Cerebral palsy research news

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Interventions and Management

1. Effect of positioning error on the Hilgenreiner epiphyseal angle and the head-shaft angle compared to the femoral neck-shaft angle in children with cerebral palsy
Emily S Sullivan, Carly Jones, Stacey D Miller, Kyoung Min Lee, Moon Seok Park, David R Wilson, Kishore Mulpuri, Agnes G d'Entremont

J Pediatr Orthop B. 2021 Oct 29. doi: 10.1097/BPB.0000000000000923. Online ahead of print.

Children with cerebral palsy (CP) often have changes in proximal femoral geometry. Neck-shaft angle (NSA), Hilgenreiner epiphyseal angle (HEA) and head-shaft angle (HSA) are used to measure these changes. The impact of femoral rotation on HEA/HSA and of ab/adduction on HEA/HSA/NSA is not well known. This study aimed to determine and compare the effect of rotation, ab/adduction and flexion/extension on HEA/HSA/NSA. Radiographic measurements from 384 patients with Gross Motor Function Classification System (GMFCS) levels I-V were utilized. NSA/HSA for affected hips were used with femoral anteversion averages to create three-dimensional models of 694 hips in children with CP. Each hip was rotated, ab/adducted and flexed/extended to simulate malpositioning. HEA/HSA/NSA of each model were measured in each joint position, and differences from correct positioning were determined. Mean HEA error at 20° of internal/external rotations were -0.60°/3.17°, respectively, with the NSA error of -6.56°/9.94° and the HSA error of -3.69°/1.21°. Each degree of ab/adduction added 1° of the HEA error, with no NSA/HSA error. NSA was most sensitive to flexion. Error for all measures increased with increasing GMFCS level. HEA/HSA were minimally impacted by rotation. NSA error was much higher than HEA/HSA in internal rotation and flexion whereas HEA was sensitive to changes in ab/adduction. Given abduction is more easily detectable on imaging than rotation, HEA may be less affected by positioning errors that are common with children with CP than NSA. HSA was least affected by position changes. HEA/HSA could be robust, complementary measures of hip deformities in children with CP.

PMID: 34723914

2. A modified surface EMG biomarker for gait assessment in spastic cerebral palsy
Maria Vinti, Manob Jyoti Saikia, John Donoghue, Kunal Mankodiya, Karen L Kerman

Hum Mov Sci. 2021 Nov 1;80:102875. doi: 10.1016/j.humov.2021.102875. Online ahead of print.

Objective: Muscle clinical metrics are crucial for spastic cocontraction management in children with Cerebral Palsy (CP). We investigated whether the ankle plantar flexors cocontraction index (CCI) normalized with respect to the bipedal heel rise (BHR) approach provides more robust spastic cocontraction estimates during gait than those obtained through the widely accepted standard maximal isometric plantar flexion (IPF). Methods: Ten control and 10 CP children with equinus gait pattern performed the BHR and IPF testing and walked barefoot 10-m distance. We compared agonist medial gastrocnemius EMG

during both testing and CCIs obtained as the ratios of antagonist EMG during swing phase of gait and either BHR or IPF agonist EMG. Results: Agonist EMG values from the BHR were: (i) internally reliable (Cronbach's $\alpha = 0.993$), (ii) ~50 ± 0.4% larger than IPF, (iii) and positively correlated. Derived CCIs were significantly smaller ($p < 0.05$) in both populations. Conclusion: The bipedal heel rise approach may be accurate enough to reveal greater agonist activity of plantar flexors than the maximal isometric plantar flexion and seems to be more appropriate to obtain cocontraction estimates during swing of gait. Significance: This modified biomarker may represent a step forward towards improved accuracy of spastic gait management in pediatric.

PMID: 34736019

3. Relationship between ankle function and walking ability for children and young adults with cerebral palsy: A systematic review of deficits and targeted interventions
Benjamin C Conner, Nushka M Remec, Cassidy M Michaels, Chase W Wallace, Emily Andrisevic, Zachary F Lerner

Review Gait Posture. 2021 Oct 25;91:165-178. doi: 10.1016/j.gaitpost.2021.10.024. Online ahead of print.

Background: A primary goal of treatment for children with cerebral palsy is improved walking ability to allow for a more active and independent lifestyle. With the importance of ankle function to walking ability, and the deficits in ankle function associated with cerebral palsy, there is good rationale for targeting this joint in an effort to improve walking ability for this population. Research question: How do deficits and targeted interventions of the ankle joint influence walking ability in children with cerebral palsy? Methods: A specific search criteria was used to identify articles that either (1) provided information on the relationship between ankle function and walking ability or (2) investigated the effect of a targeted ankle intervention on walking ability in cerebral palsy. PubMed, Embase, CINAHL, and Web of Science databases were searched from 1980-April, 2020. Resulting citations were compared against a prospective set of inclusion and exclusion criteria. Data relevant to the original research question was extracted, and the level of evidence for each intervention study was scored. Interpretation was focused on specific, pre-determined mobility measures. Results: Sixty-one citations met all criteria for data extraction, six of which were observational, and fifty-five of which were interventional. Level of evidence ranged from 2 to 4. Self-selected walking speed was the most common measure of walking ability, while physical activity level was the least common. Significance: Ankle function is an important contributor to the walking ability of children with cerebral palsy, and most interventions targeting the ankle seem to demonstrate a benefit on walking ability, but future higher-powered and/or controlled studies are necessary to confirm these findings.

