Cerebral palsy research news

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Professor Nadia Badawi AM
CP Alliance Chair of Cerebral Palsy Research

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Interventions and Management

1. Surgical approach to forearm pronation deformity in patients with cerebral palsy: a systematic review

Bram DE Lepeleere, Malcolm Forward, Manuel Martens, Frank Plasschaert

Acta Orthop Belg. 2023 Jun;89(2):183-194. doi: 10.52628/89.2.11048.

Background: Pronation deformity in patients with cerebral palsy can have a major impact on upper limb functionality. There is lack of consensus in the literature about the preferred surgical technique to address this deformity. Study aim: To evaluate and synthesize the outcome of different surgical techniques for pronation deformity in patients with cerebral palsy. Methodology: The databases MEDLINE and Embase were searched for publications up to December 2021. Articles were considered eligible for inclusion when the included patients had a pronation deformity caused by cerebral palsy and results of surgical intervention for pronation deformity were examined. Evaluation of the quality of the retrieved study was conducted using the MINORS tool. Meta-analysis was not possible due to the heterogeneity of interventions and reported outcomes. Results: Nineteen studies, involving 475 patients and eight different techniques were included. All studies reported gain of active supination in most patients. The effect of surgery on functional gain was less clear and there was a large heterogeneity of reported functional outcome measures. There were 46 reported complications. Overall quality of study design was poor, illustrated by the average MINOR score of 6.9/16. Overall, there is a high risk of bias due to poor internal and external validity of the studies. Conclusion: Despite positive reports on gain in supination and functionality after most procedures addressing pronation deformity in CP patients, no conclusions can be drawn concerning the preferred technique due to the low quality of the evidence.

PMID: 37924533

2. Upper extremity asymmetry due to nerve injuries or central neurologic conditions: a scoping review

Sandesh G Bhat, Alexander Y Shin, Kenton R Kaufman

Review J Neuroeng Rehabil. 2023 Nov 9;20(1):151. doi: 10.1186/s12984-023-01277-7.

Background: Peripheral nerve injuries and central neurologic conditions can result in extensive disabilities. In cases with unilateral impairment, assessing the asymmetry between the upper extremity has been used to assess outcomes of treatment and severity of injury. A wide variety of validated and novel tests and sensors have been utilized to determine the upper extremity asymmetry. The purpose of this article is to review the literature and define the current state of the art for describing upper extremity asymmetry in patients with peripheral nerve injuries or central neurologic conditions. Method: An electronic literature search of PubMed, Scopus, Web of Science, OVID was performed for publications between 2000 to 2022. Eligibility criteria were subjects with neurological conditions/injuries who were analyzed for dissimilarities in use between the upper extremities. Data related to study population, target condition/injury, types of tests performed, sensors used, real-world data collection, outcome measures of interest, and results of the study were extracted. Sackett's Level of Evidence was used to judge the quality of the articles. Results: Of the 7281 unique articles, 112 articles met the inclusion criteria for the review. Eight target conditions/injuries were identified (Brachial Plexus Injury, Cerebral Palsy, Multiple Sclerosis, Parkinson's Disease, Peripheral Nerve Injury, Spinal Cord Injury, Schizophrenia, and stroke). The tests performed were classified into

thirteen categories based on the nature of the test and data collected. The general results related to upper extremity asymmetry were listed for all the reviewed articles. Stroke was the most studied condition, followed by cerebral palsy, with kinematics and strength measurement tests being the most frequently used tests. Studies with a level of evidence level II and III increased between 2000 and 2021. The use of real-world evidence-based data, and objective data collection tests also increased in the same period. Conclusion: Adequately powered randomized controlled trials should be used to study upper extremity asymmetry. Neurological conditions other than stroke should be studied further. Upper extremity asymmetry should be measured using objective outcome measures like motion tracking and activity monitoring in the patient's daily living environment.

PMID: 37940959

3. Changes Induced by Early Hand-Arm Bimanual Intensive Therapy Including Lower Extremities in Young Children With

Rodrigo Araneda, Daniela Ebner-Karestinos, Julie Paradis, Anne Klöcker, Geoffroy Saussez, Josselin Demas, Rodolphe Bailly, Sandra Bouvier, Astrid Carton de Tournai, Herman Enimie, Aghiles Souki, Grégoire Le Gal, Emmanuel Nowak, Stephane V Sizonenko, Christopher J Newman, Mickael Dinomais, Inmaculada Riquelme, Andrea Guzzetta, Sylvain Brochard, Yannick Bleyenheuft

JAMA Pediatr. 2023 Nov 6:e234809. doi: 10.1001/jamapediatrics.2023.4809. Online ahead of print.

