Cerebral palsy research news

Monday 6 November 2023

Cerebral Palsy Alliance is delighted to bring you this free weekly bulletin of the latest published research into cerebral palsy. Our organisation is committed to supporting cerebral palsy research worldwide - through information, education, collaboration and funding. Find out more at cerebralpalsy.org.au/our-research

Professor Nadia Badawi AM
CP Alliance Chair of Cerebral Palsy Research

Subscribe to CP Research News

Interventions and Management

1. Efficacy of plyometric exercises on upper extremity function, selective motor control and hand grip strength in children with unilateral cerebral palsy: A randomized controlled study

Amira M Abd-Elmonem, Hazem A Ali, Sara S Saad-Eldien, Walaa A Abd El-Nabie

Physiother Res Int. 2023 Nov 3:e2061. doi: 10.1002/pri.2061. Online ahead of print.

Background: Impairment in unimanual upper limb function is frequent among children with unilateral cerebral palsy (UCP), which affects their ability to perform functional activities. Aim: To assess the efficacy of plyometric exercises on the function of upper extremity, selective motor control (SMC) and hand grip strength (HGS) in children with UCP. Design: This was a double-masked, randomized, controlled clinical trial. Setting: Outpatient Clinics of Faculty of Physical Therapy, Cairo University and Center for Physical Medicine, Rehabilitation and Rheumatology, Al-Agouza Hospital, Giza, Egypt. Population: Forty children with UCP, ranging in age from 8 to 12 years, were randomly allocated to two groups equal in numbers. Method: Children were allocated to receive conventional therapy (CONV-group; n = 20) or plyometric exercises (PLYO-group; n = 20) for 45 min. In addition, children of both groups received selected physical and occupational therapy programs (each lasted for 30 min) twice a week over 3-month. The intervention was delivered on non-consecutive days. Upper extremity function, SMC and HGS were assessed by using quality of upper extremity skills test (QUEST), Test of arm selective control and pneumatic squeeze bulb dynamometer, respectively. Results: Overall, 35 children (18 in the CONV-group, 17 in the PLYO-group) completed data collection and treatment. With-in group comparison showed significant improvement in the study groups while post-treatment comparisons revealed a significant difference from mean difference in upper extremity function is 9.55 (8.71:10.39), SMC is 2.12 (1.51:2.72) and HGS is 2.91 (2.13:3.68) ($p < 0.05$; 95% Confidence interval) in favor of the PLYO-group. Conclusions: Plyometric exercises have the capability to enhance upper extremity function and strength in children with UCP.

PMID: 37922449

2. Tendon transfers in cerebral palsy: art or science?

Michael Tonkin

Editorial J Hand Surg Eur Vol. 2023 Nov 2:17531934231210380. doi: 10.1177/17531934231210380. Online ahead of print.

Tendon transfers in cerebral palsy are commonly described as a method to improve function in the upper limb. The field can be considered by some to be more of an art than science due to the unique challenges involved, which include poor volitional control and spasticity. Surgical outcomes can be uncertain and, in certain circumstances, worsened. This presentation looks at the rationale behind tendon transfers for patients with cerebral palsy and the application of this rationale (science vs. art) to a philosophy of treatment.

PMID: 37917831

3. Effects of hip brace on coxa valga in nonambulatory children with cerebral palsy: A randomized controlled trial

Hyeonseong Woo, Bo Ryun Kim, Jin A Yoon, Hyun Jung Han, Young Il Yoon, Seung U Lee, Seon Cho, Yong Beom Shin, Hyun Jung Lee, Jee Hyun Suh, Jiwoon Lim, Jaewon Beom, Yulhyun Park, Ju Seok Ryu

Randomized Controlled Trial Medicine (Baltimore). 2023 Oct 27;102(43):e35696. doi: 10.1097/MD.0000000000035696.

Background: Coxa valga, measured as the neck-shaft angle (NSA) or head-shaft angle (HSA), is regarded as a potential risk factor for hip dislocation in patients with cerebral palsy. We investigated the effects of a novel hip brace on coxa valga. Methods: A prospective, multicenter, assessor-blinded, randomized controlled trial was conducted from July 2019 to November 2021. Children with cerebral palsy aged 1 to 10 years with Gross Motor Function Classification System levels IV and V were recruited. The study group wore a hip brace for at least 12 hour a day. A lower strap of the hip brace was designed to prevent coxa valga biomechanically. The effectiveness of the hip brace on coxa valga was assessed by measurement of the NSA and head-shaft angle at enrollment and 6 and 12 months after enrollment. Results: Sixty-six participants were enrolled, and 33 patients were assigned to each group. Changes in the mean NSA of both sides and the NSA of left side showed significant differences between the 2 groups over 12 months (mean NSA of both sides, -1.12 ± 3.64 in the study group and 1.43 ± 3.75 in the control group, P = .023; NSA of the left side, -1.72 ± 5.38 in the study group and 2.01 ± 5.22 in the control group, P = .008). Conclusions: The hip brace was effective in preventing the progression of coxa valga and hip displacement, suggesting that the prevention of coxa valga using hip brace is a contributing factor in prevention of hip displacement.

PMID: 37904376

4. Radiological assessment of hip disease in children with cerebral palsy: development of a core measurement set

Prince J S Joseph, Mohammed Khattak, Sundas T Masudi, Louise Minta, Daniel C Perry

Bone Jt Open. 2023 Nov 1;4(11):825-831. doi: 10.1302/2633-1462.411.BJO-2023-0060.R1.

