# Cerebral palsy research news

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CP Alliance Chair of Cerebral Palsy Research

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## Interventions and Management

### 1. Acute unexplained T1-T2 fracture-dislocation following posterior instrumentation and fusion for paralytic scoliosis

Joe Rassi, Diane Ghanem, Aren Joe Bizdikian, Mohammad Daher, Gaby Kreichati, Ismat Ghanem

Case Reports Int J Surg Case Rep. 2023 Nov 29:114:109099. doi: 10.1016/j.ijscr.2023.109099. Online ahead of print.

Introduction and importance: Upper thoracic fracture-dislocation following posterior instrumentation and fusion is rare, with potentially devastating neurologic consequences. The recommended treatment is an open reduction, spinal cord decompression, and a proximal extension of spinal instrumentation. To report the diagnosis and management of an acute non-traumatic T1-T2 fracture-dislocation, occurring in the early postoperative course of a posterior instrumentation and fusion for neurogenic scoliosis. Case report: A 12-year-old spastic quadriplegic cerebral palsy (CP) male patient, who underwent an uneventful T2-S1 instrumentation for scoliosis, presented to the emergency department (ED) 2 weeks later, with mild fever, urinary retention, fecaloma and hypotonia of the 4 limbs, of few days duration. His parents reported no history of trauma and denied epileptic seizures. Atypical cervicothoracic spastic movements the night preceding his symptoms were the only relevant events of the patient's history. CT and MRI were both suggestive of a complete T1-T2 fracture-dislocation and spinal cord compromise. Clinical discussion: The patient underwent immediate posterior decompression with wide lamino-arthrectomy, open reduction and proximal extension of his posterior instrumentation to C5. One year following surgery, there was only mild sensorimotor and bladder and bowel function recovery. Conclusion: To our knowledge, this is the first report of an acute non-traumatic unexplained T1-T2 fracture-dislocation following posterior instrumentation and fusion. Despite a proper management, only very mild recovery was observed one year following surgery.

PMID: 38041890

### 2. Effects of Vojta Therapy on the Motor Function of Children with Neuromotor Disorders: Study Protocol for a Randomized Controlled Trial

Mónica Menéndez-Pardiñas, Miguel Alonso-Bidegaín, Fernando Santonja-Medina, Juan Luis Sánchez-González, Jose Manuel Sanz-Mengibar

J Clin Med. 2023 Nov 28;12(23):7373. doi: 10.3390/jcm12237373.

Background: Infantile cerebral palsy is a neurological pathology that causes great morbidity, mortality, and disability in people who suffer from it, mainly affecting motor development. There are a multitude of non-pharmacological methods or therapies for its treatment. One of the main methods is Vojta therapy. This methodology acts on ontogenetic postural function and automatic postural control. Objective: This study aims to demonstrate that there are changes in the motor development of children with cerebral palsy with the application of Vojta therapy. Methods and analysis: This is a randomized controlled trial on the effectiveness of two neurorehabilitation techniques in patients with cerebral palsy conducted at the Physical Medicine and Rehabilitation Service of the Teresa Herrera Maternal and Child Hospital of the A Coruña and Cee Health Area. The study will be conducted from January 2023 to December 2024. There will be two groups: the Vojta therapy group (n = 30) and the conventional physiotherapy group (n = 30). The measurement variables will be gross motor function as measured by the Gross Motor Function Measure (GMFM) and Infant Motor Profile (IMP) scales. Ethics and dissemination: The study was approved by the Research Ethics Committee of the University of Murcia (1823/2018) and Comité de Ética de la Investigación de A Coruña-Ferrol (2022/099). Trial registration number: ClinicalTrials.gov; identifier: NCT06092619.

PMID: 38068424

### 3. From Hip Screening to Hip Surveillance: Transforming Care for Patients With Cerebral Palsy: An Analysis of a Single Institution

Alana Sadur, Curt Martinez, Sarah Dance, Ryan Travers, Ariana Gonzalez, Sean A Tabaie

J Am Acad Orthop Surg Glob Res Rev. 2023 Dec 8;7(12):e23.00236. doi: 10.5435/JAAOSGlobal-D-23-00236. eCollection 2023 Dec 1.

Introduction: Surveillance programs aimed at monitoring hip displacement in patients with cerebral palsy have been demonstrated to decrease the incidence of hip dislocations and properly time surgical intervention, ultimately improving patient outcomes. The objective of this study was to determine whether the implementation of a hip screening to surveillance program at a tertiary academic teaching hospital in 2017 increased the frequency of radiographic evaluations and changed the timing of surgical intervention. Methods: A total of 592 patients with cerebral palsy were identified, and 468 of these patients had initial radiograph date data available. In this analysis, 246 patients with initial radiograph dates after 2012 were included. The study population was divided into two groups based on the initial radiograph date, 2012 to 2016 versus 2017 to 2022. One hundred sixty patients (65%) were in the 2012 to 2016 group, and 86 (35%) were in the 2017 to 2022 group. Statistical analysis was conducted using various techniques, such as two-sample Student t-test, Mann-Whitney U test, chi square/Fisher exact test, and multivariable linear regression analysis. Results: The average number of radiographs per year in the 2017 to 2022 group was 0.11 (95% CI: 0.02, 0.20, P = 0.017) higher than the 2012 to 2016 group. After adjusting for confounders using multivariable linear regression analysis, this difference was even larger (difference 0.16, 95% CI: 0.06, 0.25, P = 0.001). The surgical intervention rate was significantly lower in the 2017 to 2022 group compared with the 2012 to 2016 group (12.9% versus 40.6%, P < 0.001). Discussion: The results of this study suggest that the implementation of a hip screening to surveillance program results in more frequent radiographic evaluations and possibly a reduced need for surgical intervention from 2017 to 2022. In the 2012 to 2016 group, more surgical interventions were performed likely because of the lack of any hip surveillance or screening program in place.

