

Cerebral palsy research news

Monday 15 January 2024

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Interventions and Management

1. Tendon transfer in spastic cerebral palsy upper limb

Hadi Gerami, G Hossain Shahcheraghi, Mahzad Javid

J Pediatr Orthop B. 2023 Dec 29. doi: 10.1097/BPB.00000000001137. Online ahead of print.

Cerebral palsy (CP) in upper limb produces functional, aesthetic and hygienic issues, and is not always amenable to surgical procedures. We are reporting a single-center, long follow-up experience with tendon transfer in wrist and forearm CP. The CP cases who had undergone tendon transfer in hand, wrist and forearm in a 14-year period were evaluated for change in motion, function and cosmetic appearance and also assessed by Manual Ability Classification System (MACS), and Dash (Disability of Arm, Shoulder, Hand) scores. Forty-two spastic CP patients with a mean age of 19.81 (10-34 years, SD: 5.36) years with a mean follow-up of 5.5 (2-14) years, entered the study. 24 cases were GMFCS I or II and 18 were III or IV. Supination beyond neutral was seen in 48.5%, and improvement in MACS scores in all the cases. Improved 'grasp' and 'release' and keyboard use was seen in 50%, 71% and 87% respectively. The satisfaction from appearance and improved function of 83-96% correlated positively with GMFSC, MACS and DASH scores. Noticeable improvement in personal hygienic care was seen in only 52% of cases. Tendon transfer in well-selected spastic upper extremity CP cases results in long-time improved function, and limb appearance - correlating with initial GMFCS and MACS scores.

PMID: <u>38189782</u>

2. Systematic Review of Total Hip Arthroplasty Outcomes in Cerebral Palsy Patients and a Comparative Analysis with Rheumatoid Arthritis

Christos Costa, Foteini Moniati, Michalis Chatzimatthaiou, Christos Papaioannou, Sapfo Athanasakopoulou, Marios Chatzimatthaiou

Review Adv Orthop. 2023 Dec 30:2023:8696116. doi: 10.1155/2023/8696116. eCollection 2023.

Background: Total hip arthroplasty (THA) is considered a successful treatment option for patients with degenerative hip arthritis. However, in the setting of neuromuscular diseases, patients with cerebral palsy (CP) are considered high-risk due to instability, contractures, and altered muscle tone. The purpose of this systematic review is to analyse the data in the setting of THA in CP patients including indications, types of implants, revision rates, and patient-reported outcomes and compare these with those of a cohort requiring THA due to degenerative arthritis unrelated to neuromuscular disorders. Methods: PubMed, Embase, and Cochrane Library were searched from inception until June 10, 2023, to identify the relevant studies for THA on CP patients. The methodological quality of the studies was evaluated using the Newcastle-Ottawa Quality Assessment Scale (NOS). Results: The initial search generated 190 studies out of which 21 met the inclusion criteria. The most frequently reported complication was dislocation affecting overall 7.5% (0-28%) of all patients, while other complications included periprosthetic fractures and heterotrophic ossification. The survival rates of primary THAs ranged from 85% to 100% at 5 years and from 73% to 86% at 10 years. Patients with CP who undergo total hip arthroplasty experience a greater overall rate of complications compared to patients with rheumatoid arthritis (RA) that undergo the same procedure.

Conclusion: The current literature suggests that THA is a beneficial procedure for patients with CP through pain reduction and functional improvement. However, the increased rates of potential complications compared to the general population require careful consideration. We suggest that further investigations on the most appropriate time of procedure, implant type, and procedure are needed.

PMID: 38188921

3. Factors associated with gait efficiency in children with cerebral palsy: association between gait abnormality and balance ability

Sosei Ishimoto, Masafumi Itokazu

J Phys Ther Sci. 2024 Jan;36(1):21-25. doi: 10.1589/jpts.36.21. Epub 2024 Jan 1.

[Purpose] Children with cerebral palsy require more gait energy than healthy children. The association between gait abnormalities, balance, and maximum step length to determine contributors to gait efficiency in children with cerebral palsy. [Participants and Methods] The study included 33 patients with cerebral palsy, who could walk without the use of walking aids. All participants were instructed to walk for 6 min, and the Total Heart Beat Index was calculated as a measure of walking efficiency. The Edinburgh Visual Gait Score was used to assess gait abnormalities. Additionally, the maximum step length was recorded, and all participants performed the Berg Balance Scale. Correlation analysis and stepwise multiple regression analysis were used to confirm the association between the aforementioned parameters and the Total Heart Beat Index. [Results] The Edinburgh Visual Gait Score was correlated with the heel lift during the stance, knee position during the terminal swing of gait as factors associated with the Total Heartbeat Index. The Berg Balance Scale was correlated with turning 360°, standing with feet together. [Conclusion] Our findings emphasize the need for treatment strategies focused on gait abnormalities and balance.

PMID: 38186965

4. Multiple Approaches of Neuro-Physiotherapy Used for Improving Balance, Normalizing Tone, and Gait Training in a Child With Ataxic Cerebral Palsy: A Case Report

Dhanashree S Upganlawar, Snehal Samal, Prishita Koul, Jaee P Kapre

Case Reports Cureus. 2023 Dec 10;15(12):e50264. doi: 10.7759/cureus.50264. eCollection 2023 Dec.