Importance: Intensive interventions are provided to young children with unilateral cerebral palsy (UCP), classically focused on the upper extremity despite the frequent impairment of gross motor function. Hand-Arm Bimanual Intensive Therapy Including Lower Extremities (HABIT-ILE) effectively improves manual dexterity and gross motor function in school-aged children. Objective: To verify if HABIT-ILE would improve manual abilities in young children with UCP more than usual motor activity. Design, setting, and participants: This prospective randomized clinical trial (November 2018 to December 2021), including 2 parallel groups and a 1:1 allocation, took place at European university hospitals, cerebral palsy specialized centers, and spontaneous applications at 3 sites: Brussels, Belgium; Brest, France; and Pisa, Italy. Matched (age at inclusion, lesion type, cause of cerebral palsy, and affected side) pairs randomization was performed. Young children were assessed at baseline (T0), 2 weeks after baseline (T1), and 3 months after baseline (T2). Health care professionals and assessors of main outcomes were blinded to group allocation. At least 23 young children (in each group) aged 12 to 59 months with spastic/dyskinetic UCP and able to follow instructions were needed. Exclusion criteria included uncontrolled seizures, scheduled botulinum toxin injections, orthopedic surgery scheduled during the 6 months before or during the study period, severe visual/cognitive impairments, or contraindications to magnetic resonance imaging. Interventions: Two weeks of usual motor activity including usual rehabilitation (control group) vs 2 weeks (50 hours) of HABIT-ILE (HABIT-ILE group). Main outcomes and measures: Primary outcome: Assisting Hand Assessment (AHA); secondary outcomes: Gross Motor Function Measure-66 (GMFM-66), Pediatric Evaluation of Disability Inventory-Computer Adaptive Test (PEDI-CAT), and Canadian Occupational Performance Measure (COPM). Results: Of 50 recruited young children (26 girls [52%], median age; 35.3 months for HABIT-ILE group; median age, 32.8 months for control group), 49 were included in the final analyses. Change in AHA score from T0 to T2 was significantly greater in the HABIT-ILE group (adjusted mean score difference [MD], 5.19; 95% CI, 2.84-7.55; P < .001). Changes in GMFM-66 (MD, 4.72; 95% CI, 2.66-6.78), PEDI-CAT daily activities (MD, 1.40; 95% CI, 0.29-2.51), COPM performance (MD, 3.62; 95% CI, 2.91-4.32), and satisfaction (MD, 3.53; 95% CI, 2.70-4.36) scores were greater in the HABIT ILE group. Conclusions and relevance: In this clinical trial, early HABIT-ILE was shown to be an effective treatment to improve motor performance in young children with UCP. Moreover, the improvements had an impact on daily life activities of these children.

PMID: 37930692

4. Treatment of Chronic Degenerative Hip Pain in a Male Patient With Cerebral Palsy Four Decades After Reverse Osteotomy

John B Hooks, David Dayya

Case Reports Cureus. 2023 Oct 4;15(10):e46495. doi: 10.7759/cureus.46495. eCollection 2023 Oct.

Patients with cerebral palsy (CP) frequently require surgical hip interventions in early adulthood due to spasticity-related gait abnormalities. In most instances, these cases are characterized by severe restrictions on mobility. This is the case of a male patient with CP who underwent right proximal femoral open reduction internal fixation (ORIF) and reverse osteotomy for right hip subluxation in young adulthood. Patients with CP who undergo total hip arthroplasty (THA) or ORIF with reverse osteotomy often require future revision. The patient was initially given an estimated 10-year longevity for his plate and screw construct (hardware). Forty-four years later, the patient presented with debilitating chronic bilateral hip pain, requiring the assistance of a cane for ambulation. There is a limited body of knowledge on ORIF and reverse osteotomy follow-up in patients with CP within a 30- to 50-year period. At the 44th-year follow-up, CT and X-ray imaging found postoperative changes in the right femur, including intact hardware, bilateral acetabular dysplasia, right femoral stress fracture, progression of hip arthritis, and right iliopsoas bursitis. Surgery for hardware revision was not indicated. Gradual restoration of function was achieved over

a 14-month period with conservative management. This case suggests that physical therapy (PT), exercise, and sporadic non-steroidal anti-inflammatory drug (NSAID) use are effective for improving chronic degenerative changes, associated bursitis, and loss of function in patients who developed CP-induced gait complications in young adulthood. These improvements can be made several decades after undergoing ORIF and osteotomies. This course of treatment was effective in improving the patient's quality of life without additional surgical interventions.

PMID: 37927768

5. Attention-Based Deep Recurrent Neural Network to Estimate Knee Angle During Walking from Lower-Limb EMG

Mohamed Abdelhady, Diane L Damiano, Thomas C Bulea

IEEE Int Conf Rehabil Robot. 2023 Sep:2023:1-6. doi: 10.1109/ICORR58425.2023.10304604.

