Cerebral palsy research news

Monday 13 May 2024

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Interventions and Management

1. Effect of Mirror Therapy on Upper Limb Function in Children and Adolescents with Hemiplegic Cerebral Palsy: A Systematic Review and Meta-Analysis

Zubina Khan, Majumi M Noohu, Sarah Parveen, Maria Usmani, Fayaz Khan, Mashael Ghazi Alsobhi, Md Dilshad Manzar, Chhavi Arora Sehgal

Review Dev Neurorehabil. 2024 May 7:1-10. doi: 10.1080/17518423.2024.2349676. Online ahead of print.

Background: This review aimed to explore the effect of mirror therapy (MT) on upper limb function in children and adolescents with hemiplegic cerebral palsy (HCP). Methods: MEDLINE, CENTRAL, Scopus, PEDro, and Web of Science were systematically searched. PEDro scale was used for the quality assessment of included trials. Risk of Bias assessment was done using Cochrane Risk-of-bias tool version 2. Meta-analysis was performed on four of the seven studies included. Results & conclusion: The majority of the trials included in this review found MT efficacious in improving motor function in HCP. Quantitative analysis of the included trials using QUEST scores for evaluation of quality of upper extremity function revealed positive but non-significant difference between the groups (MD = -0.12; 95% CI = -2.57,2.33; Z = 0.09, p = .92). Pooled analysis of the included trials using BBT, however, favored control (MD = 4.98; 95% CI = 2.32,7.63; Z = 3.67, p = .0002).

PMID: 38712882

2. How Does Postural Control of Children with CP Deal with Manipulations on Base of Support and Support Surface? A Systematic Review

Giovana Z da Conceição, Marina Gardinal, Giovanna M Kreutz, Nelci Adriana C F Rocha, Silvia Leticia Pavão

Review Dev Neurorehabil. 2024 May 8:1-13. doi: 10.1080/17518423.2024.2348002. Online ahead of print.

Aims: Systematically review literature addressing the effects of changes in base of support (BoS) configuration and characteristics of support surface (SS) on postural control of children with cerebral palsy (CP). Methods: We conducted a tailored electronic database search in PubMed/Web of Science/SCOPUS/Embase. Results: We identified 15 studies meeting inclusion criteria. Conclusion: The extant literature suggests that when children with CP experience changes in BoS and SS, they engage in fewer adaptive postural control responses than typically developing children. Documented response patterns of children with CP in the literature might guide the selection and development of rehabilitation strategies to appropriately facilitate or challenge postural control in children with CP.

PMID: 38720440

3. Optimizing Hip Replacement Procedure in Cerebral Palsy-Related Spastic Hip Dysplasia: A Case Report

Androniki Drakou, Pavlos Altsitzioglou, Spyridon Sioutis, Anastasios G Roustemis, Dimitrios S Mastrokalos, Dimitrios Koulalis

Case Reports Cureus. 2024 Apr 4;16(4):e57584. doi: 10.7759/cureus.57584. eCollection 2024 Apr.

Cerebral palsy (CP) often results in severe hip issues, disrupting musculoskeletal development and mobility due to problems such as dislocations and contractures, aggravated by spasticity and heightened muscular tone. While total hip arthroplasty (THA) is required in CP patients, the procedure carries high risks due to concerns about dislocation and wear. This study explores a method of intraoperative navigation to precisely execute preoperative strategies for spinopelvic alignment and optimal cup placement. We discuss a case of a 22-year-old male CP patient with bilateral hip dislocations who experienced significant discomfort, impeding mobility and affecting his performance as a Paralympic rower. He underwent bilateral hip replacement surgeries, guided by preoperative gait analysis and imaging, with navigation aiding in accurate acetabular component placement and correction of excessive femoral anteversion using a modular stem. The patient achieved excellent stability in both standing and rowing postures. Overall, computer navigation enhances complex hip repair by facilitating intraoperative data collection and precise execution of preoperative plans. This approach may extend the lifespan of prostheses, particularly by achieving precise acetabular component placement based on spinopelvic alignment principles, thereby offering significant benefits for CP patients undergoing THA.

PMID: 38707032

4. Pelvic floor symptoms and symptom-related quality of life in the mothers of children with cerebral palsy: a pilot cross -sectional study

Damla Korkmaz Dayican, Zeynep Hosbay, Burcin Ozyurek, Gulsena Utku Umut

Women Health. 2024 May 5:1-12. doi: 10.1080/03630242.2024.2349559. Online ahead of print.

