1. Early Intervention and Postural Adjustments During Reaching in Infants at Risk of Cerebral Palsy.
van Balen LC, Dijkstra LJ, Dirks T, Bos AF, Hadders-Algra M.


PURPOSE: To investigate postural effects of the family-centered program, COPing with and CARing for infants with special needs (COPCA), applied at 3 to 6 months' corrected age in infants at high risk of cerebral palsy. Previously, we reported postural differences between the infants at risk of CP in the control group of the current study and a group of infants developing typically. Now we focus on differences between 2 intervention groups. METHODS: We explored postural adjustments during reaching in seated infants at 4, 6, and 18 months using surface electromyography of arm, neck, and trunk muscles. Infants randomly received the family-centered program or another infant physical therapy. Using videotaped intervention sessions, we investigated correlations between time spent on specific physical therapeutic actions and direction specificity, recruitment order, and anticipatory activation at 18 months. RESULTS: Postural adjustments in both groups were similar, but development of direction specificity and anticipatory activation in COPCA infants better mimicked typical development. These 2 parameters were associated with COPCA-type physical therapeutic actions. CONCLUSIONS: Postural control was similar after both interventions. Positive outcomes were associated with fewer intervening actions of the therapist and greater allowance of spontaneous movements.

PMID: 30865144

2. Daily and Weekly Rehabilitation Delivery for Young Children With Gross Motor Delay: A Randomized Clinical Trial Protocol (the DRIVE Study).


PURPOSE: The proposed project tests the principle that frequency of rehabilitation is an important regulator of therapeutic response in infants. METHODS: We will randomize 75 infants with cerebral palsy, 6 to 24 months of age and/or Gross Motor Function Classification System levels III to V (higher severity), to determine the short-term and long-term effects of 3 dosing protocols consisting of an identical number of 2-hour sessions of the same motor learning-based therapy applied over a different total number of calendar weeks. RESULTS AND CONCLUSIONS: The results will inform clinicians, families, and scientists about dosing and will provide needed recommendations for frequency of rehabilitation to optimize motor function and development of young children with cerebral palsy.

PMID: 30865149


BACKGROUND: This systematic review evaluates the use of manual therapy for clinical conditions in the pediatric population, assesses the methodological quality of the studies found, and synthesizes findings based on health condition. We also assessed the reporting of adverse events within the included studies and compared our conclusions to those of the UK Update report. METHODS: Six databases were searched using the following inclusion criteria: children under the age of 18 years old; treatment using manual therapy; any type of healthcare profession; published between 2001 and March 31, 2018; and English. Case reports were excluded from our study. Reference tracking was performed on six published relevant systematic reviews to find any missed article. Each study that met the inclusion criteria was screened by two authors to: (i) determine its suitability for inclusion, (ii) extract data, and (iii) assess quality of study. RESULTS: Of the 3563 articles identified, 165 full articles were screened, and 50 studies met the inclusion criteria. Twenty-six articles were included in prior reviews with 24 new studies identified. Eighteen studies were judged to be of high quality. Conditions evaluated were: attention deficit hyperactivity disorder (ADHD), autism, asthma, cerebral palsy, clubfoot, constipation, cranial asymmetry, cuboid syndrome, headache, infantile colic, low back pain, obstructive apnea, otitis media, pediatric dysfunctional voiding, pediatric nocturnal enuresis, postural asymmetry, preterm infants, pulled elbow, suboptimal infant breastfeeding, scoliosis, suboptimal infant breastfeeding, temporomandibular dysfunction, torticollis, and upper cervical dysfunction. Musculoskeletal conditions, including low back pain and headache, were evaluated in seven studies. Twenty studies reported adverse events, which were transient and mild to moderate in severity. CONCLUSIONS: Fifty studies investigated the clinical effects of manual therapies for a wide variety of pediatric conditions. Moderate-positive overall assessment was found for 3 conditions: low back pain, pulled elbow, and premature infants. Inconclusive unfavorable outcomes were found for 2 conditions: scoliosis (OMT) and torticollis (MT). All other condition's overall assessments were either inconclusive favorable or unclear. Adverse events were uncommonly reported. More robust clinical trials in this area of healthcare are needed. TRIAL REGISTRATION: PROSPERA registration number: CRD42018091835.

PMID: 30866915

Germain AM, Blackmore AM, Gibson N, Newell B, Williams SA.