Cerebral palsy (CP) is a non-progressive developmental delay disorder that mainly affects children. A strategy for enhancing organizational abilities, including practices based on evidence, and improving outcomes is the base of clinical management in physiotherapy. A seven-year-old girl presented with a history of difficulty walking, standing for extended periods, and performing gross and fine motor movements. MRI revealed generalized atrophy of the cerebellum. The child was managed by medications and physiotherapy. Physiotherapy intervention was based on goal-oriented strategies, which include Rood's approach, constraint-induced movement therapy (CIMT), proprioceptive neuromuscular facilitation (PNF), passive stretching, etc. This goal-oriented program showed an improvement in the treatment outcomes of the child. The child was walking independently with a proper gait pattern and was able to maintain both static and dynamic balance. Initial physical therapy management using integrated methods promotes the achievement of developmental milestones like gross motor skills in ataxic cerebral palsy children.

PMID: 38196428

5. Short-term change of tibial torsion in children with spastic cerebral palsy after selective dorsal rhizotomy

Rui Wang, Wenbin Jiang, Min Wei, Junlu Wang, Xidan Yu, Bo Xiao, Qijia Zhan

Transl Pediatr. 2023 Dec 26;12(12):2131-2141. doi: 10.21037/tp-23-339. Epub 2023 Dec 22.

Background: Spastic cerebral palsy (CP) is a prevalent cause of motor dysfunction in children, with patients often experiencing secondary musculoskeletal deformities, including tibial torsion. This study aimed to investigate the short-term effect of selective dorsal rhizotomy (SDR) on tibial torsion in children with spastic CP. Methods: We conducted a retrospective review of children with spastic CP who underwent SDR at the Department of Neurosurgery, Shanghai Children's Hospital, between July 2019 and November 2022. Pre- and post-operative physical assessments were examined. Results: A total of 148 children were included in the study. After SDR, there was a significant decrease in muscle tone in the lower limb muscle groups. Joint range of motion in the lower limbs also increased post-surgery. Bilateral transmalleolar angle (TMA) showed a significant increase after the surgery, and 21% limbs classified as internal tibial torsion before SDR changed into normal angle post-operatively. Limbs with better improvement after SDR derived from younger patients and had lower muscle tone in the hamstring muscles when compared to those that did not show improvement. Conclusions: SDR has the potential to increase TMA in children with spastic CP. Limbs classified as internal tibial torsion are more likely to improve after SDR if they have

lower muscle tone in the hamstring muscles and are derived from younger patients.

PMID: 38197108

6. A Velocity Stretch Reflex Threshold Based on Muscle-Tendon Unit Peak Acceleration to Detect Possible Occurrences of Spasticity during Gait in Children with Cerebral Palsy

Axel Koussou, Raphaël Dumas, Eric Desailly

Sensors (Basel). 2023 Dec 20;24(1):41. doi: 10.3390/s24010041.

Spasticity might affect gait in children with cerebral palsy. Quantifying its occurrence during locomotion is challenging. One approach is to determine kinematic stretch reflex thresholds, usually on the velocity, during passive assessment and to search for their exceedance during gait. These thresholds are determined through EMG-Onset detection algorithms, which are variable in performance and sensitive to noisy data, and can therefore lack consistency. This study aimed to evaluate the feasibility of determining the velocity stretch reflex threshold from maximal musculotendon acceleration. Eighteen children with CP were recruited and underwent clinical gait analysis and a full instrumented assessment of their soleus, gastrocnemius lateralis, semitendinosus, and rectus femoris spasticity, with EMG, kinematics, and applied forces being measured simultaneously. Using a subject-scaled musculoskeletal model, the acceleration-based stretch reflex velocity thresholds were determined and compared to those based on EMG-Onset determination. Their consistencies according to physiological criteria, i.e., if the timing of the threshold was between the beginning of the stretch and the spastic catch, were evaluated. Finally, two parameters designed to evaluate the occurrence of spasticity during gait, i.e., the proportion of the gait trial time with a gait velocity above the velocity threshold and the number of times the threshold was exceeded, were compared. The proposed method produces velocity stretch reflex thresholds close to the EMG-based ones. For all muscles, no statistical difference was found between the two parameters designed to evaluate the occurrence of spasticity during gait. Contrarily to the EMG-based methods, the proposed method always provides physiologically consistent values, with median electromechanical delays of between 50 and 130 ms. For all subjects, the semitendinosus velocity during gait usually exceeded its stretch reflex threshold, while it was less frequent for the three other muscles. We conclude that a velocity stretch reflex threshold, based on musculotendon acceleration, is a reliable substitute for EMG-based ones.

PMID: 38202903

7. Effect of Exercise and Motor Interventions on Physical Activity and Motor Outcomes of Adults with Cerebral Palsy: A Systematic Review

Danielle Januszyk, Ellen Schafer, Holly J Thompson, Barbara Sargent

Review Dev Neurorehabil. 2023 Aug-Oct;26(6-7):389-412. doi: 10.1080/17518423.2023.2259978. Epub 2024 Jan 6.

Purpose: Systematically review the effect of exercise and motor interventions on physical activity and motor outcomes of adults with cerebral palsy (CP). Methods: Eight databases were searched. Results: Twenty-five studies were included, representing 439 adults with CP. Very low to low quality evidence supports that gait training is more effective than standard care or neurodevelopmental treatment for improving gait velocity, quality of gait, and ankle stiffness; balance training is more effective than seated therapeutic activities for improving walking self-confidence and perceived change in balance; whole-body vibration is no more effective than resistance training for improving strength or gait function; and resistance training is no more effective than a person's typical exercise program for improving strength or gait function of adults with CP. Adverse events were reported for balance training, functional training, resistance training, and whole-body vibration. Discussion: Further research is needed that is adequately powered and uses well-controlled study designs.