Accurate prediction of joint angle during walking from surface electromyography (sEMG) offers the potential to infer movement intention and therefore represents a potentially useful approach for adaptive control of wearable robotics. Here, we present the use of a recurrent neural network (RNN) with gated recurrent units (GRUs) and an attention mechanism to estimate knee angle during overground walking from sEMG and its initial offline validation in healthy adolescents. Our results show that the attention mechanism improved estimation accuracy by focusing on the most relevant parts of the input dataset within each time window, particularly muscles active during knee excursion. Sensitivity analysis revealed knee extensor and flexor muscles to be most salient in accurately estimating joint angle. Additionally, we demonstrate the ability of the GRU-RNN approach to accurately estimate knee angle during overground walking in a child with cerebral palsy (CP) in the presence of exoskeleton knee extension assistance. Collectively, our findings establish the initial feasibility of using this approach to estimate user movement from sEMG, which is particularly important for developing robotic exoskeletons for children with neuromuscular disorders such as CP.

PMID: 37941224

6. Asymmetry in sensory-motor function between the lower limbs in children with hemiplegic cerebral palsy: An observational study

Hsiu-Ching Chiu, Louise Ada, Rong-Ju Cherng, Chiehfeng Chen

Observational Study Chin J Physiol. 2023 Sep-Oct;66(5):345-350. doi: 10.4103/cjop.CJOP-D-23-00057.

The objective of this study was to examine the difference in sensory-motor impairments (i.e., balance, contracture, coordination, strength, spasticity, and sensation) between legs in children with hemiplegic cerebral palsy. An observational study measured both lower limbs of children with hemiplegic cerebral palsy over one session. Six sensory-motor impairments (balance, coordination, strength, spasticity, contracture, and proprioception) were measured. The between-leg differences were analyzed using the paired t-tests and presented as the mean differences (95% confidence interval (CI)). Twenty-four participants aged 10.3 years (standard deviation: 1.3) participated. The affected leg was less than the less-affected leg in terms of the strength of dorsiflexors (mean difference (MD) -2.8 Nm, 95% CI -4.2 to -1.4), plantarflexors (MD -2.6 Nm, 95% CI -4.1 to -1.0), knee extensors (MD -5.3 Nm, 95% CI -10.2 to -0.5) as well as range of ankle dorsiflexion (MD -8 deg, 95% CI -13 to -3), and balance (median difference -11.1, 95% CI -11.6 to -10.6). There was a trend toward a difference in terms of the strength of hip abductors (MD -2.6 Nm, 95% CI -5.3 to 0.1) and coordination (MD -0.20 taps/s, 95% CI -0.42 to 0.01). The legs were similar in terms of the strength of hip extensors (MD 0.3 Nm, 95% CI -4.7 to 5.3), proprioception (MD 1 deg, 95% CI 0 to 2), and spasticity (median difference 0, 95% CI 0 to 0). Examination of the difference in sensory-motor impairments between legs in children with hemiplegic cerebral palsy has given us some insights into the deficits in both legs. Not only was balance, strength, and coordination decreased compared with the less-affected leg but also the less-affected leg was markedly decreased compared with typically developing children. Therefore, an intervention aimed at increasing muscle strength and coordination in both legs might have a positive effect, particularly on more challenging physical activities. This may, in turn, lead to successful participation in mainstream sport and recreation.

PMID: 37929345

7. Accelerometer-measured physical activity, sedentary behavior, and sleep in children with cerebral palsy and their adherence to the 24-hour activity guidelines

No authors listed

Dev Med Child Neurol. 2023 Nov 11. doi: 10.1111/dmcn.15811. Online ahead of print.

No abstract available

PMID: 37950588

8. A Scoping Review of the Serious Game-Based Rehabilitation of People with Cerebral Palsy

Si Nae Ahn

Review Int J Environ Res Public Health. 2023 Nov 1;20(21):7006. doi: 10.3390/ijerph20217006.

In a serious context, individuals with Cerebral Palsy (CP) have limited opportunities to engage in social interaction experiences. Through a review, this study provides an explanation and improved evidence of the methods for rehabilitation in games used in serious contexts for people with CP. Articles published from 2010 to 2022 focusing on serious game-based rehabilitation for people with CP are extracted from MEDILINE, Academic Search Ultimate, CINAHL, and the Web of Science. The articles were assessed based on the McMaster critical review form. This study analyzes the frequencies of goal and assessment tools according to the components using the International Classification of Functioning, Disability and Health (ICF). The evidence of all the studies is presented according to the principles of Population, Intervention, Comparison, Outcome (PICO) to organize the evidence. A total of 19 articles were selected. Five articles involved Randomized Controlled Trials (RCTs), six articles involved non-randomized one-group designs, three articles involved single experimental study designs, and five articles were case report designs. In the selected articles, the average score on the McMaster critical review form was 11.8 points. In the game-based rehabilitation for CP, more articles reported goals and assessment tools focusing on body function than goals and assessment tools focusing on activity and participation, according to the ICF. These findings provide a record of past work and identify the evidence to support the application of game-based rehabilitation for people with CP.

PMID: 37947563

9. Does the Level of Focus in Serious Games in Immersive VR Correlate with the Quality of Movement? Preliminary Results from an Ongoing Study on Rehabilitation of Children with Cerebral Palsy

No authors listed

IEEE Int Conf Rehabil Robot. 2023 Sep:2023:1-6. doi: 10.1109/ICORR58425.2023.10304712.