Aims: Hip disease is common in children with cerebral palsy (CP) and can decrease quality of life and function. Surveillance programmes exist to improve outcomes by treating hip disease at an early stage using radiological surveillance. However, studies and surveillance programmes report different radiological outcomes, making it difficult to compare. We aimed to identify the most important radiological measurements and develop a core measurement set (CMS) for clinical practice, research, and surveillance programmes. Methods: A systematic review identified a list of measurements previously used in studies reporting radiological hip outcomes in children with CP. These measurements informed a two-round Delphi study, conducted among orthopaedic surgeons and specialist physiotherapists. Participants rated each measurement on a nine-point Likert scale ('not important' to 'critically important'). A consensus meeting was held to finalize the CMS. Results: Overall, 14 distinct measurements were identified in the systematic review, with Reimer's migration percentage being the most frequently reported. These measurements were presented over the two rounds of the Delphi process, along with two additional measurements that were suggested by participants. Ultimately, two measurements, Reimer's migration percentage and femoral head-shaft angle, were included in the CMS. Conclusion: This use of a minimum standardized set of measurements has the potential to encourage uniformity across hip surveillance programmes, and may streamline the development of tools, such as artificial intelligence systems to automate the analysis in surveillance programmes. This core set should be the minimum requirement in clinical studies, allowing clinicians to add to this as needed, which will facilitate comparisons to be drawn between studies and future meta-analyses.

PMID: 37909150

5. Abdominal Compartment Syndrome Secondary to Constipation in an Adult Patient With Cerebral Palsy

Jocelyn Zi Lin Ting, M Priya Dharshini, Mei Fang Chew

Case Reports Cureus. 2023 Oct 1;15(10):e46312. doi: 10.7759/cureus.46312. eCollection 2023 Oct.

The majority of patients with constipation can often be treated conservatively with laxatives, suppositories, or enemas in mild cases. However, endoscopic decompression or surgical intervention may be required in some instances. Abdominal compartment syndrome as a result of constipation is rarely seen in the literature. We report a case of faecal impaction, which led to abdominal compartment syndrome in an adult patient with cerebral palsy. With increasing life expectancy, such cases may be increasingly encountered in the adult population. Severe complications of constipation should not be overlooked, especially in this at-risk population. Early recognition of abdominal compartment syndrome is key in its management.

PMID: 37916246

6. Medium-term Results After Femoral Head Resection and Subtrochanteric Valgus Osteotomy in Children and Adolescents With Neuromuscular Disorders

Madeleine Marowsky, Oliver Jungesblut, André Strahl, Ralf Stücker, Martin Rupprecht

J Pediatr Orthop. 2023 Oct 26. doi: 10.1097/BPO.0000000000002556. Online ahead of print.

Background: Various salvage surgical procedures for painful hip dislocation in adolescent patients with cerebral palsy exist. To date, no significant differences among these surgical techniques have been described. In our institution the McHale procedure is the standard of care for painful chronically dislocated hips with or without deformity of the femoral head in patients with cerebral palsy. This study focuses on mid-term results after surgical treatment. Methods: Surgical reports and patient charts were analyzed retrospectively. All x-rays were evaluated and migration of the proximal femur and heterotopic ossification according to Brooker were recorded. In addition, we conducted a telephone interview with the caretakers with special reference to pain preoperatively and postoperatively and after implant removal, sitting tolerance, range of motion, mobility, quality of life, and personal hygiene. Results: Fifty-two patients (65 hips) with a mean age of 13.5 ± 3.6 years (range: 4 to -20 y) were included. Mean surgery time was 178.4 ± 63.4 minutes (range: 45 to 380 min) and mean follow-up was 45.17 ± 30.6 months (range: 12 to 204 mo). A significant difference between preoperative and postoperative pain levels was found, P<0.001. Personal hygiene (P=0.02) and quality of life (P=0.013) improved significantly. Eighty-five percent of the caregivers would have the surgery performed on their child again and 81% of the caregivers would recommend the surgery to others. The removal of implants leads to a significant improvement in pain (P=0.011). A total of 22 complications in 65 McHale procedures (33.9%) were related to the procedures. Conclusions: A significant reduction in pain and a significant improvement of hygiene as well as quality of life can be achieved with the McHale procedure in painful chronically dislocated hips in patients with cerebral palsy. Overall, the procedure is predominantly experienced as helpful by the caregivers and recommended to others. The removal of the implants improves pain significantly, but complications may occur in one third of the patients. Level of evidence: IV.

PMID: 37899529

7. Load modulation affects pediatric lower limb joint moments during a step-up task

Vatsala Goyal, Keith E Gordon, Theresa Sukal-Moulton

medRxiv. 2023 Oct 22:2023.10.20.23296774. doi: 10.1101/2023.10.20.23296774. Preprint

Performance in a single step has been suggested to be sensitive measure of movement quality in pediatric clinical populations. Although there is less information available in children with typical development, researchers have postulated the importance of analyzing the effect of body weight modulation on the initiation of stair ascent, especially during single limb stance where upright stability is most critical. The purpose of this study was to investigate the effect of load modulation from -20% to +15% of body weight on typical pediatric lower limb joint moments during a step-up task. Fourteen participants between 5-21 years with no known history of neurological or musculoskeletal concerns were recruited to perform multiple step-up trials. Peak extensor support and hip abduction moments were identified during the push-off and pull-up stance phases. Linear regressions were used to determine the relationship between peak moments and load. Mixed effects models were used to estimate the effect of load on hip, knee, and ankle percent contributions to peak support moments. There was a positive linear relationship between peak support moments and load in both stance phases, where these moments scaled with load. There was no relationship between peak hip abduction moments and load. While the ankle and knee were the primary contributors to the support moments, the hip contributed more than expected in the pull-up phase. Clinicians can use these results to contextualize movement differences in pediatric clinical populations including cerebral palsy and highlight potential target areas for rehabilitation for populations such as adolescent athletes.