Pelvic floor symptoms may occur in women with low back pain due to dysfunction of the spinal stabilization muscles. Low back pain is the most common musculoskeletal problem experienced by the mothers of children with cerebral palsy (CP). Therefore, our aim in this study was to examine pelvic floor symptoms and symptom-related quality of life in the mothers of children with CP. The study included 48 mothers of children with CP (n = 23) or without neurodevelopmental problems (n = 25). The mothers' pelvic floor symptoms and symptom-related quality of life were evaluated with the Pelvic Floor Distress Inventory-20 (PFDI-20) and the Pelvic Floor Impact Questionnaire-7 (PFIQ-7). PFDI-20 and PFIQ-7 scores did not differ between the two groups of mothers ($p > .05$). Additionally, moderately significant positive correlations were found between the age of the child with CP and duration of carrying and the PFDI-20 ($r = 0.419$, $p = .047$) and PFIQ-7 ($r = 0.427$, $p = .042$) scores. Pelvic floor symptoms and symptom-related quality of life in the mothers of children with CP were similar to those in the mothers of children without neurodevelopmental problems. As the age of the child with CP and duration of carrying increase, the urinary symptoms and urinary and colorectoanal symptoms-related quality of life in their mothers may worsen.

PMID: 38706221

5. Knee and ankle range of motion and spasticity from childhood into adulthood: a longitudinal cohort study of 3,223 individuals with cerebral palsy

Erika Cloodt, Anna Lindgren, Elisabet Rodby-Bousquet

Acta Orthop. 2024 May 6:95:200-205. doi: 10.2340/17453674.2024.40606.

Background and purpose: Reduced range of motion (ROM) and spasticity are common secondary findings in cerebral palsy (CP) affecting gait, positioning, and everyday functioning. These impairments can change over time and lead to various needs for intervention. The aim of this study was to analyze the development path of the changes in hamstring length, knee extension, ankle dorsiflexion, and spasticity in hamstrings and gastrosoleus from childhood into adulthood in individuals with CP at the Gross Motor Function Classification System (GMFCS) levels I-V. Methods: A longitudinal cohort study was undertaken of 61,800 measurements in 3,223 individuals with CP, born 1990-2017 and followed for an average of 8.7 years (range 0-26). The age at examination varied between 0 and 30 years. The GMFCS levels I-V, goniometric measurements, and the modified Ashworth scale (MAS) were used for repeated assessments of motor function, ROM, and spasticity. Results: Throughout the follow-up period, knee extension and hamstring length exhibited a consistent decline across all individuals, with more pronounced decreases evident in those classified at GMFCS levels III-V. Ankle dorsiflexion demonstrated a gradual reduction from 15° to 5° (GMFCS I-IV) or 10° (GMFCS V). Spasticity levels in the hamstrings and gastrosoleus peaked between ages 5 and 7, showing a propensity to increase with higher GMFCS levels. Conclusion: Passive ROM continues to decrease to 30 years of age, most pronouncedly for knee extension. Conversely, spasticity reached its peak at a younger age, with a more notable occurrence observed in the gastrosoleus compared with the hamstrings. Less than 50% of individuals had spasticity corresponding to MAS 2-4 at any age.

PMID: 38708569

6. A systematic review and meta-analysis of long-term outcomes of femoral derotation surgery for intoeing gait in cerebral palsy

Sitanshu Barik, Sunny Chaudhary, Vishal Kumar, Vikash Raj, Vivek Singh

Gait Posture. 2024 Apr 26:112:1-7. doi: 10.1016/j.gaitpost.2024.04.027. Online ahead of print.

Background: Femoral derotation osteotomy is treatment of choice in intoeing gait secondary to cerebral palsy (CP). Research question: The aim of this study was to critically appraise the literature regarding the long-term outcomes of femoral derotation surgery in CP. Methods: Electronic databases of PubMed and Scopus was used for the literature review by two researchers independently (SB, SC). The study population included patients of cerebral palsy undergoing femoral derotation surgery. The keywords used were "cerebral palsy", "intoeing gait", "femur anteversion", "hip anteversion", "femur derotation" and "femur osteotomy". Results: Nine studies which included 657 limbs in 407 patients were selected for this study of which eight were retrospective in nature. The improvement in hip rotation at stance at last follow up (more than five years) maintained a statistical significance (SMD 1.67 95 %CI 1.12-2.22). Similar statistically significant outcomes were noted for foot progression angle (SMD 1.19 95 %CI 0.92-1.47), anteversion (SMD 2.75 95 %CI 1.49-4.01) and total passive internal rotation (SMD 1.71 95 %CI 1.19-2.22). Significance: Femoral derotation surgery is the procedure of choice for correction of intoeing gait in CP. Even though, there is deterioration of results on long-term as compared to short term, majority of the patients shall maintain overall correction without recurrence of an intoeing gait. Future studies with uniform criteria for defining recurrence on the basis of functional limitations shall provide better idea about the natural course of this procedure.

PMID: 38718437

7. Filtering walking actigraphy data in children with unilateral cerebral palsy: A preliminary study

Youngsub Hwang, Jeong-Yi Kwon

Randomized Controlled Trial PLoS One. 2024 May 9;19(5):e0303090. doi: 10.1371/journal.pone.0303090. eCollection 2024.