PMID: 38183292

8. Can Clinical Assessment of Postural Control Explain Locomotive Body Function, Mobility, Self-Care and Participation in Children with Cerebral Palsy?

Blanka Vlčkova, Jiří Halámka, Markus Müller, Jose Manuel Sanz-Mengibar, Marcela Šafářová

Healthcare (Basel). 2024 Jan 1;12(1):98. doi: 10.3390/healthcare12010098.

Trunk control may influence self-care, mobility, and participation, as well as how children living with cerebral palsy (CP) move around. Mobility and Gross Motor performance are described over environmental factors, while locomotion can be understood as the intrinsic ontogenetic automatic postural function of the central nervous system, and could be the underlying element explaining the relationship between these factors. Our goal is to study the correlation among Trunk Control Measurement Scale (TCMS) and Pediatric Evaluation of Disability Inventory (PEDI) domains, as well as Locomotor Stages (LS). Methods: A feasibility observational analysis was designed including 25 children with CP who were assessed with these

scales. Results: The strong correlation confirms higher levels of trunk control in children with better self-care, mobility and participation capacities. Strong correlations indicate also that higher LS show better levels of PEDI and TCMS domains. Conclusions: Our results suggest that more mature LS require higher levels of trunk control, benefitting self-care, mobility and social functions.

PMID: 38201004

9. Oral rehabilitation of a 14-year-old quadriplegic child with cerebral palsy under general anesthesia: A case report

Mansi Baviskar, Akshaya Mudaliar, Sanjana Ratnakar Kodical, Chinmaya Chaudhary, Parnaja Valke

Case Reports J Family Med Prim Care. 2023 Nov;12(11):2983-2986. doi: 10.4103/jfmpc.jfmpc_2517_22. Epub 2023 Nov 21.

Cerebral palsy is a neuromuscular disorder in which patients deal with dysfunction in motor coordination in addition to other problems like epilepsy. Due to disrupted motor functions, the oromotor coordination is also affected, leading to higher incidence of dental caries, which is also due to increased consumption of medications. This case report deals with the oral rehabilitation of a quadriplegic 14-year-old child with cerebral palsy, suffering from severe dental pain and who was treated under general anesthesia.

PMID: <u>38186788</u>

10. The evolution of nutrition management in children with severe neurological impairment with a focus on cerebral palsy

Stina Oftedal, Siobhan McCormack, Richard Stevenson, Katherine Benfer, Roslyn N Boyd, Kristie Bell

J Hum Nutr Diet. 2024 Jan 9. doi: 10.1111/jhn.13277. Online ahead of print.

Nutritional management of children with severe neurological impairment (SNI) is highly complex, and the profile of this population is changing. The aim of this narrative review was to give the reader a broad description of evolution of the nutritional management of children with SNI in a high resource setting. In the last decade, there has been an emphasis on using multiple anthropometric measures to monitor nutritional status in children with SNI, and several attempts at standardising the approach have been made. Tools such as the Feeding and Nutrition Screening Tool, the Subjective Global Nutrition Assessment, the Eating and Drinking Ability Classification System and the Focus on Early Eating and Drinking Swallowing (FEEDS) toolkit have become available. There has been an increased understanding of how the gut microbiome influences gastrointestinal symptoms common in children with SNI, and the use of fibre in the management of these has received attention. A new diagnosis, 'gastrointestinal dystonia', has been defined. The increased use and acceptance of blended food tube feeds has been a major development in the nutritional management of children with SNI, with reported benefits in managing gastrointestinal symptoms. New interventions to support eating and drinking skill development in children with SNI show promise. In conclusion, as the life expectancy of people with SNI increases due to advances in medical and nutrition care, our approach necessitates a view to long-term health and quality of life. This involves balancing adequate nutrition to support growth, development and well-being while avoiding overnutrition and its associated detrimental long-term effects.

PMID: 38196166

11. Navigating Oral Hygiene Challenges in Spastic Cerebral Palsy Patients: A Narrative Review for Management Strategies for Optimal Dental Care

Sucharitha Palanisamy, Priyanka Cholan, Lakshmi Ramachandran, Anupama Tadepalli, Harinath Parthasarsthy, Santo G Umesh

Review Cureus. 2023 Dec 9;15(12):e50246. doi: 10.7759/cureus.50246. eCollection 2023 Dec.

In the realm of well-being, the essence of maintaining optimal oral health is gaining more recognition. This quantifying quotient is being compromised in cerebral palsy (CP) patients due to multitude variations. Spastic CP predominantly impacts bodily motions, muscle synchronization, command, muscle tone, reflexes, stance, equilibrium, and can additionally influence both delicate and large-scale motor abilities. For individuals with spastic CP, the rigidity extends its influence over both their upper and lower limbs. When this stiffness takes hold in the upper limb, it poses significant challenges in executing everyday activities, causing issues with precise grasping and coordination of muscle movements. Consequently, using a toothbrush effectively becomes a formidable task resulting in widespread caries and periodontal diseases in spastic CP patients. The central focus of this review is to explore the oral health challenges of spastic cerebral palsy patients and mapping out a path towards the most efficient time-tested and innovative dental management approaches for preserving oral health in these patients.

PMID: 38196433

12. Comparison of the prevalence of malocclusion and oral habits between children with cerebral palsy and healthy children

Fuad Lutf Almotareb, Hassan Abdulwahab Al-Shamahy

BMC Oral Health. 2024 Jan 11;24(1):72. doi: 10.1186/s12903-023-03840-z.