PMID: 37905158

8. Identifying treatment non-responders based on pre-treatment gait characteristics - A machine learning approach

Rosa M S Visscher, Julia Murer, Fatemeh Fahimi, Elke Viehweger, William R Taylor, Reinald Brunner, Navrag B Singh

Heliyon. 2023 Oct 23;9(11):e21242. doi: 10.1016/j.heliyon.2023.e21242. eCollection 2023 Nov.

Background: Paediatric movement disorders such as cerebral palsy often negatively impact walking behaviour. Although clinical gait analysis is usually performed to guide therapy decisions, not all respond positively to their assigned treatment. Identifying these individuals based on their pre-treatment characteristics could guide clinicians towards more appropriate and personalized interventions. Using routinely collected pre-treatment gait and anthropometric features, we aimed to assess whether standard machine learning approaches can be effective in identifying patients at risk of negative treatment outcomes. Methods: Observational data of 119 patients with movement disorders were retrospectively extracted from a local clinical database, comprising sagittal joint angles and spatiotemporal parameters, derived from motion capture data pre- and post-treatment (physiotherapy, orthosis, botulin toxin injections, or surgery). Participants were labelled based on their change in gait

profile score (GPS, non-responders with a decline in GPS of <1.6° vs. responders). Their pre-treatment features (sagittal joint angles, spatiotemporal parameters, anthropometrics) were used to train a support vector machine classifier with 5-fold cross-validation and Bayesian optimization within a MATLAB-based Classification Learner App. Results: An average accuracy of 88.2 ± 0.5 % was achieved for identifying participants whose gait will not respond to treatment, with 64 % true negative rate and an area under the curve of 88 %. Conclusion: Overall, a classical machine learning model was able to identify patients at risk of not responding to treatment, based on gait features and anthropometrics collected prior to treatment. The output of such a model could function as a warning signal, notifying clinicians that a certain individual might not respond well to the standard of care and that a more personalized intervention might be needed.

PMID: 37908707

9. Human lower leg muscles grow asynchronously

Brian V Y Chow, Catherine Morgan, Caroline Rae, David I Warton, Iona Novak, Suzanne Davies, Ann Lancaster, Gordana C Popovic, Rodrigo R N Rizzo, Claudia Y Rizzo, Maria Kyriagis, Robert D Herbert, Bart Bolsterlee

J Anat. 2023 Nov 2. doi: 10.1111/joa.13967. Online ahead of print.

Muscle volume must increase substantially during childhood growth to generate the power required to propel the growing body. One unresolved but fundamental question about childhood muscle growth is whether muscles grow at equal rates; that is, if muscles grow in synchrony with each other. In this study, we used magnetic resonance imaging (MRI) and advances in artificial intelligence methods (deep learning) for medical image segmentation to investigate whether human lower leg muscles grow in synchrony. Muscle volumes were measured in 10 lower leg muscles in 208 typically developing children (eight infants aged less than 3 months and 200 children aged 5 to 15 years). We tested the hypothesis that human lower leg muscles grow synchronously by investigating whether the volume of individual lower leg muscles, expressed as a proportion of total lower leg muscle volume, remains constant with age. There were substantial age-related changes in the relative volume of most muscles in both boys and girls ($p < 0.001$). This was most evident between birth and five years of age but was still evident after five years. The medial gastrocnemius and soleus muscles, the largest muscles in infancy, grew faster than other muscles in the first five years. The findings demonstrate that muscles in the human lower leg grow asynchronously. This finding may assist early detection of atypical growth and allow targeted muscle-specific interventions to improve the quality of life, particularly for children with neuromotor conditions such as cerebral palsy.

PMID: 37917014

10. Analysis of Traditional Chinese Medicine Symptoms in Children with Spastic Cerebral Palsy, a Protocol for Data Mining

Xing Wang, Fang Pang, Xiao-Gang Du

J Multidiscip Healthc. 2023 Oct 25:16:3143-3149. doi: 10.2147/JMDH.S426969. eCollection 2023.

Background: Cerebral palsy (CP) is characterized by abnormal pronunciation, posture, and movement. Clinically, CP can be categorized into various motor syndromes, including spastic hemiplegia, diplegia, quadriplegia, involuntary movement, ataxia, and mixed types. Among these, spastic CP represents over 70-80% of all CP cases. The primary objective of our study is to identify the top and core Traditional Chinese Medicine (TCM) symptoms and analysis their association rules in children with spastic cerebral palsy, thereby enhancing the theoretical foundations of TCM treatment on spastic CP. Methods: The study will be conducted on children aged 4 to 14 years with spastic CP who are undergoing treatment at Xi'an Encephalopathy Hospital Affiliated to Shaanxi University of Chinese Medicine. Basic information about the patients and their TCM symptoms will be collected on the first day of admission. This information will include age, gender, birth history, family history, disease classification, and TCM symptoms (including symptoms, tongue, and pulse). Once the data is collected, it will be exported from the electronic medical record system for further analysis. Descriptive statistics will be performed using Excel 2019, while exploratory factor analysis and cluster analysis will be conducted using SPSS Statistics 22. Additionally, association rule analysis will be carried out using SPSS Modeler 18. Results: This study will investigate the most top TCM symptoms in children with spastic CP and explore the association rules between these symptoms, mapping the presentation of spastic CP onto symptoms identified within TCM. Conclusion: Our findings will provide the distinctive characteristics of TCM symptoms in children with spastic CP, furnishing evidence-based support to clinicians and patients in making well-informed decisions collaboratively.

PMID: 37905184

11. Evaluation of online adaptive yoga for psychological well-being in adults with disabilities

Megan J Sundstrom, Alexa L Asplund, Viann N Nguyen-Feng

Rehabil Psychol. 2023 Nov 2. doi: 10.1037/rep0000520. Online ahead of print.