PMID: 38063442

### 4. Arthrodesis of the first metatarsophalangeal joint for severe hallux valgus in adolescents with cerebral palsy: A retrospective comparison study of three surgical techniques

Ken Ye, Megan Cashin, Samuel K Van de Velde, Abhay Khot, Kerr Graham, Erich Rutz

J Child Orthop. 2023 Oct 5;17(6):607-617. doi: 10.1177/18632521231200060. eCollection 2023 Dec.

Purpose: We compared the outcomes of arthrodesis of the first metatarsophalangeal joint for severe hallux valgus in 31 adolescents with cerebral palsy, using three different methods of fixation: K-wires, non-locking plates, and locking plates. Methods: Clinical outcomes included time to weight-bearing, fusion rates and surgical complications. Radiographic assessment included comparing pre- and post-operative hallux valgus angles, intermetatarsal angles, interphalangeal angles, and lateral metatarsophalangeal angles. Patient-reported outcomes included pre- and post-operative visual analogue scales addressing bunion pain and concerns, difficulties with wearing shoes and braces, and difficulties with foot hygiene. Results: Of the 31 adolescents (16 male), 10 patients had K-wire fixation, 11 had a non-locking dorsal plate, and 10 had fixation with a dorsal locking plate. Mean age at surgery was 16 years (12-18 years) and mean follow-up was 4 years (2.7-6.5 years). Patients with K-wire fixation had delayed weight-bearing and had more complications than those managed by dorsal plating. There were significant improvements in radiographic parameters (except interphalangeal angle) and in patient-reported outcomes, in all groups (p < 0.001). However, radiographic and clinical outcomes were better in the dorsal plating groups compared to the K-wire group. Conclusion: Arthrodesis of the first metatarsophalangeal joint gave good correction of deformity with improvements in symptoms and radiographic parameters in adolescents with cerebral palsy. We recommend dorsal plating that allowed early weight-bearing and had fewer complications with better clinical and radiographic outcomes, than K-wire fixation. Level of evidence: IV: Retrospective case series.

PMID: 38050598

### 5. ISB clinical biomechanics award winner 2023: Medial gastrocnemius muscle and Achilles tendon interplay during gait in cerebral palsy

Francesco Cenni, Nathalie Alexander, Maria Sukanen, Afet Mustafaoglu, Zhongzheng Wang, Ruoli Wang, Taija Finni

Clin Biomech (Bristol, Avon). 2023 Dec 6:111:106158. doi: 10.1016/j.clinbiomech.2023.106158. Online ahead of print.

Background: The interplay between the medial gastrocnemius muscle and the Achilles tendon is crucial for efficient walking. In cerebral palsy, muscle and tendon remodelling alters the role of contractile and elastic components. The aim was to investigate the length changes of medial gastrocnemius belly and fascicles, and Achilles tendon to understand their interplay to gait propulsion in individuals with cerebral palsy. Methods: Twelve young individuals with cerebral palsy and 12 typically developed peers were assessed during multiple gait cycles using 3D gait analysis combined with a portable ultrasound device. By mapping ultrasound image locations into the shank reference frame, the medial gastrocnemius belly, fascicle, and Achilles tendon lengths were estimated throughout the gait cycle. Participants with cerebral palsy were classified into equinus and non-equinus groups based on their sagittal ankle kinematics. Findings: In typically developed participants, the Achilles tendon undertook most of the muscle-tendon unit lengthening during stance, whereas in individuals with cerebral palsy, this lengthening was shared between the medial gastrocnemius belly and Achilles tendon, which was more evident in the equinus group. The lengthening behaviour of the medial gastrocnemius fascicles resembled that of the Achilles tendon in cerebral palsy. Interpretation: The findings revealed similar length changes of the medial gastrocnemius fascicles and Achilles tendon, highlighting the enhanced role of the muscle in absorbing energy during stance in cerebral palsy. These results, together with the current knowledge of increased intramuscular stiffness, suggest the exploitation of intramuscular passive forces for such energy absorption.