PURPOSE: To assess effects of adaptive bungee trampoline training for children with cerebral palsy. METHODS: This was a single-subject intervention study, A-B-A, with 4 children aged 6 to 11 years. Measurements included muscle strength, balance, functional muscle strength, functional mobility, selected Gross Motor Function Measure items, heart rate, enjoyment, and for adverse events-range of motion and spasticity. Goals were measured using the Canadian Occupational Performance Measure. RESULTS: Lower limb muscle strength improved in 3 children, and balance and functional strength in 2 children. The child who was not walking increased sitting and supported standing times. All participants had clinically significant increases on the Canadian Occupational Performance Measure. Adherence and enjoyment were high, with no adverse effects. CONCLUSION: Adaptive bungee trampoline training can improve strength, balance, and functional mobility in children with cerebral palsy.

PMID: 30865143

5. Stiffness of hip adductor myofibrils is decreased in children with spastic cerebral palsy.


Cerebral palsy (CP) is the result of a static brain lesion which causes spasticity and muscle contracture. The source of the increased passive stiffness in patients is not understood and while whole muscle down to single muscle fibres have been investigated, the smallest functional unit of muscle (the sarcomere) has not been. Muscle biopsies (adductor longus and gracilis) from pediatric patients were obtained (CP n = 9 and control n = 2) and analyzed for mechanical stiffness, in-vivo sarcomere length and titin isoforms. Adductor longus muscle was the focus of this study and the results for sarcomere length showed a significant increase in length for CP (3.6 μm) compared to controls (2.6 μm). Passive stress at the same sarcomere length for CP compared to control was significantly lower in CP and the elastic modulus for the physiological range of muscle
was lower in CP compared to control (98.2 kPa and 166.1 kPa, respectively). Our results show that CP muscle at its most reduced level (titin myofibril) is more compliant compared to normal, which is completely opposite to what is observed at higher structural levels (single fibres, muscle fibre bundles and whole muscle). It is noteworthy that at the in vivo sarcomere length in CP, the passive forces are greater than normal, purely as a functional of these more compliant sarcomeres operating at long lengths. Titin isoforms were not different between CP and non-CP adductor longus but titin:nebulin was reduced in CP muscle, which may be due to titin loss or an over-expression of nebulin in CP muscles.

PMID: 30853092

6. Computational modeling of neuromuscular response to swing-phase robotic knee extension assistance in cerebral palsy.
Lerner ZF, Damiano DL, Bulea TC.


Predicting subject-specific responses to exoskeleton assistance may aid in maximizing functional gait outcomes, such as achieving full knee-extension at foot contact in individuals with crouch gait from cerebral palsy (CP). The purpose of this study was to investigate the role of volitional and non-volitional muscle activity in subject-specific responses to knee extension assistance during walking with an exoskeleton. We developed a simulation framework to predict responses to exoskeleton torque by applying a stretch-reflex spasticity model with muscle excitations computed during unassisted walking. The framework was validated with data collected from six individuals with CP. Framework-predicted knee angle at terminal swing was within 4 ± 4° (mean ± sd) of the knee angle measured experimentally without the addition of spasticity. Kinematic responses in two-thirds of the participants could be accurately modeled using only underlying muscle activity and the applied exoskeleton torque; incorporating hamstring spasticity was necessary to recreate the measured kinematics to within 1 ± 1° in the remaining participants. We observed strong positive linear relationships between knee extension and exoskeleton assistance, and strong negative quadratic relationships between knee extension and spasticity. We utilized our framework to identify optimal torque profiles necessary to achieve full knee-extension at foot contact. An angular impulse of 0.061 ± 0.025 Nm·s·kg⁻¹·deg⁻¹ with 0.013 ± 0.002 Nm·kg⁻¹·deg⁻¹ of peak torque and 4.1 ± 1.9 W·kg⁻¹·deg⁻¹ peak mechanical power was required to achieve full knee extension (values normalized by knee excursion). This framework may aid the prescription of exoskeleton control strategies in pathologies with muscle spasticity. https://simtk.org/projects/knee-exo-pred/.

PMID: 30862380

Papageorgiou E, Nieuwenhuys A, Vandekerckhove I, Van Campenhout A, Ortibus E, Desloovere K.