Purpose/objective: The present study examined the preliminary effectiveness of an online, community-based adaptive yoga program on mindfulness, social connectedness, life satisfaction, and communicative participation. Research method/design: Adults (N = 48) were from a U.S. Midwest rehabilitation program and modally identified as White (73%), women (68%), and in their 30s (24%). One-third (35%) of participants were able to walk independently, and cerebral palsy was the most common primary disability diagnosis (32%). The 90-min adaptive yoga program and assessments were offered online weekly for 6 weeks, in which data from 29 participants across three sessions were included in the present analyses. Results: Regarding our primary outcome of interest, there was a statistically significant positive increase in mindfulness over time, $F(1, 28) = 5.66$, $p = .02$, with a strong effect size, $d = 2.43$. All secondary variables had statistically nonsignificant changes over time, although with large effect sizes: social connectedness ($d = 0.77$), life satisfaction ($d = 0.92$), and communicative participation ($d = 0.40$). Conclusions/implications: The present study provides support for well-being programs for an underserved group; specifically, an online adaptive yoga program with routine outcome monitoring assessments has preliminary effectiveness in increasing mindfulness. Consideration of such well-being programs alongside insurance would be an important policy consideration. Further controlled research is necessary to draw decisive conclusions. (PsycInfo Database Record (c) 2023 APA, all rights reserved).

PMID: 37917463

12. Cerebral Palsy - Early Diagnosis and Intervention Trial: protocol for the prospective multicentre CP-EDIT study with focus on diagnosis, prognostic factors, and intervention

Christina Engel Hoei-Hansen, Lene Weber, Mette Johansen, Rebecca Fabricius, Jonas Kjeldbjerg Hansen, Anne-Cathrine F Viuff, Gitte Rønde, Gitte Holst Hahn, Elsebet Østergaard, Morten Duno, Vibeke Andrée Larsen, Camilla Gøbel Madsen, Katrine Røhder, Ann-Kristin Gunnes Elvrum, Britt Laugesen, Melanie Ganz, Kathrine Skak Madsen, Maria Willerslev-Olsen, Nanette Mol Debes, Jan Christensen, Robin Christensen, Gija Rackauskaite

BMC Pediatr. 2023 Oct 30;23(1):544. doi: 10.1186/s12887-023-04312-7.

Background: Early diagnosis of cerebral palsy (CP) is important to enable intervention at a time when neuroplasticity is at its highest. Current mean age at diagnosis is 13 months in Denmark. Recent research has documented that an early-diagnosis set-up can lower diagnostic age in high-risk infants. The aim of the current study is to lower diagnostic age of CP regardless of neonatal risk factors. Additionally, we want to investigate if an early intervention program added to standard care is superior to standard care alone. Methods: The current multicentre study CP-EDIT (Early Diagnosis and Intervention Trial) with the GO-PLAY intervention included (Goal Oriented ParentaL supported home ActivitY program), aims at testing the feasibility of an early diagnosis set-up and the GO-PLAY early intervention. CP-EDIT is a prospective cohort study, consecutively assessing approximately 500 infants at risk of CP. We will systematically collect data at inclusion (age 3-11 months) and follow a subset of participants (n = 300) with CP or at high risk of CP until the age of two years. The GO-PLAY early intervention will be tested in 80 infants with CP or high risk of CP. Focus is on eight areas related to implementation and perspectives of the families: early cerebral magnetic resonance imaging (MRI), early genetic testing, implementation of the General Movements Assessment method, analysis of the GO-PLAY early intervention, parental perspective of early intervention and early diagnosis, early prediction of CP, and comparative analysis of the Hand Assessment for Infants, Hammersmith Infant Neurological Examination, MRI, and the General Movements method. Discussion: Early screening for CP is increasingly possible and an interim diagnosis of "high risk of CP" is recommended but not currently used in clinical care in Denmark. Additionally, there is a need to accelerate identification in mild or ambiguous cases to facilitate appropriate therapy early. Most studies on early diagnosis focus on identifying CP in infants below five months corrected age. Little is known about early diagnosis in the 50% of all CP cases that are discernible later in infancy. The current study aims at improving care of patients with CP even before they have an established diagnosis.

PMID: 37899466

13. Comparative Efficacy and Side-effect Profiles of Interventions for Pediatric Saliva Control: A Cohort Study

Apostolos Papandreou, Aoife Mahony, Anne Breaks, Michael Absoud, Charlie Fairhurst

J Pediatr. 2023 Oct 26:113803. doi: 10.1016/j.jpeds.2023.113803. Online ahead of print.

Objective: To provide comparative efficacy and side-effect profile data on conservative, behavioral, pharmacologic, and surgical treatments used for pediatric saliva control. Study design: A cohort study of children (n=483) referred to a specialty Saliva Control service between May 2014 and November 2019 was performed, using quantitative data from pre- and post-treatment questionnaires [the Drooling Impact Scale (DIS), Drooling Rating Scale (DRS)] and recording of side effects. Overall, 483 children were included; treatment choices were based on published international guidelines. Results: The largest improvement was seen post intra-glandular botulinum toxin A (BTX-A) injections (n=207; 551 courses, mean DIS change 34.7, 95% CI=29.2-35.7) or duct transpositional surgery (n=31, mean change in DIS= 29.0, CI=22.3-35.7). Oral anticholinergics were associated with good outcomes, with no significant statistical difference between glycopyrronium

bromide (n=150, mean DIS change 21.5, CI=19.1-24.0) or trihexyphenidyl (n=87, mean DIS change 22.4, CI=18.9-25.8). Inhaled ipratropium bromide was not as efficacious (n=80, mean DIS change 11.1, CI=8.9-13.3). Oromotor programs were used in a selected group with reliable outcomes (n=9, mean DIS change 13.0). Side effects were consistent with previous studies. Overall, in cases of milder severity, enterally administered therapies provided a good first-line option. With more severe problems, BTX-A injections or saliva duct transpositional surgery were more effective and well tolerated. Conclusion: We describe a large single-center pediatric saliva control cohort, providing direct comparison of efficacy and side-effect profiles for all available interventions, and inform clinical practice for specialists when considering different options. BTX-A injections or saliva duct transpositional surgery appear to be more effective for saliva control that is more severe.