PMID: 38061205

### 6. Audiovisual biofeedback amplifies plantarflexor adaptation during walking among children with cerebral palsy

Alyssa M Spomer, Benjamin C Conner, Michael H Schwartz, Zachary F Lerner, Katherine M Steele

J Neuroeng Rehabil. 2023 Dec 8;20(1):164. doi: 10.1186/s12984-023-01279-5

Background: Biofeedback is a promising noninvasive strategy to enhance gait training among individuals with cerebral palsy (CP). Commonly, biofeedback systems are designed to guide movement correction using audio, visual, or sensorimotor (i.e., tactile or proprioceptive) cues, each of which has demonstrated measurable success in CP. However, it is currently unclear how the modality of biofeedback may influence user response which has significant implications if systems are to be consistently adopted into clinical care. Methods: In this study, we evaluated the extent to which adolescents with CP (7M/1F; 14 [12.5,15.5] years) adapted their gait patterns during treadmill walking (6 min/modality) with audiovisual (AV), sensorimotor (SM), and combined AV + SM biofeedback before and after four acclimation sessions (20 min/session) and at a two-week follow-up. Both biofeedback systems were designed to target plantarflexor activity on the more-affected limb, as these muscles are commonly impaired in CP and impact walking function. SM biofeedback was administered using a resistive ankle exoskeleton and AV biofeedback displayed soleus activity from electromyography recordings during gait. At every visit, we measured the time-course response to each biofeedback modality to understand how the rate and magnitude of gait adaptation differed between modalities and following acclimation. Results: Participants significantly increased soleus activity from baseline using AV + SM (42.8% [15.1, 59.6]), AV (28.5% [19.2, 58.5]), and SM (10.3% [3.2, 15.2]) biofeedback, but the rate of soleus adaptation was faster using AV + SM biofeedback than either modality alone. Further, SM-only biofeedback produced small initial increases in plantarflexor activity, but these responses were transient within and across sessions ($p > 0.11$). Following multi-session acclimation and at the two-week follow-up, responses to AV and AV + SM biofeedback were maintained. Conclusions: This study demonstrated that AV biofeedback was critical to increase plantarflexor engagement during walking, but that combining AV and SM modalities further amplified the rate of gait adaptation. Beyond improving our understanding of how individuals may differentially prioritize distinct forms of afferent information, outcomes from this study may inform the design and selection of biofeedback systems for use in clinical care.

PMID: 38062454

### 7. The effect of uneven surfaces on inter-joint coordination during walking in children with cerebral palsy

C Dussault-Picard, Y Cherni, A Ferron, M T Robert, P C Dixon

Sci Rep. 2023 Dec 8;13(1):21779. doi: 10.1038/s41598-023-49196-w.

Clinical gait analysis on uneven surfaces contributes to the ecological assessment of gait deviations of children with spastic cerebral palsy (CP). Walking on uneven surfaces requires specific motor strategies, which can be assessed by lower-limb kinematic and inter-joint coordination analyses. This study aimed to assess and compare kinematics and inter-joint coordination between children with CP and their typically developing (TD) peers when walking on even and two levels of uneven surfaces (medium and high). A total of 17 children with CP and 17 TD children (11.5 ± 3.5 and 10.4 ± 4.5 years old, respectively) were asked to complete 6-8 gait trials on a 4-m walkway of each surface ($n = 3$) in randomized blocks while fit with retro-reflective markers on their lower-limbs. Children with CP showed proximal gait adaptations (i.e., hip and knee) on uneven surfaces. Compared with the TD group, the CP group showed decreased hip extension during late stance (49-63%, $d = 0.549$, $p < 0.001$), and a more in-phase knee-hip coordination strategy during swing phase (75-84% of gait cycle, $d = 1.035$, $p = 0.029$ and 92-100%, $d = 1.091$, $p = 0.030$) when walking on an uneven (high), compared to even surface. This study provides a better understanding of kinematic strategies employed by children with spastic CP when facing typical daily life gait challenges. Further studies are needed to evaluate the benefits of integrating uneven surfaces in rehabilitation care.

PMID: 38066308

### 8. Translating scientific recommendations into reality: a feasibility study using group-based high-intensity functional exercise training in adolescents with cerebral palsy

Alice Minghetti, Michèle Widmer, Elke Viehweger, Ralf Roth, Ramon Gysin, Martin Keller

Disabil Rehabil. 2023 Dec 2:1-10. doi: 10.1080/09638288.2023.2290204. Online ahead of print.

Purpose: To examine the feasibility and effects of a functional high-intensity exercise intervention performed in a group-setting on functionality, cardiovascular health and physical performance in adolescents with cerebral palsy (CP). Methods: Ten adolescents with a diagnosis of CP (2 females; 16.6 ± 3.4 years; GMFCS: I-II) participated in a 12-week training intervention, containing progressive resistance training using free weights and high-intensity workouts twice a week. The six-minute walking test, arterial stiffness and physical performance (strength and power tests) were measured before and after the intervention. Results: No adverse events were reported. We measured small increases in the six-minute walking test ($\Delta = 28.8$ m, 95% CI [-1.78;52.7]; $g = 0.34$ [-0.04;0.72]) and a small reduction in arterial stiffness ($\Delta = -4.65\%$ [-10.90;1.25]; $g = -0.46$ [-1.36;0.21]). All measures of physical performance increased ($0.24 \leq g \leq 0.88$). Conclusion: Functional training with free weights in high-functioning adolescents with CP is safe and effective in increasing parameters of physical performance and cardiovascular health. Positively influenced indicators of everyday independence (i.e. strength parameters) showed a transfer into movements of daily life. Concerns about adverse events through high-intensity training in adolescents with CP appear unjustified when training is performed progressively, following basic training principles.