BACKGROUND: Gait classification systems (GCSs) aim to aid clinicians and researchers in categorizing the gait of pathological populations, with the intent to improve the communication between them, to support treatment planning and enable the evaluation of patients over time. Throughout the years, various GCSs have been defined for children with cerebral palsy (CP), which were first summarized in a systematic review published in 2007. RESEARCH QUESTION: The current systematic review aimed to: a) identify GCSs that have been more recently developed, b) appraise their methodological quality and c) specify the most commonly used multiple joint gait patterns for children with CP reported in literature. METHODS: Four databases (Medline, EMBASE, CINAHL, Web of Science) were searched until July 2017. Several forms of validity and the reliability of these studies were assessed according to the principles of the consensus-based standards for the selection of health measurement instruments checklist or criteria defined in the original review. All published GCSs were also scrutinized in order to identify multiple joint patterns that have reached a predefined level of consensus. RESULTS: Thirty-six studies were considered in 15 of them being GCSs that were not included in the original review. The validity, reliability and clinical applicability of all GCSs was reported, including 3 studies from the original review. Six multiple joint patterns for children with CP reached a consensus in literature. CONCLUSION: Since the previous review, obvious progress has been made in the field of GCSs for CP, resulting in improved methodological quality of the majority of published GCSs. This encouraged the applicability of GCSs in clinical or research settings. The six reliable, valid and commonly used multiple joint patterns, emerging from this systematic review, may aid clinical and research applications and create a common language among healthcare providers.

PMID: 30851621
8. The effects of dual tasks on gait in children with cerebral palsy.


AIM: To assess the gait and cognitive performances of children with cerebral palsy (CP) during dual tasks (DT) in comparison to typically developing (TD) children. METHOD: This prospective, observational, case-control study included 18 children with CP (7 girls, 11 boys; median age 12 [10:13] years and 19 controls (9 girls, 10 boys; median age 12 [10:13y6mo] years).

Performances were recorded during a simple walking task, 5 DT (walking + cognitive tasks with increasing cognitive load), and 5 simple cognitive tasks (while sitting). Gait parameters were computed using an optoelectronic system during walking tasks. Six parameters were selected for analysis by a principal component analysis. Cognitive performance was measured for each cognitive task. The dual-task cost (DTC) was calculated for each DT.

RESULTS: Gait performance decreased in both groups as DT cognitive load increased (e.g., walking speed normalized by leg length, in simple task: 1.25 [1.15:1.46] s-1 for CP, 1.53 [1.38:1.62] s-1 for TD; DT with highest load: 0.64 [0.53:0.80] s-1 for CP, 0.95 [0.75:1.08] s-1 for TD). The CP group performed significantly worse than TD group in every task (including the simple task), but DTC were similar in both groups. A task effect was found for the majority of the gait parameters. INTERPRETATION: The reduced gait performance induced by DT may generate underestimated difficulties for children with CP in daily-life situations, where DT are common. This should be considered in clinical assessments.

PMID: 30875601

9. Validity of the Early Activity Scale for Endurance and the 6-Minute Walk Test for Children With Cerebral Palsy.
Fiss AL, Jeffries L, Yocum A, Westcott McCoy S; On Track Study Team.


PURPOSE: This study aimed to describe Early Activity Scale for Endurance (EASE) scores and 6-minute walk test (6MWT) distances of children with cerebral palsy (CP) by functional ability level, sex, and age and to examine the convergent validity of the 2 tests. METHODS: A total of 708 participants with CP, Gross Motor Function Classification System (GMFCS) levels I to V, completed the EASE, and 376 of the study participants (3-12 years), GMFCS levels I to III, completed the 6MWT.

RESULTS: Children with CP vary in EASE scores and 6MWT distances based on GMFCS level and, to a lesser extent, age. The EASE and the 6MWT demonstrate a statistically significant but low, positive correlation. CONCLUSIONS: Understanding the relationship between these outcomes and GMFCS levels and age assists clinicians in establishing plans of care targeted at improving endurance for activity and functional walking capacity for children with CP.

PMID: 30865148

10. Multiple relationships between Tardieu, Kinematic data, and Wolf Motor Function Test with children with cerebral palsy.
Hwnag J, Lee JA, Cha YJ, Lee DH, You JSH.


BACKGROUND: The World Health Organization (WHO) has developed the International Classification of Functioning, Disability and Health (ICF) model in order to provide a theoretical foundation of physical therapy diagnosis and intervention. However, the multidirectional relationships between the body structure/function domain variables (spasticity and movement kinematics) and the activity domain variables (e.g. reaching, grasping, folding, and lifting abilities) using the Wolf Motor Function Test (WMFT) remain unknown. OBJECTIVE: The purpose of the present study was to examine the directional relationships between the body function and structure domain variables and the activity domain variables using the WMFT. METHODS: Nineteen children with cerebral palsy (CP) were recruited from a major rehabilitation center. Standardized clinical tests included Tardieu scale and WMFT, which were used to measure the body function and structure domain (spasticity and activity domain (reaching, grasping, folding, and lifting abilities). An eight infrared motion capture system (VICON, Oxford, UK) was used to collect kinematics data during reaching, which represent the body function and structure domain variables. Correlational analysis was performed at P < 0.05. RESULTS: Our results revealed a fair to strong relationship between the body function and structure domain variables (11 out of 18 kinematic data) and activity domain variables (WMFT). However, no significant correlation was observed between the Tardieu score and the kinematics data or between the Tardieu score and the WMFT variables. CONCLUSIONS: The present findings suggest that the body structure/function domain variables (Kinematic
data) are closely associated with activity domain variables (WMFT). However, the body function and structure domain variables within Tardieu spasticity and kinematic data variables were not associated each other, nor between Tardieu spasticity and activity domain variables (WMFT), indicating that Tardieu spasticity test does not seem to account for or reflect active kinematic movement or WFMT variables. This finding provides an important clinical insight when developing a comprehensive assessment and intervention for children with CP.