PMID: 37898423

14. Recommendations for promoting user agency in the design of speech neuroprostheses

Narayan Sankaran, David Moses, Winston Chiong, Edward F Chang

Front Hum Neurosci. 2023 Oct 18:17:1298129. doi: 10.3389/fnhum.2023.1298129. eCollection 2023.

Brain-computer interfaces (BCI) that directly decode speech from brain activity aim to restore communication in people with paralysis who cannot speak. Despite recent advances, neural inference of speech remains imperfect, limiting the ability for speech BCIs to enable experiences such as fluent conversation that promote agency - that is, the ability for users to author and transmit messages enacting their intentions. Here, we make recommendations for promoting agency based on existing and emerging strategies in neural engineering. The focus is on achieving fast, accurate, and reliable performance while ensuring volitional control over when a decoder is engaged, what exactly is decoded, and how messages are expressed. Additionally, alongside neuroscientific progress within controlled experimental settings, we argue that a parallel line of research must consider how to translate experimental successes into real-world environments. While such research will ultimately require input from prospective users, here we identify and describe design choices inspired by human-factors work conducted in existing fields of assistive technology, which address practical issues likely to emerge in future real-world speech BCI applications.

PMID: 37920562

15. Combined surveillance and treatment register for children with cerebral palsy: the protocol of the Netherlands CP register

Aukje Andringa, Kirsten Veerkamp, Marij Roebroeck, Marjolijn Ketelaar, Martijn Klem, Hurnet Dekkers, Jeanine Voorman, Marieke van Driel, Annemieke Buizer

BMJ Open. 2023 Oct 28;13(10):e076619. doi: 10.1136/bmjopen-2023-076619.

Introduction: Cerebral palsy (CP) is a childhood onset, lifelong, condition. Early detection and timely treatment of potential problems during the child's development are important to prevent secondary impairments and improve function. Clinical management of children with CP requires a spectrum of multidisciplinary interventions, which have an impact on short-term and long-term outcomes. However, there is a lack of knowledge about a personalised approach in this heterogeneous population. Various CP registers with different aims have been developed worldwide, which has made an important contribution to our understanding of CP. The purpose of this protocol is to describe the unique design of a combined multidisciplinary surveillance and treatment register for children with CP in the Netherlands, which aims to improve quality of care and to enhance an individual treatment approach. Methods and analysis: The Netherlands CP Register combines a multidisciplinary surveillance programme with a standardised protocol for treatment registry. The register systematically collects real-life surveillance and treatment data of children with CP. The register contributes to daily care at the individual level by screening for potential secondary impairments using a decision-support tool, by visualising individual development using a dashboard, and by supporting goal setting and shared decision-making for interventions. The register provides a platform at the national level for quality of care improvement and a comprehensive database of real-life data allowing multicentre studies with a long-term follow-up. People with lived experience of CP, healthcare professionals from different disciplines and researchers collaborated in the development of the register. Ethics and dissemination: The Netherlands CP register was submitted to the Medical Ethics Review Committee of VU University Medical Center (Amsterdam, the Netherlands), who judged the register not to be subject to the Medical Research Involving Human Subjects Act. A scientific board reviews requests for dissemination of data from the register for specific research questions.

PMID: 37898490

16. Mapping health services for adults with cerebral palsy in Ireland: a pilot study

Manjula Manikandan, Shalini Jagdeo, Fiona Weldon, Sarah Harrington, Rory O'Sullivan, Jennifer Fortune, Claire Kerr, Jennifer M Ryan

HRB Open Res. 2022 Dec 15:5:61. doi: 10.12688/hrbopenres.13609.2. eCollection 2022.

Background: Cerebral palsy (CP) is a common cause of physical disability in childhood. The majority of children with CP survive to adulthood. Once discharged from children's services, adults with CP find it challenging to navigate health services. The aim of this study was to pilot and refine a methodology to map services for adults with CP in Ireland. Methods: We used a multi-informant mapping methodology consisting of: 1. Defining health services; 2. Identifying informants; 3. Designing a survey; 4. Collecting data; 5. Data checking and analysis. We collected data on services from service users and service providers using an online survey. We verified data against information available online and by asking organisations to provide details about the service. Results: Fifteen service users and nine service providers completed the online survey. Data on 265 unique services at 32 organisations were provided. The most commonly provided services were physiotherapy (12%) and occupational therapy (11%). We confirmed the name of 89 services (34%) against online information. We received further details from eight organisations about 27 services. Specifically, we received details about the organisation name for 27 of the 265 services (10%), service name for 25 services (9%), service type for 25 services (9%), a website for 19 services (7%), and data on eligibility criteria and types of supports provided for between 25 or 26 services (9% or 10%). Conclusion: This pilot study highlighted the complexity of mapping services for adults with CP in Ireland. We recommend that an alternative methodology should be used to map services for adults with CP in Ireland.

PMID: 37901656

17. Regional anesthesia and analgesia in patients with spastic cerebral palsy undergoing orthopedic surgery: a historical cohort study

Vrushali C Ponde, Vincent Chan, Neha Singh, Ashok N Johari, Jolene Lee, Anuya Gursale, Dilip Chavan

Can J Anaesth. 2023 Nov 2. doi: 10.1007/s12630-023-02617-w. Online ahead of print.