Background: Cerebral palsy (CP) represents for children an important problem of health and affects roughly 2 per 1000 live births and is the most common pediatric developmental motor disability. Therefore, the purpose of this study was to determine the prevalence, type and severity of malocclusion and oral habits in children with Cerebral Palsy (CP) and to compare them with a control group of healthy children in Sana'a city. Materials and methods: A prospective, case-control study was made of two groups, a cerebral palsy and a control group. The study population consisted of 60 children who had CP, and a control group of 60 matched children with no physical or mental disabilities. Data were collected using a questionnaire and assessment for malocclusion was done clinically. The patients were compared with equal number of age-matched controls. The inclusion criteria were individuals aged over 6 years; absence of previous orthodontic treatment; no missing permanent first molars. Results: Results showed an increased prevalence of malocclusion in children with cerebral palsy. Molar class II relationship was statistically higher in cerebral palsy children than healthy control (P = 0.001). Cerebral palsied children are likely to have a significantly increased protrusion of the anterior teeth (P < 0.001) when compared with normal children. Mouth breathing and Tongue thrust. Habits were significantly higher in the CP group (p = 0.0001) when compared with normal children. Conclusion: The prevalence of malocclusion was higher in children with Cerebral palsy than in normal children, and the present study concludes that in children with Cerebral Palsy, more oral Habits problems due to oral motor dysfunctions are common and problems of mouth breathing and Tongue thrust produce different malocclusion and poor oral hygiene complications in these children.

PMID: 38212734

13. Relation of Speech-Language Profile and Communication Modality to Participation of Children With Cerebral Palsy

Kristen M Allison, Kayla M Doherty; Cerebral Palsy Research Network

Am J Speech Lang Pathol. 2024 Jan 12:1-11. doi: 10.1044/2023_AJSLP-23-00267. Online ahead of print.

Purpose: This study aimed to examine the contribution of speech motor impairment (SMI), language impairment, and communication modality to communicative and overall participation outcomes in school-age children with cerebral palsy (CP). Method: Eighty-one caregivers of children with CP provided information about their child's speech and language skills, communication modality, and participation through a web-based survey. Caregiver responses to two validated scales were used to quantify children's communicative participation and overall participation. Children were classified into four speech-language profile groups and three communication modality groups for comparison, based on caregiver-reported information regarding their child's communication skills. Results: Children with CP who had co-occurring SMI and language impairment had significantly lower levels of communicative participation and involvement in activities overall, compared to children with SMI alone. Among children with SMI, augmentative and alternative communication (AAC) use was associated with greater overall frequency of participation and involvement in life activities. Conclusion: Children with CP who have both SMI and language impairment and those who are nonspeaking communicators should be prioritized early for communication interventions focused on maximizing participation, including consideration of AAC.

PMID: <u>38215219</u>

14. Assessing the Adequacy of the Physical, Social, and Attitudinal Environment to the Specific Needs of Young Adults With Cerebral Palsy: The European Adult Environment Questionnaire

Célia Perret, Joaquim J M Alvarelhão, Lindsay Pennington, Virginie Ehlinger, Carine Duffaut, Catherine Arnaud, Nicolas Vidart d'Egurbide Bagazgoïtia

Arch Phys Med Rehabil. 2024 Jan 11:S0003-9993(23)00693-7. doi: 10.1016/j.apmr.2023.11.012. Online ahead of print.

Objectives: To present the development of the European Adult Environment Questionnaire (EAEQ), to assess to what extent it covers the International Classification of Functioning, Disability and Health (ICF), and to describe the adequacy of the physical, social, and attitudinal environment to the specific needs of young adults with cerebral palsy (CP). Design: Cross-sectional. Setting: Administrative regions in France, Germany, Italy, Portugal, and Sweden. Participants: Young adults with CP (N=357), with varying severity profiles, aged 19-28 years at time of interview (2018-20). Interventions: Not applicable. Main outcome measure(s): Physical, social, and attitudinal environment unmet needs. Results: Relevant environmental factors (EFs)

for young adults with CP were identified during focus groups in England and Portugal. EFs were mapped to the ICF environmental classification and the EAEQ analytical structure resulted from this linking procedure. It comprised 61 items, linked to 31 ICF environmental classification categories, and covered 4 of its 5 chapters. Content validity assessed with the bandwidth index (percentage coverage of ICF Core Sets for adults with CP) was satisfactory (79.3%). A descriptive analysis was carried out. Participants had a mean age of 24 years, 56% were men, 38% had severely limited mobility. Less than 16% reported unmet needs for EFs relating to home, college/work/day placement, and communication in the Products and technology chapter. Unmet needs were higher (>20%) for the other items in the Public use and Land development categories. Social support, attitudes, and understanding of relatives were often adequate to the participants' needs. The proportion of unmet needs varied by sex (women were more often concerned) and raised with increasing gross motor impairment. Conclusion: The EAEQ describes in detail the adequacy of the environment to the specific needs of young adults with CP. Its ICF-based structure opens up possibilities for use in a universal conceptual framework.

PMID: 38206241

15. Impact of a National Follow-Up Program on the Age at Diagnosis for Cerebral Palsy

Rebecca Alison Fabricius, Mads Langager Larsen, Nanette Mol Debes, Gija Rackauskaite, Christina Engel Hoei-Hansen

Pediatr Neurol. 2023 Nov 30:152:56-61. doi: 10.1016/j.pediatrneurol.2023.11.008. Online ahead of print.