PMID: 30856130

Gonçalves RV, Fonseca ST, de Araújo PA, Souza TR, Resende RA, Mancini MC.


PURPOSE: Reduced propulsive capability can impact negatively on mobility activities of many children with spastic unilateral cerebral palsy (SUCP). This study investigated the effect of a task-oriented training program combined with functional electrical stimulation (FES) on the motor capacity of children with SUCP. METHODS: Single-case A-B design with follow-up. Gross motor function and biomechanical walking data of 4 children with SUCP were measured repeatedly across the baseline, intervention, and follow-up phases. Intervention was a task-oriented training program combined with FES applied on the gastrocnemius. Outcome variables included gait speed, impulsive torque, and ankle/hip power generation ratio. The 2-SD band and celeration line methods compared outcomes among the baseline, intervention, and follow-up periods. RESULTS: One child improved walking speed. All children improved impulsive torque and ankle/hip power ratio of the affected leg. All children improved gross motor function. CONCLUSION: The intervention improved children's propulsive capability and positively influenced their mobility.

PMID: 30865146

Kok SE, van Valenberg HFJP, van Hulst K, Jongerius P, Erasmus CE, van den Hoogen FJA.


AIM: This study evaluated whether the effect of submandibular gland botulinum neurotoxin A (BoNT-A) injection can predict the outcome of submandibular duct relocation with sublingual gland excision (SMDR) in children with drooling. Furthermore, we compared the effectiveness of both procedures. METHOD: A retrospective cohort study was performed in 42 children and adolescents (25 males, 17 females; mean [SD] age at BoNT-A injection 11y [4], range 4-20y; mean [SD] age at SMDR 15y [4], range 7-23y) with cerebral palsy or another non-progressive developmental disability who had undergone both BoNT-A injection and SMDR for drooling. Main outcomes were the drooling quotient and the visual analogue scale (VAS) on drooling severity at 8 weeks and 32 weeks follow-up. RESULTS: Failure or success of previous BoNT-A injections had no influence on success of consecutive SMDR. Relative change in main outcomes showed no significant relation between BoNT-A injection and SMDR for any follow-up measurement. After 8 weeks, SMDR was more successful than BoNT-A injection in diminishing VAS (VAS 80.0% vs 54.3%; drooling quotient 56.2% vs 51.0%). After 32 weeks, both drooling quotient (64.3% vs 29.5%) and VAS (75.7% vs 37.1%) showed significantly higher proportions of success for SMDR. INTERPRETATION: The effect of submandibular BoNT-A injection does not predict subsequent SMDR success in drooling. Furthermore, SMDR has a larger and longer-lasting positive effect on drooling than BoNT-A injections. WHAT THIS PAPER ADDS: Submandibular botulinum neurotoxin A (BoNT-A) injection effect does not predict submandibular duct relocation with sublingual gland excision outcome. Submandibular duct relocation is more effective and more permanent than BoNT-A injection.

PMID: 30854648

13. [Priorities and Goals of Botulinum Toxin A Treatment in Cerebral Palsy].
Klochkova OA, Kurenkov AL.

Botulinum toxin A (BTA) injections are an effective method of spasticity treatment in cerebral palsy (CP) but still there are a lot of questions about the selection of target muscles. The article summarizes currently accepted approaches to the goal setting and prioritizing in CP botulinum toxin therapy according to the form of CP, GMFCS level, age, spasticity level and other factors. The authors discuss the Goal Attainment Scale (GAS) and its possibilities in the BTA injections planning and evaluation of the results. Attention is also paid to the 'key muscle concept' in the multilevel spasticity treatment in CP and the additional factors that can influence the effectiveness of injections. The above approaches to the detection of patients' problems and setting of BTA treatment goals can help to prevent serious mistakes and disappointment in this effective method of treatment.