Cerebral palsy (CP), one of the diseases that cause motor deficiency, is a childhood condition characterized by a motor disability (palsy) caused by a non-progressive static lesion in the brain (cerebral) [1], [2]. Rehabilitation of patients with CP is typically accomplished through exercises performed by a team of several specialists so that the patient can act independently or with as little reliance on third parties as possible. In CP, motor disorders are often present together with sensory, perceptual, cognitive, behavioural and communication disorders. Regarding motor disorders, the most frequent outcome is hemiplegia, although diplegic and paraplegic patients are also present [1].

PMID: 37941186

10. Parents' perceptions of a modified sports intervention for children with cerebral palsy

Ricardo Rodrigues De Sousa Junior, Deisiane Oliveira Souto, Fabiane Ribeiro Ferreira, Fabiana Caetano Martins Da Silva E Dutra, Ana Cristina Resende Camargos, Georgina L Clutterbuck, Hércules Ribeiro Leite; Sports Stars Brazil Study Group

Dev Med Child Neurol. 2023 Nov 6. doi: 10.1111/dmcn.15795. Online ahead of print.

Aim: To explore the perceptions of parents of children with cerebral palsy (CP) participating in a modified sports intervention, Sports Stars Brazil. Method: Parents of children with CP (n = 15, 6-12 years old, nine males, six females), classified in Gross Motor Function Classification System levels I and II, who participated in the Sports Stars Brazil intervention, were recruited for this descriptive qualitative study. Children received the 8-week (1 hour per week) modified sports intervention. After the intervention, parents participated in a focus group. The F-word (fitness, family, fun, functioning, friends, and future) interpretation of the International Classification of Functioning, Disability and Health was used to guide the discussion. Participants were invited to report their experience of participating in the intervention using the F-words for a childhood disability model. Transcripts were coded using content analysis. Results: Parents reported positive experiences of the Sports Stars Brazil intervention, including a decrease in sedentarism (fitness), improvement of motor skills (functioning), greater connection between child and family (family), better socialization (friends), increased participation in pleasurable activities (fun), and awareness of the importance of physical activity (future). All positive experiences were interconnected. Interpretation: This study presents the ability of the Sports Stars Brazil intervention to integrate all aspects of functioning and supports its role as an engaging and promising intervention option for ambulant children with CP.

PMID: 37929798

11. Cerebral palsy registers around the world: A survey

Shona Goldsmith, Hayley Smithers-Sheedy, Nihad Almasri, Guro L Andersen, Leanne Diviney, Ecaterina Bufteac Gincota, Kate Himmelmann, Israt Jahan, Emma Waight, Sarah McIntyre

Dev Med Child Neurol. 2023 Nov 9. doi: 10.1111/dmcn.15798. Online ahead of print.

Aim: To provide a description of cerebral palsy (CP) registers globally, identify which aim to report on CP epidemiology, and report similarities and differences across topics of importance for the sustainability and collaboration between registers. Method: Representatives of all known CP registers globally (n = 57) were invited to participate. The online survey included 68 questions across aims, methodologies, output/impact, and stakeholder involvement. Responses were analysed using descriptive statistics. Results: Forty-five registers participated, including three register networks. Twenty were newly established or under development, including 12 in low- and middle-income countries (LMICs). An epidemiological aim was reported by 91% of registers. Funding is received by 85% of registers, most often from not-for-profit organizations. CP definitions are comparable across registers. While the minimum data set of a register network is used by most registers, only 25% of identified items are collected by all three register networks. Ninety per cent of registers measure research activities/output, and 64% measure research impact. People with lived experience are involved in 62% of registers. Interpretation: There has been a recent surge in CP registers globally, particularly in LMICs, which will improve understanding of CP epidemiology. Ongoing efforts to address identified methodological differences are essential to validate comparison of results and support register collaboration.

PMID: 37946559

12. Effects of voluntary exercise on muscle structure and function in cerebral palsy

No authors listed

Dev Med Child Neurol. 2023 Nov 11. doi: 10.1111/dmcn.15809. Online ahead of print.

No abstract available

PMID: 37950587

13. Two Cases of Degenerative Cervical Spondylotic Myelopathy in Adults with Athetoid and Dystonic Cerebral Palsy

Hayato Kinoshita, Michio Hongo, Yuji Kasukawa, Daisuke Kudo, Ryota Kimura, Naohisa Miyakoshi

Am J Case Rep. 2023 Nov 11:24:e941158. doi: 10.12659/AJCR.941158.