Background: The Danish National Cerebral Palsy Follow-up Program (CPOP) is a nationwide program offering standardized treatment to all children with cerebral palsy (CP) since 2004. We aimed to establish if its implementation had a positive impact on the diagnostic age of CP. Methods: Children with validated CP diagnoses were identified from the Danish Cerebral Palsy Registry and the CPOP. We then compared the age at diagnosis and the clinical features of children with CP born in 2000 to 2003 with those born in 2010 to 2013. Differences in time to diagnosis were compared using log-rank test. Results: The age at diagnosis was not different in the two periods (P = 0.23), with identical overall median diagnostic ages at 13.0 months. The number of children with severe motor disability decreased markedly from 47.5% in 2000 to 2003 to 32.0% in 2010 to 2013 (P < 0.001). There was increased usage of cerebral magnetic resonance imaging; however, this was not associated with lower diagnostic age. Conclusions: The diagnostic age of CP did not change after the implementation of a nationwide follow-up program, offering standardized and early assessments. However, central clinical aspects also changed significantly between the periods compared, which possibly affected the diagnostic age.

PMID: <u>38211417</u>

16. Distinguishing multicystic from focal encephalomalacia on delayed MRI in children with term hypoxic ischemic injury

Dana Alkhulaifat, Shyam Sunder B Venkatakrishna, César Augusto Pinheiro Ferreira Alves, Wondwossen Lerebo, Luis Octavio Tierradentro-Garcia, Mohamed Elsingergy, Fikadu Worede, Jelena Curic, Savvas Andronikou

J Neuroimaging. 2024 Jan 12. doi: 10.1111/jon.13190. Online ahead of print.

Background and purpose: To define cystic patterns resulting from term hypoxic ischemic injury (HII) on delayed Magnetic Resonance Imaging (MRI) and determine associated HII patterns and lesions that reflect the severity of injury, from a database of African children with cerebral palsy. Methods: Retrospective review of 1175 children with cerebral palsy due to term HII diagnosed on late MRI, identifying those with cystic changes. These were classified as multicystic or (multi-) focal-cystic, and were evaluated for associated injuries-thalami, basal ganglia, hippocampi, cerebellum, and presence of ulegyria. Results: Three hundred and eighty-eight of 1175 (33%) children had cystic encephalomalacia. Two hundred and seven of 388 (53.3%) had focal-cystic and 181/388 (46.6%) had multicystic injury. The focal-cystic group comprised 87.9% (182/207) with thalamic injury, 25.6% (53/207) with basal ganglia injury, and 15% (31/207) with cerebellar involvement. Basal-ganglia-thalamus (BGT) pattern was present in 43.9% (91/207) and ulegyria in 69.6% (144/207). In the multicystic group, 88.9% (161/181) had thalamic injury, 30.9% (56/181) had basal ganglia injury, and 21% (38/181) had cerebellar involvement. BGT pattern was observed in 29.8% (54/181) and ulegyria in 28.7%. (52/181). Significant associations (p<.05) were found between multicystic injury and caudate/globus pallidus involvement, and between focal-cystic pattern of injury and ulegyria. Conclusions: Cystic encephalomalacia was seen in almost one-third of patients with term HII imaged with delayed MRI, with a similar prevalence of focal-cystic and multicystic injury. Multicystic injury was associated with caudate and globus pallidi involvement, typical of the BGT pattern of HII, whereas the focal-cystic pattern was associated with ulegyria, typical of watershed injury.

PMID: 38217068

17. Bobath, NeuroDevelopmental Therapy, and clinical science: Rebranding versus rigor

Diane Damiano, Iona Novak

Dev Med Child Neurol. 2024 Jan 12. doi: 10.1111/dmcn.15844. Online ahead of print.

No abstract available

PMID: 38214960

18. Glycopyrronium 320 µg/mL in children and adolescents with severe sialorrhoea and neurodisabilities: A randomized, double-blind, placebo-controlled trial

Pierre Fayoux, Mickael Dinomais, Helen Shaw, Frédéric Villain, Déborah Schwartz, Stéphane Rondeau, Guy Letellier, Stéphane Auvin

Dev Med Child Neurol. 2024 Jan 12. doi: 10.1111/dmcn.15841. Online ahead of print.

Aim: To investigate the efficacy, safety, and impact on quality of life (QoL) of an oral formulation of 320 µg/mL glycopyrronium designed for children. Method: A double-blind, placebo-controlled SALIVA (Sialanar plus orAl rehabiLitation against placebo plus oral rehabilitation for chIldren and adolescents with seVere sialorrhoeA and neurodisabilities) trial was conducted. Children (3-17 years) with neurodisabilities and severe sialorrhoea (modified Teachers Drooling Scale \geq 6) were randomized to 320 µg/mL glycopyrronium or placebo, in addition to non-pharmacological standard care. Results: Of 87 participants, 44 were aged 10 years or under and 43 had cerebral palsy. The primary endpoint, change in total Drooling Impact Scale (DIS) score from baseline to day 84, was significantly greater (improved) with 320 µg/mL glycopyrronium versus placebo (median [quartile 1, quartile 3] -29.5 [-44.5, 0] vs -1 [-16, 5]; p < 0.001), an effect also observed at day 28 (median - 25 vs -2; p < 0.01). Significant reduction in bibs/clothes used per day was seen with glycopyrronium versus placebo at day 84 (median - 2 vs 0; p < 0.01). Glycopyrronium significantly improved DIS items 9 and 10 related to the extent that drooling affects the child's and family's life (p ≤ 0.03). Adverse events were reported by 77.3% and 69.8% of children with glycopyrronium and placebo respectively; the most common treatment-related adverse event was constipation (20.5% and 16.3%). Interpretation: The formulation of 320 µg/mL glycopyrronium significantly improved drooling and reduced its impact on QoL, with good tolerability in children with neurodisabilities.