This study aimed to determine whether filtering out walking-related actigraphy data improves the reliability and accuracy of real-world upper extremity activity assessment in children with unilateral cerebral palsy. Twenty-two children aged 4-12 years diagnosed with unilateral cerebral palsy were included in this study, which was drawn from a two-phase randomized controlled trial conducted from July 2021 to December 2022. Data were collected from a tertiary hospital in Seoul, Republic of Korea. Participants were monitored using tri-axial accelerometers on both wrists across three time points (namely, T0, T1, and T2) over 3 days; interventions were used between each time point. Concurrently, an in-laboratory study focusing on walking and bimanual tasks was conducted with four participants. Data filtration resulted in a reduction of 8.20% in total data entry. With respect to reliability assessment, the intra-class correlation coefficients indicated enhanced consistency after filtration, with increased values for both the affected and less-affected sides. Before filtration, the magnitude counts for both sides showed varying tendencies, depending on the time points; however, they presented a consistent and stable trend after filtration. The findings of this research underscore the importance of accurately interpreting actigraphy measurements in children with unilateral cerebral palsy for targeted upper limb intervention by filtering walking-induced data.

PMID: 38722902

8. Electromyographic biofeedback training in a cerebral palsy patient undergoing pronator teres rerouting and brachioradialis to extensor carpi radialis brevis tendon transfer surgery: A case report

Hande Usta Ozdemir, Ali Kitis, Ahmet Fahir Demirkan

Case Reports Jt Dis Relat Surg. 2024 Feb 13;35(2):448-454. doi: 10.52312/jdrs.2024.962. Epub 2024 Feb 13.

Several surgical procedures are used to treat dynamic pronation position of the forearm and flexion deformity of the wrist in cerebral palsy. Postoperative results of pronator teres rerouting were explored, while specially designed postoperative physiotherapy and its outcomes were limited. Herein, we present a case in whom the outcomes of electromyographic biofeedback (EMG-BF) training were assessed after pronator teres rerouting and brachioradialis tendon to extensor carpi radialis brevis tendon transfer combined with derotation osteotomy. The peak value increased, while the resting value decreased for the muscles after the intervention. Range of motion, hand function, manual ability, functional independence, and quality of life levels were improved. In conclusion, EMG biofeedback training may have a positive effect on neuromuscular control of pronator teres and brachioradialis. Free use of the upper extremity and improved manual ability positively affect the activity and quality of life of the patients.

PMID: 38727128

9. The use of shear-wave elastography for the assessment of muscle spasticity in patients with cerebral palsy, a systematic review

Edoardo Cesaro, Thomas Saliba, Paolo Simoni

Review J Clin Ultrasound. 2024 May 6. doi: 10.1002/jcu.23706. Online ahead of print.

We explore the use of shear wave elastography (SWE) for assessing muscle stiffness and treatment response in cerebral palsy (CP) children by way of a systematic review. SWE offers real-time muscle stiffness measurements, showing significant differences between CP patients and controls. Studies suggest that SWE can be used to follow muscle stiffness post-botulinum toxin treatment, correlating with clinical improvement. However, methodological variations and small sample sizes prevent comparison between different studies. Standardized protocols could enhance SWE's clinical utility. In conclusion, SWE holds promise for CP management, though standardized methodologies and larger studies are needed to validate its efficacy and integration into clinical practice.

PMID: 38708803

10. The power of Para sport: the effect of performance-focused swimming training on motor function in adolescents with cerebral palsy and high support needs (GMFCS IV) - a single-case experimental design with 30-month follow-up

Iain Mayank Dutia, Mark Connick, Emma Beckman, Leanne Johnston, Paula Wilson, Angelo Macaro, Jennifer O'Sullivan, Sean Tweedy

Br J Sports Med. 2024 May 10:bjsports-2023-107689. doi: 10.1136/bjsports-2023-107689. Online ahead of print.

Objective: This study aims to evaluate the effect of a performance-focused swimming programme on motor function in previously untrained adolescents with cerebral palsy and high support needs (CPHSN) and to determine whether the motor decline typical of adolescents with CPHSN occurred in these swimmers. Methods: A Multiple-Baseline, Single-Case Experimental Design (MB-SCED) study comprising five phases and a 30-month follow-up was conducted. Participants were two males and one female, all aged 15 years, untrained and with CPHSN. The intervention was a 46-month swimming training programme, focused exclusively on improving performance. Outcomes were swim performance (velocity); training load (rating of perceived exertion min/week; swim distance/week) and Gross Motor Function Measure-66-Item Set (GMFM-66). MB-SCED data were analysed using interrupted time-series simulation analysis. Motor function over 46 months was modelled (generalised additive model) using GMFM-66 scores and compared with a model of predicted motor decline. Results: Improvements in GMFM-66 scores in response to training were significant ($p<0.001$), and two periods of training withdrawal each resulted in significant motor decline ($p\leq0.001$). Participant motor function remained above baseline levels for the study duration, and, importantly, participants did not experience the motor decline typical of other adolescents with CPHSN. Weekly training volumes were also commensurate with WHO recommended physical activity levels. Conclusions: Results suggest that adolescents with CPHSN who meet physical activity guidelines through participation in competitive swimming may prevent motor decline. However, this population is clinically complex, and in order to permit safe, effective participation in competitive sport, priority should be placed on the development of programmes delivered by skilled multiprofessional teams. Trial registration number: ACTRN12616000326493.