PMID: 30874536

14. The efficacy of botulinum toxin a lower limb injections in addition to physiotherapy approaches in children with cerebral palsy: A systematic review.
Yana M, Tutuola F, Westwater-Wood S, Kavlak E.


BACKGROUND: To assess treatment effect of lower limb botulinum toxin type A (BTX-A) in combination with physiotherapy approaches on gross motor functions in children with cerebral palsy compared with only physiotherapy treatment. OBJECTIVE: The purpose of this review was to analyze the efficacy botulinum toxin a lower limb injections in addition to physiotherapy approaches in children with cerebral palsy. METHODS: A literature search was conducted in the following databases: Cochrane, PEDro, PubMed, MEDLINE, AMED and EMBASE. The searches were limited to the period from July 2009 to July 2015. The intervention had to contain BTX-A into the lower limb plus physiotherapy approaches and be compared with only physiotherapy. The methodological quality and clinical relevance were independently assessed by the authors. RESULTS: The database search resulted in a total of 1521 studies, of which 4(Level II of evidence) trials were included in this review. The population represented by were age between from 11 month to 15 years. Overall there were 153 children all diagnosed with CP(87 Male, 66 Female). CONCLUSIONS: The use of BTX-A injections in addition to physiotherapy approaches seems to have positive effect on spasticity and ROM. However, the question of whether the treatment of BTX-A plus physiotherapy has a greater improvement on functional capacity, such as gross motor function or gait parameter than only physiotherapy treatments, was inconclusive. Further investigation by rigorous studies is required.

PMID: 30856126

15. Inherited Ataxia and Intrathecal Baclofen for the Treatment of Spasticity and Painful Spasms.
Berntsson SG, Gauffin H, Melberg A, Holz A, Landtblom AM.


BACKGROUND: Intrathecal baclofen (ITB) treatment is considered a powerful tool in the management of severe spasticity in neurological conditions such as multiple sclerosis, cerebral palsy, and traumatic spinal cord and brain injury. OBJECTIVES: The objective of this study was to assess the effectiveness of the ITB in patients with inherited ataxia suffering from severe painful spasms and/or spasticity. METHOD: A total of 5 patients with spinocerebellar ataxia 3 or 7 or Friedreich's ataxia were included in this observational multicenter study. The patients were interviewed and completed outcome measures assessing pain (The Brief Pain Inventory), fatigue (Fatigue Severity Scale), and life satisfaction (LiSAT-9) before and 1 year after the treatment. Spasticity (Modified Ashworth Scale) and spasm frequency (SPFS) were measured objectively for each patient. RESULTS: The mean treatment time was 1.9 years. Evaluation of established standard forms revealed symptomatic relief from spasticity and/or pain. Life satisfaction improved in all patients. Conclusions: We report the potential beneficial effects of ITB treatment in patients with inherited ataxia who also suffer from spasticity/spasms. ITB treatment indication in neurological disorders allows for extension to the treatment of spasticity/spasms in patients with hereditary ataxia.

PMID: 30870851


AIM: To describe coping strategies in children and adolescents with cerebral palsy (CP), relative to age. METHOD: Patients were prospectively recruited from two paediatric rehabilitation centres in France. The Pediatric Pain Coping Inventory - French and Structured Pain Questionnaire were completed by an experienced professional for each child. RESULTS: One hundred and forty-two children with CP were included (80 males, 62 females; median age 12y; IQR=8–15y). They generally used fewer coping strategies than typically developing children ('Seeks social support and action': 12.47 vs 12.85, p=0.477; 'Cognitive self-instruction': 9.28 vs 10.90, p<0.001; 'Distraction': 4.89 vs 7.00, p<0.001; 'Problem solving': 4.43 vs 5.19, p<0.001). In the CP group, 'Seeks social support and action' decreased with age (p=0.021) and 'Cognitive self-instruction' increased with age (p<0.001). 'Problem solving' and 'Distraction' did not change with age. Coping strategies were influenced by Gross Motor Function Classification System level (p=0.022) and history of surgery (p=0.002). INTERPRETATION: Children with CP generally used fewer pain coping strategies than typically developing children and tended to rely on social support. Use of active strategies increased with age; however, they appeared later than in typically developing children and were used to a lesser extent. WHAT THIS PAPER ADDS: Children with cerebral palsy (CP) use fewer pain-coping strategies than typically developing children. Children with CP tend to use social support to cope with pain. Children with CP learn more appropriate strategies from previous painful experiences. Active coping strategies appear later but remain underused in children with CP.

PMID: 30854638

Xû N, Matsumoto H, Roye D, Hyman J.