PMID: 38042988

### 9. Effectiveness of whole-body vibration in patients with cerebral palsy: A systematic review and meta-analysis

Yong-Gu Han, Myoung-Kwon Kim

Meta-Analysis Medicine (Baltimore). 2023 Dec 1;102(48):e36441. doi: 10.1097/MD.0000000000036441.

Background: This study examined the effects of systemic vibration exercises on cerebral palsy patients. Methods: Literature published in Cumulated Index to Nursing and Allied Health Literature (CINAHL), Cochrane library, Embase, Physiotherapy Evidence Database (PEDro), and PubMed was reviewed. A total of 2978 studies were initially retrieved. After further reading of the full texts 17 articles were finally included. A quality assessment of the included studies was conducted using the risk of bias (RoB) 2.0, and the Funnel plot and the Egger test were conducted to confirm the publication bias. Subgroup analysis was carried out according to the dependent variables, the international classification of functioning, disability, and health (ICF), frequency, treatment period and age. Results: The overall effect size of homogeneity was 0.474 (CI = 0.148-0.801). The analysis of the dependent variables showed the following order of the effect size: balance, muscle strength, spasticity, bone density, range of motion of the joint, gait function, and motor function. In the ICF classification, the effect size was observed to follow the order of body structure and function, activity, and participation. The effect size in the intervention according to the treatment period showed the following order: 7 to 12 weeks, 1 to 6 weeks, and 14 to 24 weeks. The age-dependent classification showed the following order in the effect size: school age, adolescent and adult, and infant and school age. Conclusions: Systemic vibration is the most effective intervention to improve the balance and gait in patients with cerebral palsy and improve the body structure and function according to the ICF.

PMID: 38050249

### 10. An Exploration of Tri-Axial Accelerometers in Assessing the Therapeutic Efficacy of Constraint-Induced Movement Therapy in Children with Unilateral Cerebral Palsy

Youngsub Hwang, Jeong-Yi Kwon, Yoonju Na

Sensors (Basel). 2023 Nov 24;23(23):9393. doi: 10.3390/s23239393.

This study aimed to elucidate the role of tri-axial accelerometers in assessing therapeutic interventions, specifically constraint-induced movement therapy (CIMT), in children with unilateral cerebral palsy (UCP). The primary focus was understanding the correlation between the actigraphy metrics recorded during CIMT sessions and the resultant therapeutic outcomes. Children with UCP, aged between 4 and 12 years, participated in this study from July 2021 to December 2022. In conjunction with in-clinic sessions, during which participants wore tri-axial accelerometers on both limbs, we analyzed actigraphy data over three days of routine activities pre- and post-CIMT. While not all metrics derived from the accelerometers indicated significant improvements post-intervention, there was a clear trend towards a more balanced usage of both limbs, particularly evident in Axis 3, associated with vertical movement ($p = 0.017$). Additionally, a discernible correlation was observed between changes in the magnitude ratio derived from actigraphy data during CIMT (Δweek3-week1) and variations in traditional assessments pre- and post-intervention (ΔT0-T1), specifically the Assisting Hand Assessment grasp and release. Using tri-axial accelerometers has helped clarify the potential impacts of CIMT on children with UCP. The preliminary results suggest a possible link between actigraphy metrics taken during CIMT and the subsequent therapeutic outcomes determined by standardized tests.

PMID: 38067766

### 11. Profiling the Longitudinal Development of Babbling in Infants with Cerebral Palsy: Validation of the Infant Monitor of Vocal Production (IMP) Using the Stark Assessment of Early Vocal Development-Revised (SAEVD-R)

Roslyn Ward, Neville Hennessey, Elizabeth Barty, Robyn Cantle Moore, Catherine Elliott, Jane Valentine

Diagnostics (Basel). 2023 Nov 23;13(23):3517. doi: 10.3390/diagnostics13233517.

Aim: We compared early vocal development in children "at risk" for cerebral palsy (CP) with typically developing (TD) infants aged 6 to 15 months using the SAEVD-R, investigating potential pre-linguistic markers of communication impairment. Additionally, we sought to examine the agreement between the SAEVD-R and IMP, which uses parent report, in identifying departure from typical vocal development in at-risk infants. Method: Utilising a longitudinal cohort study, >10,000 vocalisations of 33 infants (15 at risk for CP and 18 TD) were assessed at 6, 9, 12, and 15 months using the SAEVD-R. Generalised linear mixed models (GLMMs) compared groups, and Spearman correlations explored IMP ceiling scores and SAEVD-R measures. Results: At 6 months, both TD and CP groups reached SAEVD-R vocalisation level 3 (expansion). By 9 months, 51% of TD infants progressed to advanced babbling (levels 4 and 5), while 80% of at-risk infants remained at level 3. At 12 and 15 months, over 90% of TD children advanced, compared to 67% at 12 months and 53% at 15 months for at-risk infants, who stayed at the pre-canonical stage. Strong correlations were found between IMP scores and vocalisation levels at 9 and 12 months. Remaining at the pre-canonical stage at 12 months correlated with delayed vocal development as per IMP scores. Interpretation: TD infants achieved higher SAEVD-R levels than at-risk infants. At 12 months, IMP scores effectively identified infants with speech-like vocalisation difficulties, demonstrating its clinical utility in identifying atypical vocal development in infants at risk for CP.