BACKGROUND Patients with athetoid and dystonic cerebral palsy (ADCP) may develop degenerative changes in the cervical spine that can aggravate their neurological symptoms in adulthood. This report is of 2 cases of ADCP associated with degenerative cervical spondylotic myelopathy in a 39-year-old woman and a 52-year-old man, requiring different surgical treatments. CASE REPORT Case 1. The patient was a 39-year-old woman who had fallen down 7 years before surgery and had since been walking with a cane. Her gait disturbance had worsened in the 2 years prior to surgery, and numbness in her upper limbs appeared. In the year before surgery, spasticity and numbness in the lower limbs worsened, and fine motor impairment also appeared. Because of mild involuntary movements of the neck, cervical laminoplasty from C3 to C6 was performed, and her symptoms remained stable until the last follow-up 4.5 years after surgery. Case 2. The patient was a 52-year-old man who had fallen down 7 years before surgery, resulting in transient limb weakness. In the year before surgery, he had developed fine motor impairment. He subsequently developed gait disturbance and requested cervical surgery. Because of involuntary movements involving the neck and trunk, he underwent cervical posterior fusion from C2 to T1. Six months after surgery, the gait disturbance had improved. CONCLUSIONS This report describes 2 adults with a history of ADCP since birth and highlights that degenerative changes of the cervical spine can occur at a relatively early age in adulthood, requiring an individualized approach to management.

PMID: 37950431

14. Unequal Cerebral Magnetic Resonance Imaging Changes in Perinatal Hypoxic Ischemic Injury of Term Neonates

Shyam Sunder B Venkatakrishna, Mohamed Elsingergy, Fikadu Worede, Jelena Curic, Savvas Andronikou

J Comput Assist Tomogr. 2023 Nov-Dec;47(6):913-918. doi: 10.1097/RCT.0000000000001486. Epub 2023 Jul 10.

Background: Perinatal hypoxic ischemic injury (HII) has a higher prevalence in the developing world. One of the primary concepts for suggesting that an imaging pattern reflects a global insult to the brain is when the injury is noted to be bilateral and symmetric in distribution. In the context of HII in term neonates, this is either bilateral symmetric (a) peripheral/watershed

(WS) injury or (b) bilateral symmetric basal-ganglia-thalamus (BGT) pattern, often with the peri-Rolandic and hippocampal injury. Unilateral, asymmetric, or unequal distribution of injury may therefore be misdiagnosed as perinatal arterial ischemic stroke. Objectives: We aimed to determine the prevalence of unequal cerebral injury in HII, identify patterns, and determine their relationship with existing classification of HII. Materials and methods: Review of brain magnetic resonance imaging from a database of children with HII. Reports with any unequal pattern of injury were included and further classified as a unilateral, bilateral asymmetric, or symmetric but unequal degree pattern of HII. Results: A total of 1213 MRI scans in patients with a diagnosis of HII revealed 156 (13%) with unequal involvement of the hemispheres: unilateral in 2 of 1213 (0.2%) (involvement only in the WS), asymmetric in 48 of 1213 (4%) (WS in 6 [0.5%], BGT in 4 [0.3%], and combined BGT and WS in 38 [3.1%]), and bilateral symmetric but unequal degree in 106 of 1213 (8.7%) (WS in 20 [1.6%], BGT in 17 [1.4%], and combined BGT and WS in 69 [5.7%]). Conclusions: The majority of children with cerebral palsy due to HII demonstrate a characteristic bilateral symmetric pattern of injury. In our study, 13% demonstrated an unequal pattern. Differentiation from perinatal arterial ischemic stroke, which is mostly unilateral and distributed typically in the middle cerebral artery territory, should be possible and recognition of the typical BGT or WS magnetic resonance imaging patterns should add confidence to the diagnosis, in such scenarios.

PMID: 37948366

15. Outpatient encounters, continuity of care, and unplanned hospital care for children and young people with cerebral palsy

Simon P Paget, Sarah McIntyre, Francisco J Schneuer, Tanya Martin, Louise Sellars, Heather Burnett, Sophie Price, Natasha Nassar

Dev Med Child Neurol. 2023 Nov 10. doi: 10.1111/dmcn.15800. Online ahead of print.

Aim: To describe the relationships between outpatient encounters, continuity of care, and unplanned hospital care in children/young people with cerebral palsy (CP). Method: In this population-based data-linkage cohort study we included children/young people with CP identified in the New South Wales/Australian Capital Territory CP Register (birth years 1994-2018). We measured the frequency of outpatient encounters and unplanned hospital care, defined as presentations to emergency departments and/or urgent hospital admissions (2015-2020). Continuity of outpatient care was measured using the Usual Provider of Care Index (UPCI). Results: Of 3267 children/young people with CP, most (n = 2738, 83.8%, 57.6% male) had one or more outpatient encounters (123 463 total encounters, median six outpatient encounters per year during childhood). High UPCI was more common in children/young people with mild CP (Gross Motor Function Classification System levels I-III, with no epilepsy or no intellectual disability), residing in metropolitan and areas of least socioeconomic disadvantage. Low UPCI was associated with four or more emergency department presentations (adjusted odds ratio [aOR] 2.34; 95% confidence interval [CI] 1.71-3.19) and one or more urgent hospital admissions (aOR 2.02; 95% CI 1.57-2.61). Interpretation: Children/young people with CP require frequent outpatient services. Improving continuity of care, particularly for those residing in regional/remote areas, may decrease need for unplanned hospital care.