PMID: 34736095

4. Use of lower extremity orthoses in patients with cerebral palsy and related factors
Mazlum Serdar Akaltun, Ozlem Altindag, Sükrü Bicer, Neytullah Turan, Savas Gursoy, Ali Gur

Prosthet Orthot Int. 2021 Nov 2. doi: 10.1097/PXR.0000000000000049. Online ahead of print.

Background: The purpose of this study was to investigate the frequency of using lower extremity orthosis prescribed for patients with cerebral palsy (CP) and to determine the factors associated with orthosis use. Study design: The study had a retrospective and descriptive design. Methods: One hundred and twenty-nine patients with CP who were prescribed orthoses were included in this study. The sociodemographic data and orthosis use status of patients were questioned with telephone calls. Motor functions were evaluated with Gross Motor Function Classification System (GMFCS). Spasticity was evaluated with the modified Ashworth scale, and range of motion was evaluated by goniometry. The study had a retrospective and descriptive design and included 129 patients with CP (53 females and 76 males) who were prescribed orthosis. The sociodemographic data and orthosis use status of patients were questioned with telephone calls. Motor functions were evaluated with Gross Motor Function Classification System (GMFCS). Spasticity was evaluated with the modified Ashworth scale, and range of motion was evaluated by goniometry. Results: A total of 53 patients who were included in the study were female, and 76 were male. The most commonly used orthosis was Ankle-Foot Orthosis. The number of patients who used orthosis every day was 45 (34.8%). The most common reason for not using orthosis was difficulty in wearing it. The mean age was significantly lower in the group that used orthosis regularly ($P < 0.05$). Spasticity and range of motion limitations were low in the group that used orthosis regularly at significant levels ($P < 0.05$). A significant relation was detected in logistic regression analysis between age and orthosis use ($P < 0.05$). Conclusion: Regular orthosis use is at quite low levels in patients with CP. The decision for prescribing orthosis should be made with a rehabilitation team, with the patient included before orthosis is prescribed. Problems related to orthosis use should be addressed at regular intervals after prescribing orthosis.

PMID: 34731872

5. Prefabricated ankle-foot orthoses for children with cerebral palsy to overcome spastic drop-foot: does orthotic ankle stiffness matter?
Harald Böhm, Chakravarthy U Dussa

Prosthet Orthot Int. 2021 Oct 28. doi: 10.1097/PXR.0000000000000050. Online ahead of print

Background: Spastic drop-foot is a common problem in children with cerebral palsy that may lead to tripping and falling. To improve ankle dorsiflexion in swing phase, prefabricated carbon-composite ankle-foot orthoses are commonly prescribed; by increasing ankle stiffness, these orthoses may also improve knee extension in stance. Objectives: To compare the effect of a stiff vs. flexible prefabricated ankle-foot orthosis on sagittal plane ankle and knee kinematics and kinetics during walking. Study design: Cross-sectional, repeated-measures, interventional study. Methods: Twenty-seven children and adolescents with cerebral palsy who had drop-foot in swing were included. Gait analysis was conducted under four conditions: barefoot, shod, with a stiff, and with a flexible orthosis. Participants were divided into two groups including children and adolescents who have a flexed knee during stance (KF, N = 12) and without flexed knee during stance (KE, N = 15). Results: Ankle dorsiflexion in swing phase was significantly improved compared with the shod condition by 6.3 degrees (SD = 3.3 degrees) only in the KE group when using the flexible orthosis. For the stiff orthosis, knee extension in stance was significantly increased by 2.4 degrees (SD = 3.3 degrees) in the KE group compared with the shod condition. No significant improvements were observed for the KF group. Further analysis indicated that only seven patients in the KF group with weak ankle plantarflexors improved knee extension while using the stiff orthosis. Conclusions: Our results suggested that in the KE group, the flexible orthosis was best suited for patients with drop-foot without a knee extension deficit. The stiff orthosis was not suitable in this group as it caused a hyperextended knee without improving dorsiflexion in swing phase. Therefore, stiffness should be considered when prefabricated orthoses are prescribed.