PMID: 38729630

11. Neuralink and Brain-Computer Interface-Exciting Times for Artificial Intelligence

Purvish Mahendra Parikh, Ajit Venniyoor

South Asian J Cancer. 2024 Apr 15;13(1):63-65. doi: 10.1055/s-0043-1774729. eCollection 2024 Jan.

Purvish Mahendra ParikhBrain-computer interfaces are becoming a tangible reality, capable of significantly aiding patients in real-world scenarios. The recent approval by the U.S. Food and Drug Administration for clinical human trials of Neuralink marks a monumental stride, comparable to Mr. Armstrong's moonwalk. Numerous other companies are also pioneering innovative solutions in this domain. Presently, over 150,000 patients in the United States possess brain implants. As technology advances, it holds the potential to alleviate various conditions, notably motor paralysis, cerebral palsy, and involuntary movements.

PMID: 38721102

12. A flexible intracortical brain-computer interface for typing using finger movements

Nishal P Shah, Matthew S Willsey, Nick Hahn, Foram Kamdar, Donald T Avansino, Chaofei Fan, Leigh R Hochberg, Francis R Willett, Jaimie M Henderson

bioRxiv [Preprint]. 2024 Apr 26:2024.04.22.590630. doi: 10.1101/2024.04.22.590630.

Keyboard typing with finger movements is a versatile digital interface for users with diverse skills, needs, and preferences. Currently, such an interface does not exist for people with paralysis. We developed an intracortical brain-computer interface (BCI) for typing with attempted flexion/extension movements of three finger groups on the right hand, or both hands, and demonstrated its flexibility in two dominant typing paradigms. The first paradigm is "point-and-click" typing, where a BCI user selects one key at a time using continuous real-time control, allowing selection of arbitrary sequences of symbols. During cued character selection with this paradigm, a human research participant with paralysis achieved 30-40 selections per minute with nearly 90% accuracy. The second paradigm is "keystroke" typing, where the BCI user selects each character by a discrete movement without real-time feedback, often giving a faster speed for natural language sentences. With 90 cued characters per minute, decoding attempted finger movements and correcting errors using a language model resulted in more than 90% accuracy. Notably, both paradigms matched the state-of-the-art for BCI performance and enabled further flexibility by the simultaneous selection of multiple characters as well as efficient decoder estimation across paradigms. Overall, the high-performance interface is a step towards the wider accessibility of BCI technology by addressing unmet user needs for flexibility.

PMID: 38712189

13. Appraising the Physical Activity Levels of Saudis with Physical Disabilities: Effects of Disability Type, Mobility Assistive Devices, and Demographic Factors

Mohamed A Said, Majed M Alhumaid

Healthcare (Basel). 2024 May 2;12(9):937. doi: 10.3390/healthcare12090937.

Physical activity (PA) has numerous health benefits for individuals with physical disabilities (IWPD). However, it is common for activity levels to fall below the suggested limits. This study aimed to evaluate the prevalence, pattern, and levels of PA among IWPD in Saudi Arabia. It also investigated the effects of individuals' type of disability, mobility assistive devices, and demographic features on PA levels. Data were collected from 238 participants, mostly male (62.2%), aged 39.76 ± 12.19 years. Among them, 19.3% had spinal conditions, 14.7% had progressive muscular dystrophy, 15.1% had multiple sclerosis, 17.6% had cerebral palsy, 16.4% had poliomyelitis, and 16.8% had limb or foot amputations. The participants were assessed using the Arabic version of the Physical Activity Scale for Individuals with Physical Disabilities (PASIPD-AR). The results showed that 62.6% (64.9% of males and 58.9% of females) met the minimum PA guidelines specified by the WHO. The average PASIPD-AR score was 10.33 ± 10.67 MET-hours/day, indicating lower PA levels, and 8.4% of individuals did not participate in any form of PA. Significant discrepancies were detected in disability type and mobility assistive device use after age adjustment. Marital status, education, and occupation greatly affected PA components. Greater attention should be paid to promoting an active lifestyle among IWPD in Saudi Arabia.

PMID: 38727494

14. Clinical characteristics and rehabilitation potential in children with cerebral palsy based on MRI classification system

Jie Yang, Congjie Chen, Ningning Chen, Helin Zheng, Yuxia Chen, Xiaoli Li, Qingxia Jia, Tingsong Li

Front Pediatr. 2024 Apr 25:12:1382172. doi: 10.3389/fped.2024.1382172. eCollection 2024.