INTRODUCTION: This study compares the current practice patterns of pain assessment and management between children with and without CP following either posterior spinal instrumentation and fusion (PSIF) or hip osteotomy (HO). METHODS: Two cohorts of CP patients were retrospectively identified and matched with non-CP patients based on age, surgical procedure, and approach to post-operative pain management. Sixteen CP patients undergoing PSIF and twenty-two undergoing HO were respectively matched with the same numbers of non-CP patients receiving the same procedures. The frequency of assessments conducted, highest pain scores recorded on each post-operative day (POD), and the amount of adjuvant analgesics administered were collected for POD 0–4. RESULTS: Patients with CP were significantly more frequently evaluated for pain post-operatively, tended to have lower pain scores as measured by current scales, and received slightly fewer analgesics. Patients with CP differed from their non-CP counterparts in both frequency and method of post-operative pain assessment. CONCLUSIONS: The purpose of this study is to elucidate the current state of post-operative pain assessment and management in children with CP undergoing major orthopaedic surgeries, to improve CP patient/caregiver understanding and expectation of the post-operative experience regarding pain, and to provide recommendations for improving the post-operative care for these patients.

PMID: 30850174

18. Spinal dorsal rhizotomy plus concurrent left and right gastrocnemii releases in a 7-year-old child with haemophilia A and spastic cerebral palsy.
Bladen M, Main E, Chugh D, Liesner R.


PMID: 30866113

Tsubouchi Y, Tanabe A, Saito Y, Noma H, Maegaki Y.


AIM: To assess the long-term natural course and prognosis of epilepsy in patients with cerebral palsy (CP). METHOD: We retrospectively collected data for 72 patients (36 males, 36 females) with CP who had epilepsy who visited our institutions between 1980 and 2015. The data from medical records, electroencephalography (EEG), and neuroimaging findings were reviewed. Time-to-event statistical analyses were performed to analyse the remission outcome and the Cox regression model.
was used for multivariate analyses. RESULTS: Median age at onset of epilepsy was 2 years 0 months, and 17 years 0 months at the latest follow-up. In total, 34 patients (47%, 0.043 per person-year) achieved seizure remission at a median age of 11 years 0 months. Favourable factors for seizure remission included older age, motor disability being able to roll over/crawl but not able to sit, intellectual disability with an IQ between 36 and 70, normal findings on neuroimaging, and CP type other than spastic quadriplegia. In multivariate analysis, spastic quadriplegia was found to be associated with continued seizure activity. Antiepileptic drugs could be discontinued without relapse in 10 patients at a median age of 16 years 6 months, occurring 11 years 6 months after the onset of epilepsy. The drugs were terminated if the patient was aged at least 10 years and had perinatal causative aetiology and normalization or amelioration of epileptiform discharges on EEG. INTERPRETATION: The remission rate of epilepsy in CP increases up to young adulthood, and termination of antiepileptic drugs can be considered in selected cases at older ages. WHAT THIS PAPER ADDS: The remission rate of epilepsy in cerebral palsy increases up to 20 years after onset. In some cases, antiepileptic drugs (AEDs) can be terminated without relapse. Older age, perinatal aetiology, and improvement on electroencephalography are favourable factors for terminating AEDs.

PMID: 30854645

20. The experience of the multidisciplinary team in epilepsy management from a resource-limited country.


OBJECTIVE: The use of multidisciplinary teams (MDTs) is a global trend in disease management, while China is still at the exploratory stage MDTs. We aimed to summarize our experience and assess the impact of MDT use in managing patients with epilepsy and optimizing their seizure outcomes. METHODS: Our MDT is staffed with skilled epileptologists, electroencephalography experts, neurosurgeons, child neurologists, radiologists, and psychiatrists. The MDT discussion has been carried out once or twice a week since 2013. We reviewed our consecutive patients discussed at our MDT from March 2013 to December 2017. The detailed clinical characteristics, suggestions, and follow-up data were collected and analyzed. RESULTS: A total of 1088 patients (604 male, 484 female) were included in this study. The median age at MDT discussion was 21 years (range 10 months to 79 years). Three hundred eighty-seven patients (35.6%) were younger than 18 years of age. The median age at seizure onset was 12 years (range 2 days to 77 years). Most patients (80.4%) had at least one seizure per month and most (77%) took 2 or more antiepileptic drugs. A total of 70.6% of patients reached the standard of drug-resistant epilepsy and 74.2% of brain magnetic resonance imaging (MRI) studies detected positive findings. After detailed MDT discussion, 18 patients were diagnosed as having nonepileptic diseases, including psychogenic nonepileptic seizure, syncope, sleep disorder, paroxysmal kinesigenic dyskinesia, withdrawal symptom, and cerebral palsy. Three hundred eighty-two patients (35.1%) were suitable for resective surgery. Among the postoperative patients successfully followed up for more than 1 year, 72.7% (136/187) received favorable outcomes (Engel class I). The seizure-free rate was 78.6% after temporal lobe surgery and 58.9% after extratemporal surgery. SIGNIFICANCE: Epilepsy management can be optimized through MDT discussion to attain accurate diagnosis and favorable seizure outcomes. There is still room for MDT improvement in resource-limited countries.