PMID: 34723908

6. Factors that Influence Acquisition of Lower Extremity Braces in the Pediatric Orthopaedic Population
Anna Rambo, Leslie Rhodes, Justin Lomax, Xueyuan Cao, Jack Steele, Karen Romer, David Spence, Benjamin W Sheffer, William C Warner, Jeffrey R Sawyer, Derek M Kelly

J Pediatr Orthop. 2021 Nov 1. doi: 10.1097/BPO.0000000000001998. Online ahead of print.

Background: Lower extremity brace-wear compliance has been studied in pediatrics, but failure to acquire a prescribed brace has not been included. The purpose of this study was to evaluate brace acquisition as a component of brace-wear compliance. Methods: Records of patients (0 to 21 y) prescribed lower extremity braces from 2017 to 2019 were reviewed. Diagnoses included cerebral palsy, spina bifida, short Achilles tendon, clubfoot, and other. Brace type was categorized as clubfoot foot abduction orthosis, ankle-foot orthosis, knee, hip, or custom/other braces. Brace prescription and acquisition dates were recorded. Insurance was classified as government, private, or uninsured. Patient demographics included age, sex, race, and calculated area deprivation index. Results: Of the 1176 prescribed lower extremity braces, 1094 (93%) were acquired while 82 (7%) were not. The odds ratios (OR) of failure to acquire a prescribed brace in Black and Hispanic patients were 1.64 and 2.71 times that in White patients, respectively (95% confidence interval: 1.01-2.71, P=0.045; 1.23-5.6, P=0.015); in patients without insurance, the OR was 8.48 times that in privately insured patients (95% confidence interval: 1.93-31.1, P=0.007). The ORs of failure to acquire were 2.12 (P=0.003) in patients 4 years or more versus 0 to 3 years, 4.17 (P<0.0001) in cerebral palsy versus clubfoot, and 4.12 (P=0.01) in short Achilles tendon versus clubfoot. There was no significant association between sex or area deprivation index and failure of brace acquisition. Conclusions: In our cohort, 7% of prescribed braces were not acquired. Black or Hispanic race, lack of insurance, and older age were associated with failure to acquire prescribed braces. Braces prescribed for clubfoot were acquired more often than for cerebral palsy or short Achilles tendon. Brace-wear compliance is an established factor in treatment success and recurrence. This study identified risk factors for failed brace acquisition, a critical step for improving compliance. These results may help effect changes in the current system that may lead to more compliance with brace wear. Level of evidence: Level III-retrospective cohort study.

PMID: 34723893

7. The effect of motor imagery training on individuals with unilateral cerebral palsy on motor imagery ability, functional mobility and muscle activity
Demet Gözaçan Karabulut, Eylem Tütün Yümin, Yusuf Öztürk

Somatosens Mot Res. 2021 Nov 3;1-8. doi: 10.1080/08990220.2021.1997983. Online ahead of print.

Aim: This study aims to examine the effect of motor imagery (MI) training on MI abilities, functional mobility, and lower extremity muscle activity in children with unilateral cerebral palsy (UCP). Method: 34 UCP and 17 typically developing participants were included. UCP was randomised into 2 groups as UCP MI and UCP control. Participants typically developing were included for baseline comparisons. UCP MI group received 8 weeks of physiotherapy and MI training, the UCP control group 8 weeks of physiotherapy training. The MI abilities, functional mobility, and lower extremity muscle activation were assessed in all groups. Results: It was found that MI training made a significant difference in favour of the UCP MI group in terms of Movement Imagery Questionnaire-For Children (MIQ-C), mental chronometry, functional mobility, and resting muscle activation ($p < 0.05$). There was no such significant change in the UCP control group. Conclusion: This current approach in UCP is a feasible method, beneficial to include it in the rehabilitation process.

PMID: 34732094

8. Oral Motor Treatment Efficacy: Feeding and Swallowing Skills in Children with Cerebral Palsy
Maria E Widman-Valencia, Luis F Gongora-Meza, Héctor Rubio-Zapata, Rita E Zapata-Vázquez, Elma Vega Lizama, Marco Ramírez Salomón, Damaris Estrella-Castillo

Behav Neurol. 2021 Oct 25;2021:6299462. doi: 10.1155/2021/6299462. eCollection 2021.

This study is aimed at identifying the relationship between oral motor treatment and the improvement of abilities for feeding and swallowing in boys and girls with CP residing in the state of Yucatán. The sample consisted of 30 patients with a diagnosis of CP and the presence of ADT, with gross motor function levels from II to V, between 3 and 14 years old, of which 50% received oral motor treatment. The predominant diagnosis was spastic CP and tetraplegia. An interview was carried out with the tutor, the application of the gross motor skills scale, and an assessment of feeding skills. The feeding and swallowing skills that improved significantly with the oral motor treatment were mandibular mobility, tongue activity, abnormal reflexes, control of breathing, and general oral motor skills ($p \leq 0.05$). Within the sample that did not receive oral motor treatment, 46% presented low or very low weight and 40% referred recurrent respiratory diseases. In the end, it was concluded that feeding skills improve significantly with oral motor treatment, regardless of the severity of gross motor involvement. Likewise, oral motor treatment was associated with a lower presence of respiratory diseases and nutritional compromise.