Background: The correlation of clinical characteristics of cerebral palsy (CP) and the magnetic resonance imaging classification system (MRICS) for (CP) is inconsistent. Specifically, the variance in rehabilitation potential across MRICS remains underexplored. Aims: To investigate the clinical characteristics and potential for rehabilitation in children with CP based on MRICS. Materials and methods: Children with CP admitted to the Department of Rehabilitation, Children's Hospital of Chongqing Medical University between 2017 and 2021 were included in the study. Qualified cases underwent a follow-up period of at least one year. The clinical characteristics of CP among different MRICS were analyzed, then the rehabilitation potential was explored by a retrospective cohort study. Results: Among the 384 initially enrolled children, the male-to-female ratio was 2.3:1, and the median age of diagnosis was 6.5 months (interquartile range: 4-12). The most prevalent MRICS categorization was predominant white matter injury (40.6%), followed by miscellaneous (29.2%) and predominant gray matter injury (15.6%). For the predominant white matter injury and miscellaneous categories, spastic diplegia emerged as the leading subtype of CP, with incidences of 59.6% and 36.6%, respectively, while mixed CP (36.7%) was the most common type in children with predominant gray matter. Notably, 76.4% of children with predominant white matter injury were classified as levels I-III on the gross motor function classification system (GMFCS), indicating significantly less severity than other groups ($\chi 2 = 12.438$, $p = 0.013$). No significant difference across MRICS categories was observed for the manual ability classification system (MACS) ($H = 8.176$, $p = 0.085$). Rehabilitation potential regarding fine motor function and adaptability based on Gesell assessment was dependent on MRICS over the follow-up period. Children with normal MRI scans exhibited superior rehabilitation outcomes. Commencing rehabilitation at an earlier stage produced consistent and beneficial results in terms of fine motor function and adaptability across all MRICS categories. Moreover, participants below 2 years of age demonstrated

enhanced rehabilitation potential regarding fine motor outcomes and adaptability within the MRICS framework. Conclusion: MRICS displayed a significant association with clinical characteristics and rehabilitation efficacy in children with CP.

PMID: 38725982

15. Early administration of umbilical cord blood cells following brief high tidal volume ventilation in preterm sheep: a cautionary tale

Nhi T Tran, Tayla R Penny, Kyra Yy Chan, Tanya Tang, Paris C Papagianis, Tara Sepehrizadeh, Lakshmi Nekkanti, Valerie A Zahra, Yen Pham, Tamara Yawno, Ilias Nitsos, Sharmony B Kelly, Alison M Thiel, Michael de Veer, Dhafer M Alahmari, Michael C Fahey, Graham Jenkin, Suzanne L Miller, Robert Galinsky, Graeme R Polglase, Courtney A McDonald

J Neuroinflammation. 2024 May 8;21(1):121. doi: 10.1186/s12974-024-03053-3.

Background: Umbilical cord blood (UCB) cells are a promising treatment for preterm brain injury. Access to allogeneic sources of UCB cells offer the potential for early administration to optimise their therapeutic capacities. As preterm infants often require ventilatory support, which can contribute to preterm brain injury, we investigated the efficacy of early UCB cell administration following ventilation to reduce white matter inflammation and injury. Methods: Preterm fetal sheep (0.85 gestation) were randomly allocated to no ventilation (SHAM; n = 5) or 15 min ex utero high tidal volume ventilation. One hour following ventilation, fetuses were randomly allocated to i.v. administration of saline (VENT; n = 7) or allogeneic term-derived UCB cells (24.5 ± 5.0 million cells/kg; VENT + UCB; n = 7). Twenty-four hours after ventilation, lambs were delivered for magnetic resonance imaging and post-mortem brain tissue collected. Arterial plasma was collected throughout the experiment for cytokine analyses. To further investigate the results from the in vivo study, mononuclear cells (MNCs) isolated from human UCB were subjected to in vitro cytokine-spiked culture medium (TNFα and/or IFNγ; 10 ng/mL; n = 3/group) for 16 h then supernatant and cells collected for protein and mRNA assessments respectively. Results: In VENT + UCB lambs, systemic IFNγ levels increased and by 24 h, there was white matter neuroglial activation, vascular damage, reduced oligodendrocytes, and increased average, radial and mean diffusivity compared to VENT and SHAM. No evidence of white matter inflammation or injury was present in VENT lambs, except for mRNA downregulation of OCLN and CLDN1 compared to SHAM. In vitro, MNCs subjected to TNFα and/or IFNγ displayed both pro- and anti-inflammatory characteristics indicated by changes in cytokine (IL-18 & IL-10) and growth factor (BDNF & VEGF) gene and protein expression compared to controls. Conclusions: UCB cells administered early after brief high tidal volume ventilation in preterm fetal sheep causes white matter injury, and the mechanisms underlying these changes are likely dysregulated responses of the UCB cells to the degree of injury/inflammation already present. If immunomodulatory therapies such as UCB cells are to become a therapeutic strategy for preterm brain injury, especially after ventilation, our study suggests that the inflammatory state of the preterm infant should be considered when timing UCB cells administration.

PMID: 38720368

16. Inter-rater reliability and agreement of the General Movement Assessment and Motor Optimality Score-Revised in a large population-based sample

Caroline Alexander, Natasha Amery, Alison Salt, Catherine Morgan, Alicia Spittle, Robert S Ware, Catherine Elliott, Jane Valentine

Early Hum Dev. 2024 Apr 25:193:106019. doi: 10.1016/j.earlhumdev.2024.106019. Online ahead of print.