PMID: 37946594

16. Dihydropteridine Reductase Deficiency - A Rare and Potentially Treatable Cause Mimicking Cerebral Palsy

Marta Ribeiro, Mafalda Rebelo, Andreia Pereira, Diana Antunes, Ana Cristina Ferreira, Sandra Jacinto

Endocr Metab Immune Disord Drug Targets. 2023 Nov 8. doi: 10.2174/0118715303279209231026044120. Online ahead of print.

Introduction: Dihydropteridine reductase deficiency (DHPRD) is a rare genetic disorder of tetrahydrobiopterin (BH4) regeneration, a cofactor for several enzymes, including phenylalanine hydroxylase. Due to hyperphenylalaninemia (HPA), patients can be detected by the newborn metabolic screening, when available. The most common symptoms of DHPRD may mimic cerebral palsy: developmental/cognitive impairment, hypotonia, peripheral hypertonia, dystonia, feeding difficulties, epilepsy, and microcephaly. The long-term neurodevelopmental outcome is strongly influenced by the early initiation of effective treatment. Case report: A 2-year-old boy, born in Guinea, was evaluated in our center with the diagnosis of "cerebral palsy". He was born after a prolonged labor, and had feeding difficulties and severe developmental delay. Examination revealed microcephaly, axial hypotonia, and dyskinetic movements without hypertension. No seizures or oculogyric crisis were reported. Brain MRI showed slight brain atrophy and hyperintensity T2/FLAIR in basal ganglia. The diagnosis of cerebral palsy was questioned, and further investigation was carried out. HPA raised the possibility of BH4 deficiency, supported by increased biopterin in urine, decreased neurotransmitters in CSF, and low DHPR enzyme activity. A variant (128_130del (p.(Val43del)) in apparent homozygosity was later detected in the QPDR gene. At 4 years old, he started L-dopa/carbidopa, oxitriptan, and a phenylalanine-restrictive diet with moderate clinical improvement. Conclusion: When the diagnosis of "cerebral palsy" is questionable, other etiologies should be investigated, particularly disorders that have specific disease-modifying treatment. In our patient, the atypical constellation of neurological signs, brain MRI findings, and the nonexistence of newborn metabolic screening in the country of origin supported additional investigation. The presence of HPA-associated dystonia was crucial to the investigation and was later confirmed as DHPRD. Unfortunately, at this stage, the reversibility of

the neurological damage in response to treatment is doubtful.

PMID: 37946348

17. Heart rate responses, agreement and accuracy among persons with severe disabilities participating in the indirect movement program: Team Twin-an observational study

Andreas Jørgensen, Mette Toftager, Martin Eghøj, Mathias Ried-Larsen, Christina Bjørk Petersen

Front Sports Act Living. 2023 Oct 24:5:1213655. doi: 10.3389/fspor.2023.1213655. eCollection 2023.

Introduction: Heart rate (HR) monitors are rarely used by people living with disabilities (PLWD), and their accuracy is undocumented. Thus, this study aims to describe the HR response during the Team Twin co-running program and, secondly, to assess the agreement and accuracy of using HR monitors among PLWD. Methods: This 16-week single-arm observational study included 18 people with various disabilities. During the study, the subjects wore a Garmin Vivosmart 4 watch (wrist). To evaluate the agreement and accuracy we applied Garmin's HRM-DUAL™ chest-worn HR monitors for comparison with the Vivosmart 4. The HR response analysis was performed descriptively and with a mixed regression model. The HR agreement and accuracy procedure was conducted on a subsample of five subjects and analyzed using Lin's concordance analysis, Bland and Altman's limits of agreement, and Cohen's kappa analysis of intensity zone agreement. This study was prospectively registered at Clinical Trials.gov (NCT04536779). Results: The subjects had a mean age of 35 (±12.6), 61% were male, 72% had cerebral palsy were 85% had GMFCS V-IV. HR was monitored for 202:10:33 (HH:MM:SS), with a mean HR of 90 ± 17 bpm during training and race. A total of 19% of the time was spent in intensity zones between light and moderate (30%-59% HR reserve) and 1% in vigorous (60%-84% HR reserve). The remaining 80% were in the very light intensity zone (<29% HR reserve). HR was highest at the start of race and training and steadily decreased. Inter-rater agreement was high (k = 0.75), limits of agreement were between -16 and 13 bpm, and accuracy was acceptable (Rc = 0.86). Conclusion: Disability type, individual, and contextual factors will likely affect HR responses and the agreement and accuracy for PLWD. The Vivosmart 4, while overall accurate, had low precision due to high variability in the estimation. These findings implicate the methodical and practical difficulties of utilizing HR monitors to measure HR and thus physical activity in adapted sports activities for severely disabled individuals.