PMID: 30868118

21. Vitamin D status of children with cerebral palsy: Should vitamin D levels be checked in children with cerebral palsy?
Akpinar P.


OBJECTIVE: We aimed to investigate the vitamin D status of children with cerebral palsy (CP). METHODS: A total of 274 children (111 females and 163 males), aged between 1 and 19 years with CP, who came to the Physical Medicine and Rehabilitation, Pediatric Rehabilitation Outpatient Clinic between October 2013 and March 2017, were included in our study. Demographics, data concerning the details of each child's comorbidity, the Gross Motor Function Classification System (GMFCS), and Manual Ability Classification System (MACS) scores were recorded. The serum 25 hydroxy vitamin D [25(OH)D], calcium (Ca), phosphate (P), and parathormone (PTH) levels were also recorded. RESULTS: The mean age of children with CP was 21 years (range 10 months to 79 years). Seven patients (35.6%) were younger than18 years of age. The 25(OH)D level <5 ng/ml, considered as vitamin D deficiency; 62 children at the 25(OH)D level 5-<12 ng/ml, regarded as vitamin D insufficiency; 62 children at the 25(OH)D level 12-<20 ng/ml, considered as vitamin D insufficiency, 43 children at the 25(OH)D level 20-<30 ng/ml, considered as vitamin D sufficiency, and 15 children at the 25(OH)D level >30 ng/ml. A total of 36 children were already taking vitamin D supplements. There was a significant correlation between the 25(OH)D levels and GMFCS and MACS levels.
and associated impairments such as the epilepsy history, intellectual delay, teeth problems, and growth retardation (p<0.05). CONCLUSION: Our results revealed that the children with CP who are not ambulatory (GMFCS levels IV-V) and have associated impairments were prone to vitamin D deficiency, and thus should be checked for vitamin D.

PMID: 30860516

van Aswegen T, Myezwa H, Potterton J, Stewart A.


BACKGROUND: Caregivers of children with cerebral palsy (CP) are at risk of having high stress levels and poor quality of life (QOL) which could have a detrimental effect on themselves and their children. Taking caregivers' well-being into consideration is therefore important when providing rehabilitation to children with CP. Interventions to mediate primary caregiver stress and QOL using an educational tool have not been tested in this population in South Africa. OBJECTIVES: The aim of this study was to determine the effect of a group-based educational intervention, Hambisela, on stress levels and QOL of primary caregivers of children with CP in Mamelodi, a township in Gauteng, South Africa. METHOD: Eighteen primary caregivers of children with CP participated in a quasi-experimental pretest-post-test pilot study. Hambisela, a group-based educational intervention, was carried out once a week over 8 consecutive weeks. Caregiver stress and QOL were assessed before and after the intervention using the Parenting Stress Index-Short Form (PSI-SF) and the Paediatric Quality of Life-Family Impact Module (PedsQLTM-FIM). Sociodemographic information was assessed using a demographic questionnaire. The Gross Motor Function Classification System (GMFCS) was used to assess the gross motor level of severity of CP in the children. RESULTS: Data were collected for 18 participants at baseline and 16 participants at follow-up. At baseline, 14 (87.5%) participants had clinically significant stress which reduced to 11 (68.8%) at follow-up. There was no significant change in primary caregiver's stress levels (p = 0.72) and QOL (p = 0.85) after the Hambisela programme. Higher levels of education were moderately associated with lower levels of primary caregiver stress (r = -0.50; p = 0.03). CONCLUSION: Most primary caregivers in this pilot study suffered from clinically significant stress levels. Hambisela, as an educational intervention, was not effective in reducing the stress or improving the QOL in these primary caregivers of children with CP. Future studies with a larger sample size are needed to investigate the high stress levels of primary caregivers of children with CP. CLINICAL IMPLICATIONS: Rehabilitation services for children with disabilities should include assessments to identify caregivers with high stress levels. Holistic management programmes should also include care for the carers.