PMID: 38066758

### 12. Revisiting Dysarthria Treatment Across Languages: The Hybrid Approach

Erika S Levy, Gemma Moya-Galé

J Speech Lang Hear Res. 2023 Dec 6:1-10. doi: 10.1044/2023_JSLHR-23-00629. Online ahead of print.

Purpose: Ten years after Miller and Lowit's (2014) groundbreaking book providing a cross-linguistic perspective on motor speech disorders, we ask where we are regarding dysarthria treatment across languages in two specific populations: adults with Parkinson's disease (PD) and children with cerebral palsy (CP). Method: In this commentary, we consider preliminary evidence for both language-independent and language-specific approaches to treatment and propose a hybrid approach to speech treatment across languages, centered on the individual with dysarthria who speaks any given language. Conclusions: Treatment research on individuals with dysarthria secondary to PD and CP is advancing, but several areas remain to be explored. Next steps are suggested for addressing the paucity and complexity of cross-linguistic speech treatment research.

PMID: 38056466

### 13. Validity of measurement of femoral anteversion angle using FEMORA® software based on two-dimensional radiographic imaging examination femur in children with cerebral palsy in Indonesia

Tri Wahyu Martanto, Yusuf Rizal, Irwanto, Sulis Bayu Sentono, Rosy Setiawati, Sri Andreani Utomo, Prastiya Indra Gunawan, Nurul Kusuma Wardani, Prima Hari Nastiti, Rachmat Agung Widodo, Moon Seok Park, Arif Zulkarnain, Hizbillah Yazid, Hendra Cahaya Kumara, Muhammad Ihsan Kitta

Heliyon. 2023 Nov 11;9(11):e22243. doi: 10.1016/j.heliyon.2023.e22243. eCollection 2023 Nov.

Introduction: Children with spastic cerebral palsy (CP) often show an increase in femoral anteversion angle (FAA). Computed tomography (CT) scan is the main modality for evaluating FAA in these patients, however, due to significant radiation exposure, it carries a high carcinogenic risk. FEMORA® software is expected to be able to accurately assess FAA even with conventional X-ray images that only require low radiation exposure. However, its validity has not been tested in various populations or CT devices. This study aimed to validate the FEMORA® software by comparing it to CT scans done on an Indonesian population. Material and methods: All spastic CP patients of the outpatient clinic at Dr. Soetomo Hospital between March and November 2022, were included. The FEMORA® Software evaluation was performed by three examiners. The calculation results were averaged and compared with those of the CT scan. Intraclass correlation coefficient (ICC), reliability, and correlation were be assessed. Results: There were 36 patients included in this study. Most were female (n = 22; 61,1 %) and the average age was 7,28 years old. Interobserver preoperative analysis using ICC showed good outcomes (p = 0.918; 95 % CI, 0.858-0.955). FAA measurement results using FEMORA® and CT scans were 41,71 ± 12,90 and 32,68 ± 11,85, respectively. Correlation coefficient between the two values is 0.634 (p < 0.001). Conclusion: FEMORA® software demonstrates a good and significant correlation with FAA measurement using CT scan.

PMID: 38045176

### 14. Telehealth services for children with neuro-developmental disabilities in the Asia-Pacific region: A systematic review

Bhagya Devagiri, Akila Dinethra K Ariyasena, Dhammika Deepani Siriwardhana, Samanmali Sumanasena

Review Child Care Health Dev. 2023 Dec 4. doi: 10.1111/cch.13192. Online ahead of print.

Background: In recent years telehealth became a popular and a rational health service delivery approach, especially amidst multiple challenges posed while providing health care interventions during the COVID-19 pandemic. Aim: We synthesized available evidence on telehealth for managing children with NDDs in the Asia-Pacific region with the aim of identifying successful methods. Methodology: We systematically reviewed six electronic databases: MEDLINE, AMED, EMBASE, PsychInfo, Web of Science, and (CINAHL plus) using the keywords and database-specific subject headings from their inception to 25th August 2021. Review findings were synthesized narratively, and harvest plots were used to demonstrate the effect of interventions. The protocol and reporting the findings of this review adhered to PRISMA 2020 guidelines. PROSPERO registration: CRD 340690. Results: We harvested 30,823 records; 17,563 duplicates were removed, and 196 full-text articles were assessed for eligibility. 16 studies with multiple research designs were included in the review. Eight were from the Pacific region and eight represented Asia. The interventions targeted families and children with a variety of NDDs (autism spectrum disorder, Down syndrome, cleft lip and palate, hearing impairment, cerebral palsy etc.) via telehealth. Telehealth packages consisted of direct and indirect methods of synchronous, asynchronous, and hybrid approaches. All studies used parent-led intervention strategies. Telehealth reported a positive effect in 7/16 studies while five showed a neutral effect. Conclusions: According to published evidence telehealth for children with NDDs is an evolving, evidence-based service facilitation modality in the Asia-Pacific region, with only a few published randomized controlled trials. The systematic review shows promising telehealth practices emerging across the region despite the diversity in NDDs studied.