PMID: 37941848

18. Heterogeneity of Tasks and Outcome Measures in Dual Tasking Studies in Children with Cerebral Palsy: A Scoping Review

Meaghan Rubsam, Gay L Girolami, Tanvi Bhatt

Phys Ther. 2023 Nov 6:pzad151. doi: 10.1093/ptj/pzad151. Online ahead of print.

Objective: The aims of this scoping review are to examine the available literature regarding dual tasking in children with cerebral palsy (CP) and to identify and categorize both the motor and cognitive tasks and outcome measures used primarily through the International Classification of Functioning, Disability and Health model. Methods: Five electronic databases were searched. Studies were included if they: (1) were published in English; (2) included at least 1 group of children or adolescents with a diagnosis of CP; (3) assessed dual tasking as part of the study; (4) reported the method for performing the dual task; and (5) reported the outcome measures utilized. Results: Twenty-three studies with 439 children with CP were included. All studies utilized motor activities as the primary task, including walking, balance, and a functional transition. Motor secondary tasks occurred in 10 studies, cognitive secondary tasks in 12 studies, and 1 study used both. Forty-one outcome measures over 23 studies assessed the body structure and function domain, 7 measures over 6 studies assessed activity limitations, and 2 outcomes over 2 studies assessed participation. Conclusions: The 23 included studies demonstrated heterogeneity in the age and function of participants, secondary tasks, and outcome measures. Future studies on dual tasking in children with CP should consider the difficulty of the primary motor or cognitive task and compare secondary tasks to establish this contribution to motor performance. Studies should incorporate activity and participation measures to assess meaningful functional outcomes. Impact: Children with CP experience challenges when exposed to dual task situations. This scoping review highlights the importance of considering multiple factors when designing dual tasking studies involving children with CP to facilitate results translation, improved participation, and enhanced function. Similarly, studies should utilize activity and participation outcomes to assess quality of life.

PMID: 37941469

19. Spastic Diplegia and Visual Defects in CTNNB1 Gene Mutation: Genetic Mimic of Cerebral Palsy

Arushi Gahlot Saini, Pradeep Kumar Gunasekaran, Rahul Ranjan, Vikas Bhatia

Indian J Pediatr. 2023 Nov 8. doi: 10.1007/s12098-023-04923-z. Online ahead of print.

No abstract available

PMID: 37938513

20. Neurodevelopmental outcomes in preterm or low birth weight infants with germinal matrix-intraventricular hemorrhage: a meta-analysis

Meicen Zhou, Shaopu Wang, Ting Zhang, Surong Duan, Hua Wang

Pediatr Res. 2023 Nov 7. doi: 10.1038/s41390-023-02877-8. Online ahead of print.

Background: This meta-analysis aimed to identify the near- and long-term neurodevelopmental prognoses of preterm or low birth weight (LBW) infants with different severities of intraventricular hemorrhage (IVH). Methods: Four databases were searched for observational studies that were qualified using the Newcastle-Ottawa Scale. Results: 37 studies involving 32,370 children were included. Compared to children without IVH, children with mild IVH had higher incidences of neurodevelopmental impairment (NDI), cerebral palsy (CP), motor/cognitive delay, hearing impairment and visual impairment, as well as lower scores of the mental development index (MDI) and psychomotor development (PDI). Moreover, compared to mild IVH, severe IVH increased susceptibilities of children to NDI, motor delay, CP, hearing impairment and visual impairment, with worse performances in MDI, PDI, motor score and IQ. Mild IVH was not associated with seizures or epilepsy. Conclusions: Adverse neurodevelopmental outcomes positively associated with the occurrence and severity of IVH in preterm or LBW infants, providing evidence for counseling and further decisions regarding early therapeutic interventions. Impact: Adverse neurodevelopmental outcomes later in life were closely associated with the occurrence and severity of IVH in preterm or LBW infants. Our results highlight the importance to make prediction of the neurodevelopmental outcomes of children born preterm or LBW with a history of IVH, which will guide affected parents when their children need clinical interventions to reach the full potential. We emphasize the importance of identifying specific developmental delays that may exist in children with IVH, providing detailed information for the development of comprehensive intervention measures.