Background: Prechtl's General Movement Assessment (GMA) at fidgety age (3-5 months) is a widely used tool for early detection of cerebral palsy. Further to GMA classification, detailed assessment of movement patterns at fidgety age is conducted with the Motor Optimality Score-Revised (MOS-R). Inter-rater reliability and agreement are properties that inform test application and interpretation in clinical and research settings. This study aims to establish the inter-rater reliability and agreement of the GMA classification and MOS-R in a large population-based sample. Methods: A cross-sectional study of 773 infants from birth-cohort in Perth, Western Australia. GMA was conducted on home-recorded videos collected between 12 + 0 and 16 + 6 weeks post term age. Videos were independently scored by two masked experienced assessors. Inter-rater reliability and agreement were assessed using intraclass correlation coefficient and limits of agreement respectively for continuous variables, and Cohen's Kappa and Gwet's Agreement Coefficient, and percentage agreement respectively for discrete variables. Results: The classification of GMA showed almost perfect reliability (AC1 = 0.999) and agreement (99.9 %). Total MOS-R scores showed good-excellent reliability (ICC 0.857, 95 % CI 0.838-0.876) and clinically acceptable agreement (95 % limits of agreement of ±2.5 points). Substantial to almost perfect reliability and agreement were found for all MOS-R domain subscores. While MOS-R domains with higher redundancy in their categorisation have higher reliability and agreement, inter-rater reliability and agreement are substantial to almost perfect at the item level and are consistent across domains. Conclusion: GMA at fidgety age shows clinically acceptable inter-rater reliability and agreement for GMA classification and MOS-R for population-based cohorts assessed by experienced assessors.

PMID: 38718464

17. Exome sequencing in a Chinese cohort of children with cerebral palsy identifies likely pathogenic variants

No authors listed

Nat Med. 2024 May 7. doi: 10.1038/s41591-024-02967-y. Online ahead of print.

No abstract available

PMID: 38714899

18. A Holistic Approach for Physiotherapy Rehabilitation of Girdlestone Arthroplasty With Infection and Concomitant Contralateral Spastic Hemiplegic Cerebral Palsy: A Case Report

Sojwal P Nandanwar, Swapnil U Ramteke

Case Reports Cureus. 2024 Apr 5;16(4):e57689. doi: 10.7759/cureus.57689. eCollection 2024 Apr.

Girdlestone arthroplasty is a traditional approach for complicated infections occurring with contralateral spastic hemiplegic cerebral palsy, which presents intricate challenges in rehabilitation. In this case report, an 18-year-old girl came to a multispecialty hospital with a history of falls. She was an identified case of femoral head dislocation with acute osteomyelitis and a history of spastic hemiplegic cerebral palsy. She underwent girdlestone arthroplasty with additional upper tibial and ankle pin traction. After that, she was referred to physiotherapy management. To further aid recovery, rehabilitation protocol included a combination of static exercises, ankle pumps on the affected side, and stretching, bimanual hand-arm training with lower limb training on the unaffected side to reduce spasticity. Once the stitches were removed and traction discontinued, the focus shifted to improving mobility through basic activities like rolling and transitioning to sitting, gradually progressing to standing with the assistance of a walker and bimanual hand-arm training with lower limb training for spasticity. Outcome measures like functional independence measure, numerical pain rating scale, range of motion, and manual ability classification system were used to record patient progress during rehabilitation. This case report serves the crucial role physiotherapy plays in the treatment of orthopedic and neurological conditions in younger patients, with the ultimate goal of regaining functional independence and enhancing overall quality of life.

PMID: 38711685

19. 'High hopes for treatment': Australian stakeholder perspectives of the clinical translation of advanced neurotherapeutics for rare neurological diseases

Christina Q Nguyen, Didu S T Kariyawasam, Tsz Shun Jason Ngai, James Nguyen, Kristine Alba-Concepcion, Sarah E Grattan, Elizabeth E Palmer, Kate Hetherington, Claire E Wakefield, Russell C Dale, Sue Woolfenden, Shekeeb Mohammad, Michelle A Farrar

Health Expect. 2024 Jun;27(3):e14063. doi: 10.1111/hex.14063.

Introduction: Advanced therapies offer unprecedented opportunities for treating rare neurological disorders (RNDs) in children. However, health literacy, perceptions and understanding of novel therapies need elucidation across the RND community. This study explored healthcare professionals' and carers' perspectives of advanced therapies in childhood-onset RNDs. Methods: In this mixed-methodology cross-sectional study, 20 healthcare professionals (clinicians, genetic counsellors and scientists) and 20 carers completed qualitative semistructured interviews and custom-designed surveys. Carers undertook validated psychosocial questionnaires. Thematic and quantitative data analysis followed. Results: Participants described high positive interest in advanced therapies, but low knowledge of, and access to, reliable information. The substantial 'therapeutic gap' and 'therapeutic odyssey' common to RNDs were recognised in five key themes: (i) unmet need and urgency for access; (ii) seeking information; (iii) access, equity and sustainability; (iv) a multidisciplinary and integrated approach to care and support and (v) difficult decision-making. Participants were motivated to intensify RND clinical trial activity and access to advanced therapies; however, concerns around informed consent, first-in-human trials and clinical trial procedures were evident. There was high-risk tolerance despite substantial uncertainties and knowledge gaps. RNDs with high mortality, increased functional burdens and no alternative therapies were consistently prioritised for the development of advanced therapies. However, little consensus existed on prioritisation to treatment access. Conclusions: This study highlights the need to increase clinician and health system readiness for the clinical translation of advanced therapeutics for RNDs. Co-development and use of educational and psychosocial resources to support clinical decision-making, set therapeutic expectations and promotion of equitable, effective and safe delivery of advanced therapies are essential. Patient or public contribution: Participant insights into the psychosocial burden and information need to enhance the delivery of care in this formative study are informing ongoing partnerships with families, including co-production and dissemination of psychoeducational resources featuring their voices hosted on the Sydney Children's Hospitals Network website SCHN Brain-Aid Resources.