PMID: 38214675

19. Neurodevelopmental Treatment in Children With Cerebral Palsy: A Review of the Literature

Sandeep Khanna, Ranganathan Arunmozhi, Chanan Goyal

Review Cureus. 2023 Dec 12;15(12):e50389. doi: 10.7759/cureus.50389. eCollection 2023 Dec.

This review aimed to explore the current literature on neurodevelopmental treatment (NDT) in children with cerebral palsy (CP). It also sought to determine what outcome measures are used to analyze the effect of NDT and whether these parameters are in line with the components of the International Classification of Functioning, Disability and Health (ICF). The studies published in the English language between 2000 and 2023 were included based on a search of the databases PEDro, PubMed, and Google Scholar. Studies that examined the effect of NDT on children with CP were included. We found a total of 54 studies describing the effect of NDT in children with CP and these were included in this literature review. NDT in children with CP was found to have positive outcomes in 41 studies, while 13 studies had contradictory conclusions. Based on our findings, NDT is widely used for the rehabilitation of children with CP globally. The parameters used to assess the improvement mostly included gross motor function, balance, and postural control. The outcome measures used in studies are usually linked to body structure and function or activities domain of the ICF model by the World Health Organization (WHO). However, there is a scarcity of studies on the effect of NDT on participation, which should be the outcome of any rehabilitation program. There is scope for future research to demonstrate the effect of NDT on the participation of children with CP. Further studies with larger sample sizes and homogenous groups are recommended.

PMID: <u>38213384</u>

20. Intrathecal Baclofen Infusion-Botulinum Toxin Combined Treatment Efficacy in the Management of Spasticity due to Cerebral Palsy

Riccardo Marvulli, Giuseppa Lagioia, Giancarlo Ianieri, Lucrezia Dell'Olio, Alessandra Zonno, Mariagrazia Riccardi, Rosa Bianca Sinisi, Laura Belinda Rizzo, Giacomo Farì, Marisa Megna, Maurizio Ranieri

CNS Neurol Disord Drug Targets. 2024 Jan 11. doi: 10.2174/0118715273250973230919121808. Online ahead of print.

Background: Cerebral Palsy (CP) is a group of permanent, but not unchanging, disorders of movement and/or posture and motor function, which are due to a non-progressive interference, lesion, or abnormality of the developing/immature brain. One clinical presentation is muscle spasticity, which leads to a significant impact on the individual's functionality and quality of life.

Spasticity treatment is multidisciplinary and includes pharmacological and physical intervention; intrathecal baclofen shows a positive effect in severe spasticity and suboptimal response to oral drugs, while local injection of Botulinum toxin type A (BTXA) improves muscle tone, motion and pain. Objective: The aim of this study was to evaluate the efficacy of the combined intrathecal baclofen infusion (ITB) - botulinum toxin treatment in the management of spasticity in CP. Methods: 8 patients with spastic tetraparesis were enrolled. All patients were treated with intrathecal Baclofen; in lower limbs, no spastic symptoms appeared, while marked spasticity was noted in upper limbs. We injected the right and left Biceps Brachial (BB) and Flexor Digitorum Superficialis (FDS) muscles with botulinum toxin type A. All patients underwent Myometric measurement, Ashworth Scale, Numerical Rating Scale, and Visual Analogic Scale evaluation before infiltration (T0), 30 days after injection (T1), 60 days after injection (T2), and 90 days after treatment (T3). Results: All data demonstrated an improvement in spasticity, pain, quality of life, and self-care during the study, with p < 0.05. No side effects appeared. Conclusion: This study demonstrated the efficacy and safety of intrathecal baclofen infusion and botulinum toxin combined treatment in the management of spasticity, pain, quality of life, and selfcare in CP patients.

PMID: 38213169

21. Delivery room intubation and neurodevelopment among extremely preterm infants

Kei Tamai, Naomi Matsumoto, Takashi Yorifuji, Akihito Takeuchi, Makoto Nakamura, Kazue Nakamura, Misao Kageyama; Neonatal Research Network of Japan

Pediatr Res. 2024 Jan 11. doi: 10.1038/s41390-023-02993-5. Online ahead of print.

Background: The impact of delivery room intubation (DRI) on neurodevelopment in extremely preterm infants remains unclear. Methods: We retrospectively analyzed data for infants born at 24-27 gestational weeks between 2003 and 2018. The primary outcome was neurodevelopmental impairment (NDI), defined as cerebral palsy or hearing, visual, or cognitive impairment at age 3 years. Secondary outcomes were NDI components and death before and after discharge from the neonatal intensive care unit. We conducted robust Poisson regression analyses, adjusting for perinatal confounders. Results: The full cohort included 4397 infants with NDI data, of whom 3703 were intubated in the delivery room and 694 were not intubated in the delivery room. The mean gestational age and birth weight were 26.0 ± 1.1 weeks and 778 ± 184 g for infants with DRI and 26.6 ± 1.0 weeks and 873 ± 184 g for infants without DRI. Compared with infants without DRI, those with DRI had a higher risk for NDI (32.4% vs. 23.3%; adjusted risk ratio 1.18, 95% confidence interval: 1.01-1.37). There were no differences in secondary outcomes between infants with and without DRI. Conclusions: DRI was associated with an increased risk for NDI at age 3 years among extremely preterm infants. Impact: Few studies have examined the impact of delivery room intubation on neurodevelopment in infants born extremely preterm, and the results have been inconsistent. A total of 4397 infants born at 24-27 gestational weeks who had neurodevelopmental data at age 3 years were included in the present study. The present study found that delivery room intubation was associated with an increased risk for neurodevelopmental impairment at age 3 years among extremely preterm infants.