PMID: 38049949

### 15. Caregiver's Quality of Life Among Children with Cerebral Palsy in the Kingdom of Saudi Arabia, and Various Influencing Factors: A Single Cohort Study

Jaya Shanker Tedla, Faisal Asiri, Ravi Shankar Reddy, Kumar Gular, Venkata Nagaraj Kakaraparthi, Devika Rani Sangadala, Snehil Dixit, Ahlam Mohammed Alamri, Vijaya Kumar Narne, Rawan Abdullah Mohammed Alasmari, Omer Abdullah Dhafer, Mohammed Ahmed Al Shamer

J Multidiscip Healthc. 2023 Nov 29:16:3705-3714. doi: 10.2147/JMDH.S440190. eCollection 2023.

Purpose: Quality of life (QOL) among disabled children and their caregivers is an important concern in healthcare. We aim to evaluate the quality of life among caregivers of children with cerebral palsy and to observe the effects of various demographic factors and affected child-related factors on caregivers' quality of life. Patients and methods: After ethical approval and written consent was obtained from the participants. One hundred six caregivers of children with cerebral palsy from the Asir region were recruited for the study. Caregivers provided details, including their demographic characteristics, social factors, and information regarding their affected children regarding age, gender, mobility levels, etc. They also completed the Arabic version of the World Health Organization Quality of Life-BREF (WHOQOL-BREF) questionnaire for assessing their QOL. Results: All the caregivers were women; their mean age was 40.38 years and SD7.09, and the overall QOL mean and standard deviations were 66.38 ± 12.88. There was a moderately significant correlation between total QOL in comparison with caregivers' educational level and mobility capacity, with R values of 0.54 (p<0.001) and 0.62 (p<0.001), respectively. Conclusion: All the subdomains of WHOQOL-BREF were found to be very closely related to the total scores for QOL. The caregivers of children with cerebral palsy had better QOL scores than the cutoff scores proposed in the WHOQOL-BREF scale. Factors such as increased mobility and education of the affected child contributed to better total QOL scores.

PMID: 38050486

### 16. Health care resource utilization and costs among patients with spasticity or cervical dystonia

Michael Hull, Natalya Danchenko, Vamshi Ruthwik Anupindi, Mitchell DeKoven, Jing He, Jonathan Bouchard

J Manag Care Spec Pharm. 2023 Dec 6:1-12. doi: 10.18553/jmcp.2023.22205. Online ahead of print.

Background: Spasticity and cervical dystonia (CD) are movement disorders with considerable direct and indirect health care cost implications. Although several studies have discussed their clinical impact, few have calculated the economic burden of these disorders. Objective: To assess the all-cause health care resource utilization (HCRU) and costs in adults and children with spasticity or CD. Methods: This retrospective, observational cohort-based study was conducted using administrative insurance claims from the IQVIA PharMetrics Plus database from October 1, 2015, to December 31, 2019. Patients were selected based on International Classification of Diseases, Tenth Revision, Clinical Modification diagnosis codes for first evidence of spasticity (associated with a spasticity etiology) or CD (index date) during the selection window, from April 1, 2016, through December 31, 2018. Cases were stratified into 3 mutually exclusive cohorts: adult patients with spasticity, pediatric patients with spasticity, and patients with CD; those with spasticity who had a history of stroke or cerebral palsy were also evaluated in subcohorts. Patients without evidence of spasticity or CD during the study period were identified as a matched comparator group and were randomly assigned an index date. Patients with spasticity were matched 1:1 to the comparator group based on age, sex, index year, and payer type using descriptive analyses. Results: 215,739 adult patients with spasticity, 29,644 pediatric patients with spasticity, and 9,035 adult patients with CD were identified after matching. Adult patients with spasticity and CD had mean (SD) ages of 48.4 (15.6) years and 48.0 (13.1) years, respectively. Stroke was identified in 31.9% (n = 68,928) of adult patients with spasticity, and cerebral palsy was identified in 11.3% (n = 3,364) of pediatric patients with spasticity. Adult and pediatric patients with spasticity and patients with CD had significantly higher HCRU (including mean number of outpatient, emergency department, and inpatient visits and proportions of patients with prescription fills) and higher mean total health care costs per patient (adult patients with spasticity $29,912 vs $7,464; pediatric patients with spasticity $16,089 vs $2,963; and patients with CD $20,168 vs $7,141) than matched comparators (all P<0.0001). Conclusions: The management of patients with spasticity or CD results in considerably higher health care expenses. Within managed health care systems, more effective management of spasticity and CD in adult and pediatric patients represents a significant opportunity for cost savings.