PMID: 38711219

20. Early Neurodevelopmental Assessments for Predicting Long-Term Outcomes in Infants at High Risk of Cerebral Palsy

Abdul Razak, Emily Johnston, Vathana Sackett, Marissa Clark, Margaret Charlton, Lindsay Zhou, Pramod Pharande, Courtney A McDonald, Rod W Hunt, Suzanne L Miller, Atul Malhotra

JAMA Netw Open. 2024 May 1;7(5):e2413550. doi: 10.1001/jamanetworkopen.2024.13550.

Importance: Studies suggest that early neurodevelopmental assessments are beneficial for identifying cerebral palsy, yet their effectiveness in practical scenarios and their ability to detect cognitive impairment are limited. Objective: To assess the effectiveness of early neurodevelopmental assessments in identifying cerebral palsy and cognitive and other neurodevelopmental impairments, including their severity, within a multidisciplinary clinic. Design, setting, and participants: This diagnostic study was conducted at Monash Children's Hospital, Melbourne, Australia. Participants were extremely preterm infants born at less than 28 weeks' gestation or extremely low birth weight infants less than 1000 g and term encephalopathic infants who received therapeutic hypothermia, attending the early neurodevelopmental clinic between January 2019 and July 2021. Data were analyzed from December 2023 to January 2024. Exposures: Early cerebral palsy or high risk of cerebral palsy, the absence of fidgety movements, and Hammersmith Infant Neurological Examination (HINE) scores at corrected age (CA) 3 to 4 months. Early cerebral palsy or high risk of cerebral palsy diagnosis was based on absent fidgety movements, a low HINE score (<57), and medical neurological examination. Main outcome and measures: The outcomes of interest were cerebral palsy, cognitive and neurodevelopmental impairments and their severity, diagnosed at 24 to 36 months' CA. Results: A total of 116 infants (median [IQR] gestational age, 27 [25-29] weeks; 65 [56%] male) were included. Diagnosis of early cerebral palsy or high risk of cerebral palsy demonstrated a sensitivity of 92% (95% CI, 63%-99%) and specificity of 84% (95% CI, 76%-90%) for predicting cerebral palsy and 100% (95% CI, 59%-100%) sensitivity and 80% (95% CI, 72%-87%) specificity for predicting moderate to severe cerebral palsy. Additionally, the accuracy of diagnosis of early cerebral palsy or high risk of cerebral palsy was 85% (95% CI, 77%-91%) for predicting cerebral palsy and 81% (95% CI, 73%-88%) for predicting moderate to severe cerebral palsy. Similarly, the absence of fidgety movements had an 81% (95% CI, 73%-88%) accuracy in predicting cerebral palsy, and HINE scores exhibited good discriminatory power with an area under the curve of 0.88 (95% CI, 0.79-0.97) for cerebral palsy prediction. However, for cognitive impairment, the predictive accuracy was 44% (95% CI, 35%-54%) for an early cerebral palsy or high risk of cerebral palsy diagnosis and 45% (95% CI, 36%-55%) for the absence of fidgety movements. Similarly, HINE scores showed poor discriminatory power for predicting cognitive impairment, with an area under the curve of 0.62 (95% CI, 0.51-0.73). Conclusions and relevance: In this diagnostic study of infants at high risk for cerebral palsy or other cognitive or neurodevelopmental impairment, early neurodevelopmental assessments at 3 to 4 months' CA reliably predicted cerebral palsy and its severity at 24 to 36 months' CA, signifying its crucial role in facilitating early intervention. However, for cognitive impairment, longer-term assessments are necessary for accurate identification.

PMID: 38709738

Prevention and Cure

21. Magnesium sulphate for women at risk of preterm birth for neuroprotection of the fetus

Emily S Shepherd, Shona Goldsmith, Lex W Doyle, Philippa Middleton, Stéphane Marret, Dwight J Rouse, Peter Pryde, Hanne T Wolf, Caroline A Crowther

Review Cochrane Database Syst Rev. 2024 May 10;5(5):CD004661. doi: 10.1002/14651858.CD004661.pub4.

