or NDI defined as cognitive delay (Bayley Scales of Infant and Toddler Development-III score <85), cerebral palsy with Gross Motor Function Classification System level II or greater, or severe vision or hearing impairment. Results: A total of 179 infants (45 [44.6%] male) with mean (SD) gestational age 25.9 (1.5) weeks were enrolled, and valid data were captured from 101 infants during 237 transfusion events. Transfusion was associated with a significant increase in mean Csat of 4.8% (95% CI, 2.7%-6.9%) in the lower-hemoglobin threshold group compared to 2.7% (95% CI, 1.2%-4.2%) in the higher-hemoglobin threshold group, while mean Msat increased 6.7% (95% CI, 2.4%-11.0%) vs 5.6% (95% CI, 2.7%-8.5%). Mean cFTOE and mFTOE decreased in both groups to a similar extent. There was no significant change in peripheral oxygen saturation (SpO2) in either group (0.2% vs -0.2%). NDI or death occurred in 36 infants (37%). Number of transfusions with mean pretransfusion Csat less than 50% was associated with NDI or death (odds ratio, 2.41; 95% CI, 1.08-5.41; P = .03). Conclusions and relevance: In this secondary study of the TOP randomized clinical trial, Csat and Msat were increased after transfusion despite no change in SpO2. Lower pretransfusion Csat may be associated with adverse outcomes, supporting further investigation of targeted tissue saturation monitoring in preterm infants with anemia.

PMID: 37733345

17. The transformational potential of the traumatic experience of parents of children with cerebral palsy

Ruslan Vasyutin

Dev Med Child Neurol. 2023 Sep 21. doi: 10.1111/dmcn.15733. Online ahead of print.

No abstract available

PMID: 37732710

18. Self-reported Health-related Quality of Life in Adolescents With Cerebral Palsy

Nancy Lennon, Faithe Kalisperis, Chris Church, Tim Niiler, Freeman Miller, Isabel Biermann, Jennifer Davey, Julieanne P Sees, M Wade Shrader

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Background: Happiness, comfort, and motor function contribute to satisfaction with life for individuals with cerebral palsy (CP). Evidence-based medical care can improve motor function and physical health of youth with CP. Less is known about medical care and its relationship to health-related quality of life (HRQOL) in adolescents and young adults with CP. This study aimed to describe HRQOL among adolescents with CP to examine differences between adolescent (self) and parent (proxy) reports of HRQOL and to explore associations of pain, age, and gross motor function with HRQOL. Methods: This is a retrospective study including adolescents with CP classified as Gross Motor Function Classification System levels I to V, ages 11 to 20 years, reading ≥ a fourth-grade level, and who completed the self-reported Pediatric Outcomes Data Collection Instrument (PODCI). Parents completed the PODCI concurrently or within 12 months and scores were compared. In addition, self-reported scores were compared between age bands, across Gross Motor Function Classification System levels, with typically developing youth (TDY), and between youth with/without pain. Results: PODCI scores from 102 adolescents [59 males; 15.0 (SD: 2.6) years old] were examined. Scores from 50 adolescents and parents were matched. Mean self-reported scores were significantly higher than mean parent-reported scores in 4 domains: upper extremity and physical function (P=0.018), sports and physical function (P=0.005), happiness (P=0.023), and global functioning (P=0.018). All domains, except

Happiness, were significantly < TDY (P<0.01). The presence of pain was associated with lower scores in all domains (P<0.05). Conclusion: Examining HRQOL with the PODCI revealed significant limitations in physical function and higher pain in adolescents with CP compared with TDY. Self- and parent-reported PODCI results should be considered separately. Adolescents report higher HRQOL compared with parent proxy. Recognizing and validating the perspectives of youth and their parents presents an opportunity for providers to discuss different points of view with families. Such engagement can help promote self-efficacy in youth with CP as they transition to the responsibility of guiding their own care in adulthood. Level of evidence: III, Retrospective comparative study.

PMID: 37728111

19. Genetic Predisposition to Adverse Neurodevelopmental Outcome of Extremely Low Birth Weight Infants

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Objective: This study aimed to evaluate whether there are genetic variants associated with adverse neurodevelopmental outcomes in extremely low birth weight (ELBW) infants. Study design: We conducted a candidate gene association study in two well-defined cohorts of ELBW infants (<1,000 g). One cohort was for discovery and the other for replication. The discovery case-control analysis utilized anonymized DNA samples and evaluated 1,614 single-nucleotide polymorphisms (SNPs) in 145 genes concentrated in inflammation, angiogenesis, brain development, and oxidation pathways. Cases were children who died by age one or who were diagnosed with cerebral palsy (CP) or neurodevelopmental delay (Bayley II mental developmental index [MDI] or psychomotor developmental index [PDI] < 70) by 18 to 22 months. Controls were survivors with normal neurodevelopment. We assessed significant epidemiological variables and SNPs associated with the combined outcome of CP or death, CP, mental delay (MDI < 70) and motor delay (PDI < 70). Multivariable analyses adjusted for gestational age at birth, small for gestational age, sex, antenatal corticosteroids, multiple gestation, racial admixture, and multiple comparisons. SNPs associated with adverse neurodevelopmental outcomes with $p < 0.01$ were selected for validation in the replication cohort. Successful replication was defined as $p < 0.05$ in the replication cohort. Results: Of 1,013 infants analyzed (452 cases, 561 controls) in the discovery cohort, 917 were successfully genotyped for >90% of SNPs and passed quality metrics. After adjusting for covariates, 26 SNPs with $p < 0.01$ for one or more outcomes were selected for replication cohort validation, which included 362 infants (170 cases and 192 controls). A variant in SERPINE1, which encodes plasminogen activator inhibitor (PAI1), was associated with the combined outcome of CP or death in the discovery analysis ($p = 4.1 \times 10\text{-}4$) and was significantly associated with CP or death in the replication cohort (adjusted odd ratio: 0.4; 95% confidence interval: 0.2-1.0; $p = 0.039$). Conclusion: A genetic variant in SERPINE1, involved in inflammation and coagulation, is associated with CP or death among ELBW infants.