PMID: 38055046

### 17. Clinical features associated with epilepsy occurrence, resolution, and drug resistance in children with cerebral palsy: A population-based study

Nimra Feroze, Tasneem Karim, Katarina Ostojic, Sarah Mcintyre, Elizabeth H Barnes, Byoung Chan Lee; Cerebral Palsy Alliance study group; Russell C Dale, Deepak Gill, Kavitha Kothur

Dev Med Child Neurol. 2023 Dec 7. doi: 10.1111/dmcn.15807. Online ahead of print.

Aim: To investigate clinicoradiological features associated with epilepsy, its resolution, and drug resistance in children with cerebral palsy (CP). Method: Data were gathered from the New South Wales/Australian Capital Territory CP Register, encompassing children with CP born between 2003 and 2015 (n = 1916). Clinical features and the severity of impairments were compared among three groups: children with current epilepsy (n = 604), those with resolved epilepsy by age 5 years (n = 109), and those without epilepsy (n = 1203). Additionally, a subset of the registry cohort attending Children's Hospital Westmead (n = 256) was analysed to compare epilepsy and treatment characteristics between drug-responsive (n = 83) and drug-resistant groups (n = 147) using logistic regression and hierarchical cluster analysis. Results: Manual Ability Classification System levels IV and V, intellectual impairment, and vision impairment were found to be associated with epilepsy in children with CP on multivariable analysis ($p < 0.01$). Moderate to severe intellectual impairment and bilateral spastic CP were independent positive and negative predictors of epilepsy persistence at the age of 5 years respectively ($p < 0.05$). Microcephaly and multiple seizure types were predictors of drug-resistant epilepsy (area under the receiver operating characteristic curve of 0.83; 95% confidence interval 0.77-0.9). Children with a known genetic cause (14%) and CP epilepsy surgery group (4.3%) formed specific clinical subgroups in CP epilepsy. Interpretation: Our study highlights important clinical associations of epilepsy, its resolution, and treatment response in children with CP, providing valuable knowledge to aid in counselling families and identifying distinct prognostic groups for effective medical surveillance and optimal treatment.

PMID: 38059324

### 18. Gamification and neurological motor rehabilitation in children and adolescents: A systematic review

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Introduction: Gamification consists of the use of games in non-playful contexts. It is widely employed in the motor rehabilitation of neurological diseases, but mainly in adult patients. The objective of this review was to describe the use of gamification in the rehabilitation of children and adolescents with neuromotor impairment. Methods: We performed a systematic review of clinical trials published to date on the MEDLINE (PubMed), Scielo, SCOPUS, Dialnet, CINAHL, and PEDro databases, following the PRISMA protocol. The methodological quality of the studies identified was assessed using the PEDro scale. Results: From a total of 469 studies, 11 clinical trials met the inclusion criteria. We analysed the gamification systems used as part of the rehabilitation treatment of different neuromotor conditions in children and adolescents. Cerebral palsy was the most frequently studied condition (6 studies), followed by developmental coordination disorder (3), neurological gait disorders (1), and neurological impairment of balance and coordination (1). Conclusion: The use of gamification in rehabilitation is helpful in the conventional treatment of neuromotor disorders in children and adolescents, with increased motivation and therapeutic adherence being the benefits with the greatest consensus among authors. While strength, balance, functional status, and coordination also appear to improve, future research should aim to determine an optimal dosage.

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### 19. New Frontier: The First Year of an Adult Neurodevelopmental Disabilities Clinic

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Cogn Behav Neurol. 2023 Dec 11. doi: 10.1097/WNN.0000000000000362. Online ahead of print.

Background: As patients with neurodevelopmental disorders (NDDs) transition from pediatric to adult health care systems, they often have difficulty finding physicians to address their NDD-related needs. In response to this care gap, we established a new consultation clinic within a behavioral neurology clinic in an adult neurology department to address the neurodevelopmental concerns of these adult patients. Objective: To characterize the population of adult patients with NDDs seen in the adult NDD clinic in its first year. Method: Data were obtained by a retrospective chart review of all patients with NDDs seen in the adult NDD clinic from September 2020 through December 2021. Results: Of the 86 patients who were seen in the adult NDD clinic, the average age was 34 years (SD = 15, range = 18-74 years). Developmental diagnoses included intellectual disability (63%), autism spectrum disorder (47%), Down syndrome (15%), cerebral palsy (9%), and other genetic disorders (26%). Comorbidities addressed included behavioral concerns (34%), anxiety (29%), seizure disorders (22%), and depression (15%). Behavioral and/or mental health concerns prompted 65% of the initial clinic visits. The most common recommendation made was to begin or increase exercise (59%), followed by facilitating connection to community, social, and employment resources. Conclusion: Adults with NDDs have diagnoses, comorbidities, and concerns that are similar to, but also distinct from, those addressed in other adult neurology clinics. This study addresses the need for, and feasibility of, caring for the diverse population of adults with NDDs in an adult neurology setting.