Background: Magnesium sulphate is a common therapy in perinatal care. Its benefits when given to women at risk of preterm birth for fetal neuroprotection (prevention of cerebral palsy for children) were shown in a 2009 Cochrane review. Internationally, use of magnesium sulphate for preterm cerebral palsy prevention is now recommended practice. As new randomised controlled trials (RCTs) and longer-term follow-up of prior RCTs have since been conducted, this review updates the previously published version. Objectives: To assess the effectiveness and safety of magnesium sulphate as a fetal neuroprotective agent when given to women considered to be at risk of preterm birth. Search methods: We searched Cochrane Pregnancy and Childbirth's Trials Register, ClinicalTrials.gov, and the World Health Organization (WHO) International Clinical Trials Registry Platform (ICTRP) on 17 March 2023, as well as reference lists of retrieved studies. Selection criteria: We included RCTs and cluster-RCTs of women at risk of preterm birth that assessed prenatal magnesium sulphate for fetal neuroprotection compared with placebo or no treatment. All methods of administration (intravenous, intramuscular, and oral) were eligible. We did not include studies where magnesium sulphate was used with the primary aim of preterm labour tocolysis, or the prevention and/or treatment of eclampsia. Data collection and analysis: Two review authors independently assessed RCTs for inclusion, extracted data, and assessed risk of bias and trustworthiness. Dichotomous data were presented as summary risk ratios (RR) with 95% confidence intervals (CI), and continuous data were presented as mean differences with 95% CI. We assessed the certainty of the evidence using the GRADE approach. Main results: We included six RCTs (5917 women and their 6759 fetuses alive at randomisation). All RCTs were conducted in high-income countries. The RCTs compared magnesium sulphate with placebo in women at risk of preterm birth at less than 34 weeks' gestation; however,

treatment regimens and inclusion/exclusion criteria varied. Though the RCTs were at an overall low risk of bias, the certainty of evidence ranged from high to very low, due to concerns regarding study limitations, imprecision, and inconsistency. Primary outcomes for infants/children: Up to two years' corrected age, magnesium sulphate compared with placebo reduced cerebral palsy (RR 0.71, 95% CI 0.57 to 0.89; 6 RCTs, 6107 children; number needed to treat for additional beneficial outcome (NNTB) 60, 95% CI 41 to 158) and death or cerebral palsy (RR 0.87, 95% CI 0.77 to 0.98; 6 RCTs, 6481 children; NNTB 56, 95% CI 32 to 363) (both high-certainty evidence). Magnesium sulphate probably resulted in little to no difference in death (fetal, neonatal, or later) (RR 0.96, 95% CI 0.82 to 1.13; 6 RCTs, 6759 children); major neurodevelopmental disability (RR 1.09, 95% CI 0.83 to 1.44; 1 RCT, 987 children); or death or major neurodevelopmental disability (RR 0.95, 95% CI 0.85 to 1.07; 3 RCTs, 4279 children) (all moderate-certainty evidence). At early school age, magnesium sulphate may have resulted in little to no difference in death (fetal, neonatal, or later) (RR 0.82, 95% CI 0.66 to 1.02; 2 RCTs, 1758 children); cerebral palsy (RR 0.99, 95% CI 0.69 to 1.41; 2 RCTs, 1038 children); death or cerebral palsy (RR 0.90, 95% CI 0.67 to 1.20; 1 RCT, 503 children); and death or major neurodevelopmental disability (RR 0.81, 95% CI 0.59 to 1.12; 1 RCT, 503 children) (all low-certainty evidence). Magnesium sulphate may also have resulted in little to no difference in major neurodevelopmental disability, but the evidence is very uncertain (average RR 0.92, 95% CI 0.53 to 1.62; 2 RCTs, 940 children; very low-certainty evidence). Secondary outcomes for infants/children: Magnesium sulphate probably reduced severe intraventricular haemorrhage (grade 3 or 4) (RR 0.76, 95% CI 0.60 to 0.98; 5 RCTs, 5885 infants; NNTB 92, 95% CI 55 to 1102; moderate-certainty evidence) and may have resulted in little to no difference in chronic lung disease/bronchopulmonary dysplasia (average RR 0.92, 95% CI 0.77 to 1.10; 5 RCTs, 6689 infants; low-certainty evidence). Primary outcomes for women: Magnesium sulphate may have resulted in little or no difference in severe maternal outcomes potentially related to treatment (death, cardiac arrest, respiratory arrest) (RR 0.32, 95% CI 0.01 to 7.92; 4 RCTs, 5300 women; low-certainty evidence). However, magnesium sulphate probably increased maternal adverse effects severe enough to stop treatment (average RR 3.21, 95% CI 1.88 to 5.48; 3 RCTs, 4736 women; moderate-certainty evidence). Secondary outcomes for women: Magnesium sulphate probably resulted in little to no difference in caesarean section (RR 0.96, 95% CI 0.91 to 1.02; 5 RCTs, 5861 women) and postpartum haemorrhage (RR 0.94, 95% CI 0.80 to 1.09; 2 RCTs, 2495 women) (both moderate-certainty evidence). Breastfeeding at hospital discharge and women's views of treatment were not reported.

PMID: 38726883