PMID: 34733374

9. Classification of pain in children with cerebral palsy
Michael N Vinkel, Gija Rackauskaite, Nanna B Finnerup

Review Dev Med Child Neurol. 2021 Nov 2. doi: 10.1111/dmcn.15102. Online ahead of print.

Pain in patients with cerebral palsy (CP) is a major health issue strongly associated with reduced quality of life. In this study, we provide an overview of pain conditions in children with CP using the International Classification of Diseases, 11th Revision (ICD-11), which has been updated with a classification of chronic pain. Common causes of pain in children with CP, including hip displacement, muscle spasms, and procedures, are discussed; less studied pain types including headaches, neuropathic pain, visceral pain, and acute versus chronic pain are also highlighted. The addition of chronic pain to the ICD-11 is an important step forward in optimizing both the registration and assessment of pain conditions. However, a tool designed specifically for the different types of pain in patients with CP is imperative. In this paper, we propose a Cerebral Palsy Pain Classification that is aligned with the underlying mechanisms of pain and the ICD-11 pain classification.

PMID: 34726259

10. Epilepsy and related challenges in children with COL4A1 and COL4A2 mutations: A Gould syndrome patient registry
Danielle Boyce, Sheena McGee, Lisa Shank, Sheel Pathak, Douglas Gould

Epilepsy Behav. 2021 Nov 1;125:108365. doi: 10.1016/j.yebeh.2021.108365. Online ahead of print.

Recently, patient advocacy groups started using the name Gould syndrome to describe clinical features of COL4A1 and COL4A2 mutations. Gould syndrome is increasingly identified in genetic screening panels, and because it is a rare disease, there is a disproportionate burden on families to understand the disease and chart the course for clinical care. Among the chief concerns for caregivers of children with Gould syndrome are the challenges faced because of epilepsy, including severe manifestations such as infantile spasms. To document the concerns of the patient population, the Gould Syndrome Foundation established the Gould Syndrome Global Registry (GSGR). Methods: The Gould Syndrome Foundation developed questions for the GSGR with iterative input from patients and caregivers. An institutional review board issued an exemption determination before data collection began. Participants were recruited through social media and clinician referrals. All participants consented electronically, and the data were collected and managed using REDCap electronic data capture tools. De-identified data representing responses received between October 2019 and February 2021 were exported and analyzed with IBM SPSS 27 using descriptive statistics (mean, standard deviation, frequency, range, and percent). Results: Seventy families from twelve countries provided data for the registry, representing 100 affected people (40 adults and 60 children). This analysis represents a subanalysis of the 35 out of 60 children <=18 years of age who reported a history of seizures. Nearly half of these participants were diagnosed with infantile spasms. Participants with epilepsy frequently reported developmental delays (88.6%), stroke (60.0%), cerebral palsy (65.7%), and constipation (57.1%). Ten (28.6%) children use a feeding tube. Despite the fact that more than half of respondents reported stroke, only 34.3% reported ever receiving education on stroke recognition. Conclusion: Here we describe the development and deployment of the first global registry for individuals and family members with Gould syndrome, caused by mutations in COL4A1 and COL4A2. It is important for pediatric neurologists to have access to resources to provide families upon diagnosis. Specifically, all families with Gould Syndrome must have access to infantile spasms awareness and stroke education materials. The Gould Syndrome Foundation is planning several improvements to this patient registry which will encourage collaboration and innovation for the benefit of people living with Gould syndrome.

PMID: 34735964

11. The international Perinatal Outcomes in the Pandemic (iPOP) study: protocol
Sarah J Stock, Helga Zoega, Meredith Brockway, Rachel H Mulholland, Jessica E Miller, Jasper V Been, Rachael Wood, Ishaya I Abok, Belal Alshaikh, Adejumoke I Ayede, Fabiana Bacchini, Zulfiqar A Bhutta, Bronwyn K Brew, Jeffrey Brook, Clara Calvert, Marsha Campbell-Yeo, Deborah Chan, James Chirombo, Kristin L Connor, Mandy Daly, Kristjana Einarsdóttir, Ilaria Fantasia, Meredith Franklin, Abigail Fraser, Siri Eldevik Håberg, Lisa Hui, Luis Huicho, Maria C Magnus, Andrew D Morris, Livia Nagy-Bonnard, Natasha Nassar, Sylvester Dodzi Nyadanu, Dedeke Iyabode Olabisi, Kirsten R Palmer, Lars Henning Pedersen, Gavin Pereira, Amy Racine-Poon, Manon Ranger, Tonia Rihs, Christoph Saner, Aziz Sheikh, Emma M Swift, Lloyd Tooke, Marcelo L Urquia, Clare Whitehead, Christopher Yilgwan, Natalie Rodriguez, David Burgner, Meghan B Azad, iPOP Study Team

Wellcome Open Res. 2021 Feb 2;6:21. doi: 10.12688/wellcomeopenres.16507.1. eCollection 2021.