PMID: 38212388

22. The causal association between maternal depression, anxiety, and infection in pregnancy and neurodevelopmental disorders among 410 461 children: a population study using quasi-negative control cohorts and sibling analysis

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Psychol Med. 2024 Jan 11:1-9. doi: 10.1017/S0033291723003604. Online ahead of print.

Background: To address if the long-standing association between maternal infection, depression/anxiety in pregnancy, and offspring neurodevelopmental disorder (NDD) is causal, we conducted two negative-control studies. Methods: Four primary care cohorts of UK children (pregnancy, 1 and 2 years prior to pregnancy, and siblings) born between 1 January 1990 and 31 December 2017 were constructed. NDD included autism/autism spectrum disorder, attention-deficit/hyperactivity disorder, intellectual disability, cerebral palsy, and epilepsy. Maternal exposures included depression/anxiety and/or infection. Maternal (age, smoking status, comorbidities, body mass index, NDD); child (gender, ethnicity, birth year); and area-level (region and level of deprivation) confounders were captured. The NDD incidence rate among (1) children exposed during or outside of pregnancy and (2) siblings discordant for exposure in pregnancy was compared using Cox-regression models, unadjusted and adjusted for confounders. Results: The analysis included 410 461 children of 297 426 mothers and 2 793 018 person-years of follow-up with 8900 NDD cases (incidence rate = 3.2/1000 person years). After adjustments, depression and anxiety consistently associated with NDD (pregnancy-adjusted HR = 1.58, 95% CI 1.46-1.72; 1-year adj. HR = 1.49, 95% CI 1.39-1.60; 2-year adj. HR = 1.62, 95% CI 1.50-1.74); and to a lesser extent, of infection (pregnancy adj. HR = 1.16, 95% CI 1.10-1.22; 1-year adj. HR = 1.20, 95% CI 1.14-1.27; 2-year adj. HR = 1.19, 95% CI 1.12-1.25). NDD risk did not differ among siblings discordant for pregnancy exposure to mental illness HR = 0.97, 95% CI 0.77-1.21 or infection HR = 0.99, 95% CI 0.90-1.08. Conclusions: Maternal risk appears to be unspecific to pregnancy: our study provided no evidence of a specific, and therefore causal, link between in-utero exposure to infection, common mental illness, and later development of NDD.

23. Co-creation: Pioneering progress in cerebral palsy research

Rachel Byrne, Bronya Metherall

Dev Med Child Neurol. 2024 Jan 10. doi: 10.1111/dmcn.15828. Online ahead of print.

No abstract available

PMID: <u>38204327</u>

24. Evaluation of a Modified Mindfulness-Based Stress Reduction Intervention for Adults with Cerebral Palsy and Anxiety and/or Emotion Regulation Difficulties-A Randomised Control Trial

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J Clin Med. 2023 Dec 19;13(1):1. doi: 10.3390/jcm13010001.

Mindfulness-Based Stress Reduction (MBSR) has not yet been evaluated for people with cerebral palsy (CP). The aims of this randomised control trial were to investigate whether a modified telehealth MBSR program could improve mindfulness and reduce depression, anxiety, and emotion regulation difficulties among adults with CP with elevated anxiety and/or emotional regulation difficulties. Participants (n = 31) with elevated anxiety and/or emotion regulation difficulties and no/mild intellectual impairment were randomised to a modified telehealth MBSR program (90 min weekly, 9 weeks) group or a wait-list group. Measurements were collected prior to (T1), after (T2), and 8 weeks post-intervention (T3). The primary outcome was the mean between-group difference in the change in Cognitive and Affective Mindfulness Scale-R (CAMS-R) scores in T1-T2. The secondary outcomes included mean within-group differences over time for the CAMS-R total scores, Depression Anxiety and Stress Scale-21 subscales, and Difficulties in Emotion Regulation Scale (DERS) total t-score. We found no statistically significant between-group difference in mean change in mindfulness scores for T1-T2 and T1-T3; improved mean scores for Depression and Stress subscales for T1-T2; and improved DERS t-scores for T1-T2 and T1-T3. In conclusion, this study found no significant between-group difference for the primary outcome of mindfulness. The MBSR program was successfully modified for adults with CP and was effective in improving depression, stress, and emotion regulation. ACTRN12621000960853.

PMID: 38202008

25. Adaptive Intervention to Prevent Respiratory Illness in Cerebral Palsy: Protocol for a Feasibility Pilot Randomized Controlled Trial

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JMIR Res Protoc. 2024 Jan 8:13:e49705. doi: 10.2196/49705.