PMID: 37935882

21. When the Lung Refuses to Expand: A Deep Dive Into a Pneumothorax Ex Vacuo Case

Mena Louis, Lucas Canaan, John Hastings, Navneeth Bongu, Hardeep Singh

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Pneumothorax ex vacuo and trapped lung represent challenging clinical entities, especially in the context of pre-existing comorbidities. This case report outlines the diagnostic and management pathway of a 38-year-old patient with cerebral palsy who initially presented with empyema. Following the evacuation of the empyema, the patient developed pneumothorax ex vacuo, a rare phenomenon occurring due to a vacuum-like negative intrapleural pressure initiated by lung collapse. Initially suspected to have an infectious etiology based on laboratory findings, the patient was later found to have a large hydropneumothorax through a combination of imaging, laboratory studies, and clinical evaluations without confirming infection or malignancy. Despite interventions including Tissue Plasminogen Activator (TPA) and Deoxyribonuclease (DNAse) administration to facilitate pleural drainage, the patient's condition persisted, necessitating a surgical intervention that evolved from a minimally invasive video-assisted thoracoscopic surgery (VATS) to a more invasive thoracotomy due to unforeseen pleural thickening. The patient's pre-existing condition of cerebral palsy increased his susceptibility to respiratory complications, including empyema, due to the risk of aspiration. This case highlights the importance of a multidisciplinary approach in managing such complex clinical scenarios. It also serves as a clinical reminder that pneumothorax ex vacuo is generally benign and does not typically require chest tube placement, as the primary issue is an unexpandable lung that is unresponsive to pleural drainage. The report emphasizes the need for flexible surgical planning and robust postoperative management to optimize patient outcomes. It also clarifies the distinct pathophysiology of pneumothorax ex vacuo compared to primary or secondary pneumothorax, advocating for a comprehensive diagnostic approach and the crucial role of a multidisciplinary team in the management of such intricate cases.

PMID: 37933350

22. One-year review of disability certificates issued by the neurology unit of a medical college in Maharashtra, India

Makarand M Kanjalkar, Prabhanjan V Kulkarni

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In this article, we describe the profile of patients who were issued disability certificates from the neurology unit of a medical college in Maharashtra, India. In a retrospective cross-sectional study of 1-year duration, the demographic and clinical details of 102 participants were studied, of which 100 were issued the certificate. The mean age was 36 ± 14.04 years. There were

more number of male applicants (65%) than females. Motor disability (71%) was the most common manifestation for which disability certificate was sought. Cerebral palsy was the most common etiology in the age group of <40 years, while stroke was the most common etiology in the age group >40 years. During our study period, four patients' diagnoses were changed during revisit. Additional studies are required to assess the causes of low female enrollment and lack of motivation to apply for the disability certificate at an earlier age.

PMID: 37929398

23. Arginase deficiency masked by cerebral palsy and coagulopathy-Three varied presentations of Latin American origin

Shelby L Mills, Paige Roberts, Myla Ashfaq, Kathryn Leal, Hope Northrup, Deborah L Brown, David Rodriguez-Buritica, Laura S Farach

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Arginase deficiency (ARG1-D) is an autosomal recessive inborn error of metabolism that is often misdiagnosed. Classic presentation of ARG1-D includes progressive symptoms of spasticity, delayed development, cognitive impairment, protein avoidance, and seizures. Patients who present atypically may evade diagnosis and require a thoughtful diagnostic workup. Here, we discuss three females of Latin American origin with differing clinical presentations, but who all have the same intronic pathogenic variant in ARG1. Importantly, we found that each case included elevated coagulopathy on laboratory testing and discussed one case in particular with manifestation of bleeding. When diagnosed early, treatment is favorable and can prevent progressive decline. While many states have added ARG1-D to their expanded newborn screening panels, still many states and countries do not screen for ARG1-D, and it can be missed in a healthy newborn. We aim to bring awareness to not only the classic presentation as a necessary consideration for otherwise unexplained spastic diplegia but also to the varied presentations of ARG1-D.

PMID: 37927486

24. Associations between the aetiology of preterm birth and mortality and neurodevelopment up to 11 years

Linda Grönroos, Päivi Rautava, Sirkku Setänen, Anna Nyman, Eeva Ekholm, Liisa Lehtonen, Milla Ylijoki; PIPARI Study Group

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Aim: To investigate how the aetiology of very preterm birth/very low birth weight is associated with mortality and later neurodevelopmental outcomes. Methods: Very preterm/very low-birth weight singletons were categorised based on the aetiology of preterm birth: spontaneous preterm birth (n = 47, 28.1%), preterm premature rupture of membranes (n = 56, 33.5%) or placental vascular pathology (n = 64, 38.3%). Mortality, cerebral palsy, severe cognitive impairment by 11 years of age (<2SD) and mean full-scale intelligence quotient at 11 years were studied in association with birth aetiology. Results: There was no difference in mortality or rate of cerebral palsy according to birth aetiologies. The rate of severe cognitive impairment was lower (4.9% vs. 15.3%) in the preterm premature rupture of the membrane group in comparison to the placental vascular pathology group (OR 0.2, 95% CI 0.03-0.9, adjusted for gestational age). At 11 years, there was no statistically significant difference in the mean full-scale intelligence quotient. Conclusion: Placental vascular pathology, as the aetiology of very preterm birth/very low birth weight, is associated with a higher rate of severe cognitive impairments in comparison to preterm premature rupture of membranes, although there was no difference in the mean full-scale intelligence quotient at 11 years. The aetiology of very preterm birth/very low birth weight was not associated with mortality or the rate of cerebral palsy.

PMID: 37926858

25. Newly diagnosed Parkinson's disease in a middle-aged cerebral palsy patient with schizencephaly

Sangjee Lee, Yu Jin Jung

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No abstract available

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