Preterm birth is the leading cause of infant death worldwide, but the causes of preterm birth are largely unknown. During the early COVID-19 lockdowns, dramatic reductions in preterm birth were reported; however, these trends may be offset by increases in stillbirth rates. It is important to study these trends globally as the pandemic continues, and to understand the underlying cause(s). Lockdowns have dramatically impacted maternal workload, access to healthcare, hygiene practices, and air pollution - all of which could impact perinatal outcomes and might affect pregnant women differently in different regions of the world. In the international Perinatal Outcomes in the Pandemic (iPOP) Study, we will seize the unique opportunity offered by the COVID-19 pandemic to answer urgent questions about perinatal health. In the first two study phases, we will use population-based aggregate data and standardized outcome definitions to: 1) Determine rates of preterm birth, low birth weight, and stillbirth and describe changes during lockdowns; and assess if these changes are consistent globally, or differ by region and income setting, 2) Determine if the magnitude of changes in adverse perinatal outcomes during lockdown are modified by regional differences in COVID-19 infection rates, lockdown stringency, adherence to lockdown measures, air quality, or other social and economic markers, obtained from publicly available datasets. We will undertake an interrupted time series analysis covering births from January 2015 through July 2020. The iPOP Study will involve at least 121 researchers in 37 countries, including obstetricians, neonatologists, epidemiologists, public health researchers, environmental scientists, and policymakers. We will leverage the most disruptive and widespread "natural experiment" of our lifetime to make rapid discoveries about

preterm birth. Whether the COVID-19 pandemic is worsening or unexpectedly improving perinatal outcomes, our research will provide critical new information to shape prenatal care strategies throughout (and well beyond) the pandemic.

PMID: 34722933

12. Distinctive Neuroimaging Pattern in Term Newborns With Neonatal Placental Encephalopathy: A Case Series
Fatema Al Amrani, Guillaume Sébire, Moy Fong Chen, Pia Wintermark, Christine Saint-Martin

Pediatr Neurol. 2021 Oct 4;126:74-79. doi: 10.1016/j.pediatrneurol.2021.09.020. Online ahead of print.

Background: Identifying antepartum versus intrapartum timing and the cause of neonatal encephalopathy (NE) often remains elusive owing to our limited understanding of the underlying pathophysiological processes and lack of appropriate biomarkers. Objectives: This retrospective observational study describes a case series of term newborns with NE who displayed a recognizable magnetic resonance imaging pattern of immediately postnatal brain abnormalities that rapidly evolved toward cavitation. Our aim is to (1) report this neuroimaging pattern, (2) look for placental determinants, and (3) depict the outcome. Design/methods: This is a unicentric retrospective case series reporting the clinical, radiological, and laboratory findings of NE associated with a distinctive neuroimaging pattern, that is, immediately postnatal extensive corticosubcortical T2 hyperintensities, followed by rapid corticosubcortical cavitation that does not match the neuroimaging picture of intrapartum hypoxic-ischemic encephalopathy (HIE). Results: Seven term newborns presented bilateral corticosubcortical hyperintensities that were detected on T2 between day of life (DOL) 1-4, which rapidly evolved toward cystic encephalomalacia, that is, between DOL9 and DOL12. All these newborns presented with moderate/severe NE. The outcome was either neonatal death or quadriplegic cerebral palsy and epilepsy. None of the reported patients fulfilled the criteria of a high likelihood of acute intrapartum hypoxic-ischemic or quadriplegic cerebral palsy. All these newborns were exposed to chronic and/or acute placental inflammation and/or hypoxic-ischemic. Conclusions: To further define the antepartum causes of NE, early neuroimaging and a placental examination are recommended. Brain T2 hyperintense injuries before DOL4 followed by rapid cavitation before DOL12 might be biomarkers of NE from an antepartum/placental origin.

PMID: 34740136

13. Variation in Bacterial Respiratory Culture Results in Children With Neurologic Impairment
Amanda Warniment, Rebecca Steuart, Jonathan Rodean, Matt Hall, Sofia Chinchilla, Samir S Shah, Joanna Thomson

Hosp Pediatr. 2021 Nov;11(11):e326-e333. doi: 10.1542/hpeds.2020-005314.