Purpose: We sought to examine the incidence of severe postoperative pain in patients with cerebral palsy (CP) in the first 48 hr after surgery performed under combined regional and general anesthesia and its association with patient and surgical factors. Methods: In a historical cohort study, we reviewed the electronic records of 452 patients with spastic CP who underwent orthopedic surgeries of the upper and lower extremities from April 2016 to February 2020. Collected data included patient characteristics, American Society of Anesthesiologists Physical Status, details of anesthesia and surgery, types of regional anesthesia applied, success rate of anesthesia, incidence of severe pain, and adverse events. Results: We analyzed data from 440 patients; 404 patients underwent lower extremity surgery, 20 upper extremity surgery, and 15 both, and one patient required stem cell injection. All patients received general anesthesia before block performance. Single-injection neuraxial anesthesia was performed in 241 (54.8%) patients, brachial plexus block in 27 (6.1%) patients, and femoral/sciatic nerve blocks in 17 (3.9%) patients. Continuous neuraxial, brachial plexus, and femoral/sciatic nerve blocks were performed in 149 (33.9%), four (0.9%), and seven (1.6%) of the patients, respectively. Major and complex major surgeries were performed in 161 (36.6%) and 72 (16.4%) patients, respectively and continuous catheters were inserted in 50.3% of patients undergoing major surgery and in 91.7% of patients undergoing complex major surgery. Severe pain was reported by the caregivers of 68 (15.5%) patients who received nonopioid analgesic interventions. Conclusion: Despite the use of regional anesthesia, approximately 15% of patients with spastic CP undergoing orthopedic surgery for spastic cerebral palsy experienced severe pain that responded to treatment adjustments.

PMID: 37919631

18. Prechtl's General Movements Assessment at writhing age guides MRI use in clinical implementation network

Allison J Chirigos, Betsy Ostrander, Vera Joanna Burton, Marissa Mirecki, Nathalie L Maitre; Cerebral Palsy Foundation Early Detection and Intervention Network

Pediatr Res. 2023 Nov 2. doi: 10.1038/s41390-023-02856-z. Online ahead of print.

No abstract available

PMID: 37919380

19. Polyhandicap, profound intellectual multiple disabilities : Concept and definition of a highly specific public health issue [Article in French]

Marie-Christine Rousseau, Myriam Winance, Karine Baumstarck

Rev Epidemiol Sante Publique. 2023 Oct 31;71(6):102184. doi: 10.1016/j.respe.2023.102184. Online ahead of print.

Objectives: The concept of polyhandicap first emerged in the late '60s in France, with actually a consensus on its definition.

This consensus has yet to be reached internationally. The absence of an international consensus on a definition and name for persons with polyhandicap limits progress in research and health planning for these people. Methods: This article describes the history of the emergence of the concept of polyhandicap in France and internationally. Results: The emergence of the concept and definition of polyhandicap is part of the history of the development of special education and care for children with disabilities started at the end of the 19th century and during the first half of the 20th century. In France, between 1970 and 2002, working groups composed of professionals and family associations gradually developed and refined the definition of polyhandicap, differentiating it from other clinical entities such as cerebral palsy. Internationally, the term polyhandicap is used in 4 European countries: in France where it first appeared, in Italy, in French-speaking Belgium, and in French-speaking Switzerland but also outside the EU. Various terms may be used around the world to describe clinical entities similar to polyhandicap; the most frequently used in the literature is the term Profound Intellectual and Multiple Disabilities (PIMD) or PIMD Spectrum which does not systematically refer to an early brain injury. Discussion: We are currently in the process of internationalizing the concept and definition of polyhandicap, and hopefully, as was the case for cerebral palsy in the 2000s, the various research teams working on this subject around the world will create collaborations and research networks targeting this specific population. Conclusion: A consensus around a precise definition of polyhandicap is important to ensure that these people are recognized for their uniqueness and specific qualities and to provide them adapted care.

PMID: 37918043

20. Maternal and Neonatal Outcomes at Periviable Gestation throughout Delivery Admission

Angela R Seasely, Victoria C Jauk, Jeff M Szychowski, Namasivayam Ambalavanan, Alan T Tita, Brian M Casey

Am J Perinatol. 2023 Nov 1. doi: 10.1055/s-0043-1776347. Online ahead of print.

Objective: The threshold of viability, as well as cutoffs for delivery interventions and neonatal resuscitation, vary by hospital and involve complex counseling. With improvements in neonatal resuscitation and intensive care, the threshold of viability has been decreasing. Decisions regarding delivery planning and neonatal resuscitation efforts should be based on the best available evidence. Our objective was to characterize survival rates and neonatal outcomes following periviable birth at different milestones beginning with prenatal admission through 1 year of life in a contemporary cohort. Study design: We performed a retrospective cohort study of all inborn infants without major congenital anomalies who delivered at the University of Alabama at Birmingham from 2013 to 2019 at gestational ages 22+0/7 to 25+6/7. Our primary outcome was to compared survival milestones throughout the pre- and postdelivery periods and neonatal complications in surviving newborns through 1 year of life at each gestational age. Results: The survival rate to 1 year of life was 49% (48-56%, 95% confidence interval [CI]) for the entire cohort and varied according to gestational age at delivery (22 weeks 15% [10-23%, 95% CI], 23 weeks 48% [43-58%, 95% CI], 24 weeks 57% [52-67%, 95% CI], 25 weeks 71% [67-82%, 95% CI]). Overall for the entire cohort, the rate of lung disease requiring respiratory support at discharge was 51%, intraventricular hemorrhage was 42%, retinopathy of prematurity was 74%, pulmonary hypertension was 30%, and concerns for cerebral palsy at 1 year of life was 25%. All outcomes improved with advancing gestational age at delivery. Of infants who delivered during the 22nd week of gestation, 50% received antenatal corticosteroids. Infants exposed to antenatal corticosteroids had more interventions, less pulmonary hypertension, and improved survival to 1 year of life. Conclusion: Knowledge of maternal complications, longitudinal survival rates, and neonatal outcomes of periviable deliveries according to gestational age throughout the admission enhances obstetric and perinatal counseling after hospital admission.