Background: This study will pilot-test an innovative just-in-time adaptive intervention to reduce severe respiratory illness among children with severe cerebral palsy (CP). Our intervention program, Respiratory Exacerbation-Plans for Action and Care Transitions (RE-PACT), delivers timely customized action planning and rapid clinical response when hospitalization risk is elevated. Objective: This study aims to establish RE-PACT's feasibility, acceptability, and fidelity in up to 90 children with severe CP. An additional aim is to preliminarily estimate RE-PACT's effect size. Methods: The study will recruit up to 90 caregivers of children with severe CP aged 0 to 17 years who are cared for by a respiratory specialist or are receiving daily respiratory treatments. Participants will be recruited from pediatric complex care programs at the University of Wisconsin-Madison (UW) and the University of California, Los Angeles (UCLA). Study participants will be randomly assigned to receive usual care through the complex care clinical program at UW or UCLA or the study intervention, RE-PACT. The intervention involves action planning, rapid clinical response to prevent and manage respiratory illness, and weekly SMS text messaging surveillance of caregiver confidence for their child to avoid hospitalization. RE-PACT will be run through 3 successively larger 6-month trial waves, allowing ongoing protocol refinement according to prespecified definitions of success for measures of feasibility, acceptability, and fidelity. The feasibility measures include recruitment and intervention time. The acceptability measures include recruitment and completion rates as well as intervention satisfaction. The fidelity measures include observed versus expected rates of intervention and data collection activities. The primary clinical outcome is a severe respiratory illness, defined as a respiratory diagnosis requiring hospitalization. The secondary clinical outcomes include hospital days and emergency department visits, systemic steroid courses, systemic antibiotic courses, and death from severe respiratory illness. Results: The recruitment of the first wave began on April 27, 2022. To date, we have enrolled 30 (33%) out of 90 participants, as projected. The final wave of recruitment will end by October 31, 2023, and the final participant will complete the study by April 30, 2024. We will start analyzing the complete responses by April 30, 2024, and the publication of results is expected at

the end of 2024. Conclusions: This pilot intervention, using adaptive just-in-time strategies, represents a novel approach to reducing the incidence of significant respiratory illness for children with severe CP. This protocol may be helpful to other researchers and health care providers caring for patients at high risk for acute severe illness exacerbations. Trial registration: ClinicalTrials.gov/NCT05292365; https://clinicaltrials.gov/study/NCT05292365.

PMID: 38190242

26. Effects of video game-based therapy in an adolescent with cerebral palsy: A case report

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Case Reports World J Clin Cases. 2023 Dec 26;11(36):8595-8602. doi: 10.12998/wjcc.v11.i36.8595.

Background: Herein, we report the case of a 13-year-old boy with spastic quadriplegia cerebral palsy (CP) at Gross Motor Function Classification System (GMFCS) level II, engaging in a 6-wk video game-based therapy (VBT) program. This study aimed to offer essential insights regarding VBT's impact on enhancing the physical function and improving the quality of life (QoL) of adolescents diagnosed with CP. This report provides a distinctive viewpoint that can inform and direct future clinical practices and research endeavors. Case summary: The boy presented with moderate mobility, balance, and overall well-being. He faced challenges with diminished lower limb strength, which affected his daily living and physical fitness capabilities. Our participant was diagnosed with spastic quadriplegic CP at GMFCS level II. He participated in a 6-wk program of VBT using a play station. This innovative approach incorporates warm-up exercises, interactive activities, and cool-down routines, targeting various movements, including single-leg stance, weight shifting, kicking, jumping, marching, and squatting. After VBT, the strength of the left hip extensor significantly increased from 199.3 N to 541.3 N. Distance covered as part of a 6-min walk test increased by 82 m. His Paediatric QoL Inventory score increased dramatically by 25.9%. Conclusion: VBT is an innovative, individualized therapy that enhances physical function and QoL in CP, emphasizing its role in ambulatory patients.

PMID: 38188206

27. Corrected Age at Bayley Assessment and Developmental Delay in Extreme Preterms

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Pediatrics. 2024 Jan 8:e2023063654. doi: 10.1542/peds.2023-063654. Online ahead of print.

Background and objectives: Research on outcomes of prematurity frequently examines neurodevelopment in the toddler years as an end point, but the age range at examination varies. We aimed to evaluate whether the corrected age (CA) at Bayley-III assessment is associated with rates of developmental delay in extremely preterm children. Methods: This retrospective cohort study included children born at <29 weeks' gestation who were admitted in the Canadian Neonatal Network between 2009 and 2017. The primary outcomes were significant developmental delay (Bayley-III score <70 in any domain) and developmental delay (Bayley-III score <85 in any domain). To assess the association between CA at Bayley-III assessment and developmental delay, we compared outcomes between 2 groups of children: those assessed at 18 to 20 months' CA and 21-24 months. Results: Overall, 3944 infants were assessed at 18-20 months' CA and 881 at 21-24 months. Compared with infants assessed at 18-20 months, those assessed at 21-24 months had higher odds of significant development delay (20.0% vs 12.5%; adjusted odds ratio, 1.75; 95% confidence interval [CI], 1.41-2.13) and development delays (48.9% vs 41.7%, adjusted odds ratio 1.33; 95% CI, 1.11-1.52). Bayley-III composite scores were on average 3 to 4 points lower in infants evaluated at 21-24 months' CA (for instance, adjusted mean difference and 95% CI for language: 3.49 [2.33-4.66]). Conversely, rates of cerebral palsy were comparable (4.6% vs 4.7%) between the groups. Conclusions: Bayley-III assessments performed at 21-24 months' CA were more likely to diagnose a significant developmental delay compared with 18- to 20-month assessments in extremely preterm children.

PMID: 38186292