Objectives: To examine bacterial respiratory cultures in children with neurologic impairment (NI) (eg, cerebral palsy), both with and without tracheostomies, who were hospitalized with acute respiratory infections (ARIs) (eg, pneumonia) and to compare culture results across hospitals and age groups. Methods: This multicenter retrospective cohort study included ARI hospitalizations for children aged 1 to 18 years with NI between 2007 and 2012 who had a bacterial respiratory culture obtained within 2 days of admission. Data from 5 children's hospitals in the Pediatric Health Information System Plus database were used. Organisms consistent with oral flora and nonspeciated organisms were omitted from analysis. The prevalence of positive respiratory culture results and the prevalence of organisms identified were compared across hospitals and age groups and in subanalyses of children with and without tracheostomies by using generalized estimating equations to account for within-patient clustering. Results: Of 4900 hospitalizations, 693 from 485 children had bacterial respiratory cultures obtained. Of these, 54.5% had positive results, although this varied across hospitals (range 18.6%-83.2%; $P < .001$). Pseudomonas aeruginosa and Staphylococcus aureus were the most commonly identified organisms across hospitals and age groups and in patients with and without tracheostomies. Large variation in growth prevalence was identified across hospitals but not age groups. Conclusions: The bacteriology of ARI in hospitalized children with NI differs from that of otherwise healthy children. Significant variation in prevalence of positive bacterial respiratory culture results and organism growth were observed across hospitals, which may be secondary to local environmental factors and microbiology reporting practices.

PMID: 34716209

14. Impact of fetal presentation on neurodevelopmental outcome in a trial of preterm vaginal delivery: a nationwide, population-based record linkage study
Anna Toijonen, Seppo Heinonen, Mika Gissler, Laura Seikku, Georg Macharey

Arch Gynecol Obstet. 2021 Oct 31. doi: 10.1007/s00404-021-06146-z. Online ahead of print.

Purpose: To assess the risk of adverse neurodevelopmental outcomes at the age of four after an attempted vaginal delivery according to the fetal presentation in birth. Methods: This retrospective record linkage study evaluated the risks of cerebral palsy, epilepsy, intellectual disability, autism spectrum disorder, attention-deficit/hyperactivity disorder, and speech, visual, and auditory disabilities among preterm children born after an attempted vaginal breech delivery. The control group comprised children born in a cephalic presentation at the same gestational age. This study included 23 803 singleton deliveries at gestational weeks 24 + 0-36 + 6 between 2004 and 2014. Results: From 1629 women that underwent a trial of vaginal breech delivery, 1122 (66.3%) were converted to emergency cesarean sections. At extremely preterm and very preterm gestations (weeks 24 + 0-31 + 6), no association between a trial of vaginal breech delivery and neurodevelopmental delay occurred. At gestational weeks 32 + 0-36 + 6, the risks of visual disability (aOR 1.67, CI 1.07-2.60) and autism spectrum disorders (aOR 2.28, CI 1.14-4.56) were increased after an attempted vaginal breech delivery as compared to vaginal cephalic delivery. Conclusion: A trial of vaginal breech delivery at extremely preterm and very preterm gestations appears not to increase the risk of adverse neurodevelopmental outcomes at the age of four. In moderate to late preterm births, a trial of vaginal breech delivery was associated with an increased risk of visual impairment and autism spectrum disorders compared to children born in cephalic presentation. A trial of vaginal preterm breech delivery requires distinctive consideration and careful patient selection.

PMID: 34718843

15. Nasal trumpet as a long-term remedy for obstructive sleep apnea syndrome in a child
Tammy Yau, Ruchi Bansal, Kimberly Hardin, Craig Senders, Kiran Nandalike

Case Reports SAGE Open Med Case Rep. 2021 Oct 26;9:2050313X211055303. doi: 10.1177/2050313X211055303. eCollection 2021.

We present a case of successful long-term use of nasal trumpet for severe obstructive sleep apnea syndrome in a child with cerebral palsy and complex medical issues. Obstructive sleep apnea syndrome is frequently seen in pediatric patients with cerebral palsy due to their abnormal airway tone and pulmonary vulnerability. Identifying children with cerebral palsy who are at risk for obstructive sleep apnea syndrome is important because its treatment can improve quality of life and seizure control. Although first-line treatment for obstructive sleep apnea syndrome is adenotonsillectomy, children with cerebral palsy are more likely to have residual obstructive sleep apnea syndrome postoperatively. Other options such as positive airway pressure therapy and other upper airway surgeries may pose significant challenges and tolerance issues, as in our patient. As demonstrated in our report, the low rate of complications and ease of use make nasal trumpets a potential long-term treatment option for children with obstructive sleep apnea syndrome who fail or cannot comply with the traditional treatment options.

PMID: 34721876

16. Motor functions, quality of life and maternal anxiety and depression in children with cerebral palsy of different intelligence levels
Elif Akçay, Yaşar Tanır, Serap Tıraş Teber, Birim Günay Kılıç

Turk J Pediatr. 2021;63(5):846-854. doi: 10.24953/turkjped.2021.05.012.