PMID: 37913782

21. [Recent research on the long-term neurodevelopmental outcomes of very preterm infants] [Article in Chinese] [Abstract in English, Chinese]

Ye Feng

Review Zhongguo Dang Dai Er Ke Za Zhi. 2023 Oct 15;25(10):1066-1071. doi: 10.7499/j.issn.1008-8830.2305072.

With the increase in the survival rate of very preterm infants, the long-term neurodevelopmental outcomes of such infants have attracted more and more attention. Very preterm infants tend to develop movement disorders and psychological and behavioral problems, including cerebral palsy, developmental coordination disorders, autism spectrum disorders, attention deficit hyperactivity disorders, specific learning disorders, and intellectual developmental disorders. It is of vital importance to improve the long-term prognosis of very preterm infants, and early comprehensive intervention measures can minimize disability and achieve optimal parenting outcomes. This article provides a review of the research progress on the long-term neurodevelopmental outcomes in extremely preterm infants.

PMID: 37905765

22. Case report: Suspecting guanine nucleotide-binding protein beta 1 mutation in dyskinetic cerebral palsy is important

Han-Byeol Choi, Yoonju Na, Jiwon Lee, Jeehun Lee, Ja-Hyun Jang, Jong-Won Kim, Jeong-Yi Kwon

Case Reports Front Pediatr. 2023 Oct 13:11:1204360. doi: 10.3389/fped.2023.1204360. eCollection 2023.

Herein, we describe the case of a 43-month-old girl who presented with clinical manifestations of dyskinetic cerebral palsy (CP), classified as the Gross Motor Function Classification System (GMFCS) V. The patient had no family history of neurological or perinatal disorders. Despite early rehabilitation, serial assessments using the Gross Motor Function Measure (GMFM) showed no significant improvements in gross motor function. Brain magnetic resonance imaging showed nonspecific findings that could not account for developmental delay or dystonia. Whole-genome sequencing identified a heterozygous NM_002074.5(GNB1):c.239T>C (p.Ile80Thr) mutation in guanine nucleotide-binding protein beta 1 (GNB1) gene. Considering this case and previous studies, genetic testing for the etiology of dyskinetic CP is recommended for children without relevant or with nonspecific brain lesions.

PMID: 37900673

23. Lessons learned from a pediatric powered mobility lending program

Naomi Gefen, Patrice L Weiss, Amihai Rigbi, Lori Rosenberg

Disabil Rehabil Assist Technol. 2023 Oct 28:1-10. doi: 10.1080/17483107.2023.2276232. Online ahead of print.

Purpose: To evaluate children's characteristics and impact of a powered wheelchair lending program including comparisons of diagnostic sub-groups, and validation of a predictive model of powered mobility proficiency. Methods and materials: This retrospective study included 172 children who participated in the ALYN powered mobility lending program from 3/2009-7/2022. Demographics and functional levels were measured via questionnaires; driving proficiency was evaluated when the wheelchair was returned, and parents and children were interviewed following their participation in the program. Results: Two diagnostic groups were identified: cerebral palsy (CP) (n = 136, median = 9.75 yrs) and other neuromuscular diseases (NMD) (n = 30, median = 5.83 yrs). They differed significantly in the age they commenced PM training, the male/female ratio, walking ability and access mode. Fifty-seven percent of the participants with CP achieved powered mobility proficiency, a rate that was significantly lower than the 73% proficiency found for the NMD group. Four significant predictors were identified: communication, manual wheelchair operation, access mode and go-stop upon request. They predicted proficiency in approximately 80% of cases. Overall feedback from the parents and children indicated that their personal and family's quality of life improved as a result of their child's ability to use a powered wheelchair. Conclusions: A lending program provides children with opportunities to improve mobility skills in an appropriate powered wheelchair. Children who can communicate verbally, propel a manual wheelchair, use a joystick and go-stop upon request are significantly more likely to become proficient drivers; however, many who were unable to complete these tasks also improved and even became proficient drivers.

PMID: 37897432

Prevention and Cure

24. Methylxanthine for the prevention and treatment of apnea in preterm infants

Keri A Marques, Matteo Bruschettini, Charles C Roehr, Peter G Davis, Michelle Fiander, Roger Soll

Review Cochrane Database Syst Rev. 2023 Oct 31;10(10):CD013830. doi: 10.1002/14651858.CD013830.pub2.