PMID: 38063505

### 20. Childhood outcomes after maternal antenatal sildenafil treatment for severe early-onset fetal growth restriction: a randomized trial (STRIDER NZAus)

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In this follow-up at 2.5 years of children from the STRIDER NZAus Trial (N = 112), in which women with singleton pregnancies affected by severe early fetal growth restriction were randomized to sildenafil citrate 75 mg daily or placebo until 32 weeks, there was no difference between groups in survival without neurosensory impairment, defined as any of cerebral palsy, deafness, blindness, cognitive delay (Bayley III cognition or language score >1 SD below mean) or motor delay: 30/56 [54%] vs. 34/56[61%]; aOR = 0.74, 95%CI: 0.31, 1.77. However, children exposed to sildenafil appeared to be more likely to have cognitive delay (13/45[29%] vs. 4/40[10%]; aOR = 3.71, 95% CI: 1.01, 13.63) but less likely to have emotional-behavioural difficulties (2/43[5%] vs. 8/38[21%]; aOR = 0.19, 95%CI: 0.03, 1.00). Conclusion: maternal sildenafil treatment for severe early-onset FGR was not associated with altered survival free of neurosensory impairment at 2.5 years' corrected age.

PMID: 38057497

### 21. Editorial: The use of repetitive peripheral magnetic stimulation (rPMS) in neurological disorders and neurorehabilitation

Volker Zschorlich, Tomofumi Yamaguchi, Cyril Schneider

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No abstract available

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## 22. Genetic pathways in cerebral palsy: a review of the implications for precision diagnosis and understanding disease mechanisms

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Cerebral palsy is a diagnostic term utilized to describe a group of permanent disorders affecting movement and posture. Patients with cerebral palsy are often only capable of limited activity, resulting from non-progressive disturbances in the fetal or neonatal brain. These disturbances severely impact the child's daily life and impose a substantial economic burden on the family. Although cerebral palsy encompasses various brain injuries leading to similar clinical outcomes, the understanding of its etiological pathways remains incomplete owing to its complexity and heterogeneity. This review aims to summarize the current knowledge on the genetic factors influencing cerebral palsy development. It is now widely acknowledged that genetic mutations and alterations play a pivotal role in cerebral palsy development, which can be further influenced by environmental factors. Despite continuous research endeavors, the underlying factors contributing to cerebral palsy remain are still elusive. However, significant progress has been made in genetic research that has markedly enhanced our comprehension of the genetic factors underlying cerebral palsy development. Moreover, these genetic factors have been categorized based on the identified gene mutations in patients through clinical genotyping, including thrombosis, angiogenesis, mitochondrial and oxidative phosphorylation function, neuronal migration, and cellular autophagy. Furthermore, exploring targeted genotypes holds potential for precision treatment. In conclusion, advancements in genetic research have substantially improved our understanding of the genetic causes underlying cerebral palsy. These breakthroughs have the potential to pave the way for new treatments and therapies, consequently shaping the future of cerebral palsy research and its clinical management. The investigation of cerebral palsy genetics holds the potential to significantly advance treatments and management strategies. By elucidating the underlying cellular mechanisms, we can develop targeted interventions to optimize outcomes. A continued collaboration between researchers and clinicians is imperative to comprehensively unravel the intricate genetic etiology of cerebral palsy.

PMID: 38051892

## 23. How well does the EQ-5D-Y-5L describe children with intellectual disability?: "There's a lot more to my child than that she can't wash or dress herself."

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Value Health. 2023 Dec 1:S1098-3015(23)06196-X. doi: 10.1016/j.jval.2023.11.005. Online ahead of print.

Objectives: The EQ-5D-5L is a generic health utility instrument for measuring health-related quality of life (HRQoL), with self-report and proxy-report versions for children (EQ-5D-Y-5L). Children with intellectual disability (ID) are a heterogeneous population whose impairments and comorbidities place them at risk of poor HRQoL. This study aimed to describe the content validity and suitability for children with ID of a proxy-report version of the EQ-5D-Y-5L as seen by their caregivers. Methods: A proxy-report EQ-5D-Y-5L was administered to caregivers of children with ID. Using cognitive think-aloud interviewing, participants were encouraged to provide the reasoning for their choices, assess the questions' relevance, comprehensibility, and comprehensiveness, and comment on the tool's strengths and weaknesses. Qualitative content analysis used both directed (deductive) and conventional (inductive) methods. Results: There were 28 interviews with 30 caregivers of children with ID (aged 8 to 22 years, 17 boys, with autism spectrum disorder, cerebral palsy, Down syndrome, and rare genetic disorders). The EQ-5D-Y-5L was considered clear, concise, and largely relevant, but insufficiently comprehensive for this population. Interviewees sought clarification of the definition of HRQoL, whether it included unchanging impairments (versus fluctuating health states), and what basis of comparison to use (child or peer). Many interviewees suggested inclusion of questions for other domains, including communication and social engagement, equipment and human supports required, and a wider range of mental health questions. Conclusions: The study suggests that further work is required to ensure accurate responses to the EQ-5D-Y-5L from caregivers of children with ID and to describe these children adequately.

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