Background: Cerebral palsy (CP) is the most common motor disability in childhood. In addition to motor impairment, it is frequently accompanied by intellectual disability (ID). We aimed to investigate the associations between motor functions, quality of life (QoL) and maternal psychopathology in children with CP of different intelligence levels. Methods: In total, 37 children and adolescents (16 females and 21 males) between 4 and 18 years of age diagnosed with CP were recruited from a Pediatric Neurology Outpatient Clinic. Gross Motor Function Classification System (GMFCS) and Bimanual Fine Motor Function (BFMF) were used for the children`s motor functions assessment. Quality of life was determined by the caregivers with Pediatric Quality Of Life Inventory-Parent version (PedsQL-P). Maternal anxiety and depression levels were assessed using Beck Anxiety Inventory (BAI) and Beck Depression Inventory (BDI). Results: Moderate-severe ID (n=19)(13.5%,

37.8%) and normal IQ-mild ID (n=18) (32.4 %,16.2%) groups were evaluated in this study. GMFCS level 2 was more frequent in both groups. The majority of the severe-moderate ID group was at BFMF level 4, while the normal IQ-mild ID group was at BFMF level 2. PedsQL-P scores of children with CP, maternal BAI scores, and maternal BDI scores did not differ between the two groups ($p > 0.05$). Psychosocial PedsQL scores had a moderate negative correlation with the maternal BAI scores ($r=-0.41$, $p < 0.05$). There was also a moderate positive correlations between the ages of children and maternal BDI scores ($r=0.34$, $p < 0.05$). Conclusions: Our results demonstrated that maternal anxiety was correlated with psychosocial QoL in children with CP. Maternal depression scores increasing with the ages of the children with CP may also indicate the social support needs for mothers with children of chronic diseases. Further studies may reveal the associations with other biopsychosocial factors in children with CP of different intelligence levels by using longitudinal study designs with larger sample sizes.

PMID: 34738366

17. Impact of social support for mothers as caregivers of cerebral palsy children in Japan
Mutsuko Moriwaki, Hitomi Yuasa, Masayuki Kakehashi, Hideaki Suzuki, Yasuki Kobayashi

J Pediatr Nurs. 2021 Nov 1;S0882-5963(21)00307-9. doi: 10.1016/j.pedn.2021.10.010. Online ahead of print.

Purpose: Social supports are critical to alleviate the psychological and physical burden of primary caregivers of children with disabilities. This study aims to (1) clarify how cerebral palsy in children affects caregiving burden of the mother, and (2) identify the social supports that can effectively reduce that burden. Design and methods: This is a cross-sectional study in which mothers of children with cerebral palsy completed questionnaires and provided data regarding their child's condition, family support, social support usage, degree of satisfaction with supports, and caregiving burden. Results: We analyzed responses from 1190 mothers. Support usage, particularly of home-visit nursing, home care, home-visit rehabilitation, and mobility support, was higher in severely burdened groups. However, the proportion of satisfaction with social support in groups with light or no burden were higher, particularly in home care, home-visit rehabilitation, training/treatment, and short stays. Mothers whose children have an intellectual disability and gross exercise ≥1 in addition to tube feeding or intravenous nutrition especially felt a strong sense of burden. The most effective measure in reducing mother's sense of burden was short stays. Conclusions: Mothers with children who can move and have an intellectual disability felt more burden compared with mothers of bedridden children. The findings clarify that supports, such as home care and short stays, have a significant impact on reducing the mother's sense of burden. Practice implications: Due to the large sample size, we believe that the results can inform efforts to increase social support for caregivers.

PMID: 34736819

18. Global positioning system activity profile in male para footballers with cerebral palsy: Does training meet the match -play intensity in a three-day national tournament?
Alvin M Goh, Fadi Ma'ayah, Craig A Harms, Robert U Newton, Eric J Drinkwater

Am J Phys Med Rehabil. 2021 Nov 3. doi: 10.1097/PHM.0000000000001919. Online ahead of print.

Objective: To compare training and match loads for CP football athletes in a state-level development program to assess the specificity of training for competition in the para sport. Design: Time-motion analysis and heart-rate monitoring were applied during 20 training sessions of the Western Australia CP football 2017 training season, and seven competition matches of the 2017 Australian National Championships. The study employed an observational study design with between-subjects and within -subjects comparisons of training and match loads. Results: There were no significant differences between training and match loads in players' measurements of speed components and heart-rates, with two exceptions: higher match distance covered in very-high-speed ($g = 0.33$) and time in heart rate zone 1 (<75% HRmax,$g = 0.45$), and less time in HR Zone 2 ($g = 0.42$). Conclusion: Our results indicate that the athletes were conditioned to perform at similar intensities implemented during training, as demonstrated by the consistency between competition and training data in total distance, relative speed zones and relative heart rate zones. Coaches should apply this knowledge in the programming of training volume and intensities to obtain peak match fitness and specifically prepare athletes for competition in CP football.