PMID: 37726016

20. Prospective cohort study of neurodevelopmental outcomes following extreme neonatal hyperbilirubinaemia in Australia

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Aim: This study aimed to establish the incidence and nature of neurodevelopmental outcomes following extreme neonatal hyperbilirubinaemia in an Australian cohort. Methods: A prospective cohort study of neurodevelopmental outcomes up to 3 years of age of infants born between 2010 and 2013 at ≥34 weeks gestation, with total serum bilirubin ≥450 μmol/L and/or clinical signs of acute bilirubin encephalopathy. Outcome measures comprised neurological examination, Bayley Scales of Infant and Toddler Development, 3rd edition and Ages and Stages Questionnaire, 3rd edition. Results: The Australian estimated incidence of kernicterus is 0.35 per 100 000 live births. Within the follow-up cohort of 26, three children have clinical neurodevelopmental impairment: one has gross motor function classification system level 4 cerebral palsy, audiological deficiency and visual impairment; the second has gross motor function classification system level 1 cerebral palsy and the third has global developmental delay with autism spectrum disorder. Mean Bayley Scales of Infant and Toddler Development, 3rd edition scores were: cognition 10.3 (SD 1.5), receptive communication 9.4 (SD 1.8), expressive communication 9.2 (SD 2.4), fine motor 10.4 (SD 2.6) and gross motor 9.2 (SD 2.3). Conclusion: The Australian national rate of kernicterus compares favourably with global estimates. Future preventative strategies in this context include universal neonatal hyperbilirubinaemia assessment and mandated adverse outcome reporting and investigation.

PMID: 37724614

21. Growth variables and obstetrical risk factors in newborns are associated with psychomotor development at preschool age

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Background: Low birthweight resulting from preterm birth or fetal growth restriction is associated with poor neurocognitive development and child psychopathology affecting school performance and educational success. Prediction of developmental performance may therefore serve as a basis for early intervention strategies to improve educational success and mental health of our children in a timely manner. Objective: This study aimed to explore the predictive capacity of morphometric variables taken at birth and that of obstetrical risk factors to predict developmental performance at 4.3 (standard deviation, 0.8) years preschool age. We examined predicted Total psychomotor development score, predicted Developmental disability index, calculated Morphometric vitality index, and predicted Intelligence quotient, Maze test, and Neurologic examination optimality score in a large prospective screening (cranial ultrasound screening, n=5,301) and validation cohort (n=508,926). Study design: In a single-center cohort observational study design (data collection done from 1984-1988, analysis done in 2022), a prospective cranial ultrasound screening study (1984-1988) was carried out on 5,301 live-born infants, including 571 (10.8%) preterm infants (≤36 weeks gestation), on the day of discharge of the mother at 5 to 8 days postpartum from a level 3 perinatal center. Predicted psychomotor development as assessed by predicted Total psychomotor development score, predicted Developmental disability index, calculated Morphometric vitality index, and predicted Intelligence quotient, Maze test, and Neurologic examination optimality score, was calculated. We related growth variables and obstetrical risk factors to Psychomotor development indices, and calculated Morphometric vitality index using odds ratios, receiver operating characteristics, analysis of variance, and multivariate analysis of variance. Results: The key result of our study is the observation that simple morphometric measures from newborns at birth like weight/head circumference ratio predict overall psychomotor development at 4.3 years (standard deviation, 0.8) of preschool age. Psychomotor development was assessed by predicted Total psychomotor development score, predicted Intelligence quotient, Maze test, and Neurologic examination optimality score, and related to weight/head circumference ratio in linear regression (P<.001) and ROC curve analyses (P<.001). Further, white matter damage strongly predicted adverse outcome in predicted Developmental disability index (P<.001). There was also a close correlation between calculated Morphometric vitality index and predicted Developmental disability index (P<.001). Finally, brain body weight ratio, weight/head circumference ratio, preterm birth, reduced Apgar at 10 minutes, weight/length ratio, and white matter damage yielded highest odds ratios for adverse outcome in predicted Total psychomotor development score and in predicted Developmental disability index (P<.001) and high effect sizes in reduced predicted Intelligence quotient, Maze test, and Neurologic examination optimality scores. Conclusion: Simple morphometric data, birth variables, and obstetrical risk factors bear predictive capacity for neurocognitive performance in children at 4.3 years (standard deviation, 0.8) of age and hence provide a basis for parental consultation and early intervention to improve school performance, educational success, and mental health in developed and developing countries.

PMID: 37719644

22. The value of continuing research on epidemiology of cerebral palsy (CP) - What have we learned?

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No abstract available

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