Background: Very preterm infants often require respiratory support and are therefore exposed to an increased risk of chronic lung disease and later neurodevelopmental disability. Although methylxanthines are widely used to prevent and treat apnea associated with prematurity and to facilitate extubation, there is uncertainty about the benefits and harms of different types of methylxanthines. Objectives: To assess the effects of methylxanthines on the incidence of apnea, death, neurodevelopmental disability, and other longer-term outcomes in preterm infants (1) at risk for or with apnea, or (2) undergoing extubation. Search methods: We searched CENTRAL, MEDLINE, Embase, two other databases, and three trial registers (November 2022). Selection criteria: We included randomized trials in preterm infants, in which methylxanthines (aminophylline, caffeine, or theophylline) were compared to placebo or no treatment for any indication (i.e. prevention of apnea, treatment of apnea, or prevention of re-intubation). Data collection and analysis: We used standard Cochrane methods and GRADE to assess the certainty of evidence. Main results: We included 18 studies (2705 infants), evaluating the use of methylxanthine in preterm infants for: any indication (one study); prevention of apnea (six studies); treatment of apnea (five studies); and to prevent re-intubation (six studies). Death or major neurodevelopmental disability (DMND) at 18 to 24 months. Only the Caffeine for

Apnea of Prematurity (CAP) study (enrolling 2006 infants) reported on this outcome. Overall, caffeine probably reduced the risk of DMND in preterm infants treated with caffeine for any indication (risk ratio (RR) 0.87, 95% confidence interval (CI) 0.78 to 0.97; risk difference (RD) -0.06, 95% CI -0.10 to -0.02; number needed to treat for an additional beneficial outcome (NNTB) 16, 95% CI 10 to 50; 1 study, 1869 infants; moderate-certainty evidence). No other trials reported DMND. Results from the CAP trial regarding DMND at 18 to 24 months are less precise when analyzed based on treatment indication. Caffeine probably results in little or no difference in DMND in infants treated for prevention of apnea (RR 1.00, 95% CI 0.80 to 1.24; RD -0.00, 95% CI -0.10 to 0.09; 1 study, 423 infants; moderate-certainty evidence) and probably results in a slight reduction in DMND in infants treated for apnea of prematurity (RR 0.85, 95% CI 0.71 to 1.01; RD -0.06, 95% CI -0.13 to 0.00; NNTB 16, 95% CI 7 to > 1000; 1 study, 767 infants; moderate-certainty evidence) or to prevent re-intubation (RR 0.85, 95% CI 0.73 to 0.99; RD -0.08, 95% CI -0.15 to -0.00; NNTB 12, 95% CI 6 to >1000; 1 study, 676 infants; moderate-certainty evidence). Death. In the overall analysis of any methylxanthine treatment for any indication, methylxanthine used for any indication probably results in little or no difference in death at hospital discharge (RR 0.99, 95% CI 0.71 to 1.37; I2 = 0%; RD -0.00, 95% CI -0.02 to 0.02; I2 = 5%; 7 studies, 2289 infants; moderate-certainty evidence). Major neurodevelopmental disability at 18 to 24 months. In the CAP trial, caffeine probably reduced the risk of major neurodevelopmental disability at 18 to 24 months (RR 0.85, 95% CI 0.76 to 0.96; RD -0.06, 95% CI -0.10 to -0.02; NNTB 16, 95% CI 10 to 50; 1 study, 1869 infants; moderate-certainty evidence), including a reduction in the risk of cerebral palsy or gross motor disability (RR 0.60, 95% CI 0.41 to 0.88; RD -0.03, 95% CI -0.05 to -0.01; NNTB 33, 95% CI 20 to 100; 1 study, 1810 infants; moderate-certainty evidence) and a marginal reduction in the risk of developmental delay (RR 0.88, 95% CI 0.78 to 1.00; RD -0.05, 95% CI -0.09 to -0.00; NNTB 20, 95% CI 11 to > 1000; 1 study, 1725 infants; moderate-certainty evidence). Any apneic episodes, failed apnea reduction after two to seven days (< 50% reduction in apnea) (for infants treated with apnea), and need for positive-pressure ventilation after institution of treatment. Methylxanthine used for any indication probably reduces the occurrence of any apneic episodes (RR 0.31, 95% CI 0.18 to 0.52; I2 = 47%; RD -0.38, 95% CI -0.51 to -0.25; I2 = 49%; NNTB 3, 95% CI 2 to 4; 4 studies, 167 infants; moderate-certainty evidence), failed apnea reduction after two to seven days (RR 0.48, 95% CI 0.33 to 0.70; I2 = 0%; RD -0.31, 95% CI -0.44 to -0.17; I2 = 53%; NNTB 3, 95% CI 2 to 6; 4 studies, 174 infants; moderate-certainty evidence), and may reduce receipt of positive-pressure ventilation after institution of treatment (RR 0.61, 95% CI 0.39 to 0.96; I2 = 0%; RD -0.06, 95% CI -0.11 to -0.01; I2 = 49%; NNTB 16, 95% CI 9 to 100; 9 studies, 373 infants; low-certainty evidence). Chronic lung disease. Methylxanthine used for any indication reduces chronic lung disease (defined as the use of supplemental oxygen at 36 weeks' postmenstrual age) (RR 0.77, 95% CI 0.69 to 0.85; I2 = 0%; RD -0.10, 95% CI -0.14 to -0.06; I2 = 18%; NNTB 10, 95% CI 7 to 16; 4 studies, 2142 infants; high-certainty evidence). Failure to extubate or the need for re-intubation within one week after initiation of therapy. Methylxanthine used for the prevention of re-intubation probably results in a large reduction in failed extubation compared with no treatment (RR 0.48, 95% CI 0.32 to 0.71; I2 = 0%; RD -0.27, 95% CI -0.39 to -0.15; I2 = 69%; NNTB 4, 95% CI 2 to 6; 6 studies, 197 infants; moderate-certainty evidence). Authors' conclusions: Caffeine probably reduces the risk of death, major neurodevelopmental disability at 18 to 24 months, and the composite outcome DMND at 18 to 24 months. Administration of any methylxanthine to preterm infants for any indication probably leads to a reduction in the risk of any apneic episodes, failed apnea reduction after two to seven days, cerebral palsy, developmental delay, and may reduce receipt of positive-pressure ventilation after institution of treatment. Methylxanthine used for any indication reduces chronic lung disease (defined as the use of supplemental oxygen at 36 weeks' postmenstrual age).

PMID: 37905735