PMID: 34740223

19. Cerebral Palsy, COVID-19, and Neurolipidosis in an 18-Year-Old Female
George S Stoyanov, Deyan L Dzhenkov, Hristo Popov, Emran Lyutfi, Lilyana Petkova

Case Reports Cureus. 2021 Sep 26;13(9):e18294. doi: 10.7759/cureus.18294. eCollection 2021 Sep.

Since the novel coronavirus (COVID-19) pandemic started, children and young adults have seldom been placed in high-risk groups, despite reports that they are at increased risk of severe forms of the disease and death in the presence of comorbidities. Herein we report an autopsy case of an 18-year-old female with a history of cerebral palsy (CP), recurrent respiratory infections, and newly diagnosed COVID-19, and who expired 22 days after presenting with symptoms of the disease. Gross findings were concurrent with CP-significant hypotrophy, with deep and wide brain sulci. The lungs grossly were with increased weight and blood-filled. Histopathology of the respiratory system showed the well-established COVID-19-associated alveolar multinucleated cells, type two pneumocyte hyperplasia, and vascular changes. Furthermore, foci of groups of enlarged cells with foamy cytoplasm were identified in the pulmonary interstitium. Similar changes were also seen in the spleen, liver, and central nervous system, concurrent with an unrecognized lipid storage disease. The clinically unrecognized neurolipidosis, corresponding morphologically and clinically to Niemann-Pick disease type B, leading to interstitial lung disease and recurrent respiratory infections, inevitably played a role in the severity and progression of COVID-19 in our case, despite the age.

PMID: 34722070

20. Death by fecaloma
Yosuke Usumoto, Michael S Pollanen

Forensic Sci Med Pathol. 2021 Nov 4. doi: 10.1007/s12024-021-00438-1. Online ahead of print.

A 59-year-old man with a history of cerebral palsy and dextroscoliosis died in a group home. He required supplemental oxygen and had no bowel movement for weeks prior to death. At autopsy, the abdomen was markedly distended and there were flexion contractures of the legs. Postmortem computed tomography revealed a dilated digestive tract and fecal loading in the sigmoid and rectum, marked upwardly displaced diaphragm and scoliosis. On internal examination, the diaphragm was displaced rostrally and the rectosigmoid colon contained 2.5 kg of fecaloma with two rectal fecaliths. Severe scoliosis with marked reduction in volume of thoracic cavity was present. Microscopic examination revealed chronic aspiration pneumonia and chronic pulmonary hypertension. Overall, four factors led to respiratory failure: fecaloma; cerebral palsy; scoliosis; and chronic aspiration pneumonia. Based on clinicopathological correlation, the cause of death was determined to be a combination of these factors, and the key acute factor was the fecaloma.

PMID: 34735683

21. EBNEO Commentary: Should We Abandon Therapeutic Hypothermia for Neonatal Encephalopathy in Low and Middle-Income Countries?
Nehad Nasef, Islam Nour, Hesham Abdel-Hady

Acta Paediatr. 2021 Nov 5. doi: 10.1111/apa.16175. Online ahead of print.

Therapeutic hypothermia (TH) is the standard of care in neonates with moderate and severe hypoxic ischemic encephalopathy (HIE) as it lowers the rates of mortality, cerebral palsy, hearing and visual impairment, and neurodevelopmental delay. The majority of evidence towards the effectiveness and safety of TH has been pooled out from data of well-designed randomized controlled trials which were conducted in high-income countries.

PMID: 34738256

Prevention and Cure

22. Cell-based treatment of cerebral palsy: still a long way ahead
Mohammad Shariati, Reza Jafarzadeh Esfahani, Hamid Reza Bidkhori, Ehsan Sabouri, Shadi Mehrzad, Ariane Sadr-Nabavi

Curr Stem Cell Res Ther. 2021 Nov 1. doi: 10.2174/1574888X16666211102090230. Online ahead of print.

Background: Cerebral palsy (CP) is a permanent neurodevelopmental disorder with considerable global disability. Various rehabilitation strategies are currently available. However, none represents a convincing curative result. Cellular therapy recently holds much promise as an alternative strategy to repair neurologic defects. Method: In this narrative review, a comprehensive search of the MEDLINE and ClinicalTrials.gov was made, using the terms: "cell therapy" and "cerebral palsy", including published and registered clinical studies, respectively. Results: The early effects of these studies demonstrated that using cell therapy in CP patients is safe and improves the deficits for a variable duration. Despite such hopeful early bird results, the long-term outcomes are not conclusive. Conclusions: Due to the heterogeneous nature of CP, personal factors seem essential to consider. Cell dosage, routes of administration, and repeated dosing are pivotal to establish optimal personalized treatments. Future clinical trials should consider employing other cell types, specific cell modifications before administration, and cell-free platforms.

PMID: 34727864