PMID: 30863797

23. Longitudinal validation of the CPCHILD in a Dutch sample of non-ambulatory children with severe disabilities.


BACKGROUND: The purpose of this study is to evaluate the longitudinal validity of the Dutch version of the Caregiver Priorities and Child Health Index of Life with Disabilities (CPCHILD), a health-related quality of life instrument for non-ambulatory children with severe motor disabilities and accompanying disorders. METHODS: The effect of two interventions, Botox injections in the hip region and percutaneous endoscopic gastrostomy, was followed over time. Caregivers (n=38) of non-ambulatory children (26 boys, 12 girls; mean age: 9y, 5mo (4y, 9mo)) with severe disabilities completed the questionnaire prior to the intervention, at three months and six months follow-up. Seven a priori hypotheses were formulated. Longitudinal validity was analysed by paired t-test of the pre-post scores and correlation analysis between the change-scores and two external criteria, a caregivers' perceived change in health-related quality of life of the child questionnaire and a general health-related quality of life instrument. RESULTS: The results reported here follow completely the pattern we hypothesised for four analyses and partially in the remaining three. In the Botox group the mean change-score at three months was 6.9 points (p<0.05) which exceeds the minimal clinically important difference of 5.8 points. At six months the effect was diminished to 4.5 points, in line with the temporary effect of Botox. There were moderate positive correlations between the change-scores and an external criterion (Spearman's rho: 0.46 - 0.58). CONCLUSIONS: This study indicates that the CPCHILD-Dutch Version has sufficient longitudinal validity statistically and clinically in non-ambulatory children with severe disabilities.

PMID: 30870582


OBJECTIVE: To determine the efficacy of a hospital-based intervention that transitions into existing community support, in enhancing developmental outcomes at 2 years of corrected age in infants born at less than 32 weeks. STUDY DESIGN: In total, 323 families of 384 infants born <32 weeks were randomized to receive intervention or care-as-usual. The intervention teaches parents coping skills, partner support, and effective parenting strategies over 4 hospital-based and 4 home-phone sessions. At 2 years of corrected age maternally reported child behavior was assessed by the Infant and Toddler Social Emotional Adjustment Scale. Observed child behavior was coded with the Revised Family Observation Schedule. Cognitive, language, and motor skills were assessed with the Bayley Scales of Infant and Toddler Development III. RESULTS: Mean gestational age of infants was 28.5 weeks (SD = 2.1), and mothers' mean age was 30.6 years (SD = 5.8). A total of 162 families (n = 196 infants) were allocated to intervention and 161 families (n = 188 infants) received care-as-usual. There was no significant adjusted difference between treatment groups on dysregulation (0.2; 95% CI -2.5 to 3.0, P = .9) externalizing (0.3; 95% CI 1.6 to 2.2, P = .8), internalizing (-1.5; 95% CI -4.3 to 1.3, P = .3), observed aversive (0.00; -0.04 to 0.04, P = .9), or nonaversive behavior (-0.01; 95% CI -0.05 to 0.03, P = .7). Intervention children scored significantly higher on cognition (3.5; 95% CI 0.2-6.8, P = .04) and motor skill (5.5; 95% CI 2.5-8.4, P < .001, and approached significance on language (3.8; 95% CI 0.3 to 7.9, P = .07). CONCLUSIONS: Baby Triple P for Preterm Infants increases cognitive and motor skills but does not impact behavior. The results are evidence that hospital-based interventions can improve some developmental outcomes for infants <32 weeks. TRIAL REGISTRATION: ACTRN 12612000194864.

PMID: 30857773

25. Feasibility of parent communication training with remote coaching using smartphone apps.
Stockwell K, Alabdulqader E, Jackson D, Basu A, Olivier P, Pennington L.


BACKGROUND: Communication training for parents of young children with neurodisability is often delivered in groups and includes video coaching. Group teaching is problematic when there is wide variation in the characteristics and needs amongst participants. AIMs: To assess the potential feasibility and acceptability of delivering one-to-one parent training supported by remote coaching using smartphone apps and of conducting further trials of the intervention. METHODS & PROCEDURES: We aimed to recruit eight children aged 12-48 months with motor disorders and communication difficulties and to provide families with individual parent training in six weekly home visits supplemented by remote coaching via smartphone apps. For outcome measurement, parents recorded their interaction with their child thrice weekly during baseline (3 weeks), intervention (3 weeks) and follow-up (1 week). Measures comprised parent responsiveness and counts of children's communication and vocalization. Research design feasibility was measured through rates of recruitment, attrition, outcome measure completion and agreement between raters on outcome measurement. Intervention feasibility was assessed through the proportion of therapy sessions received, the number of videos and text messages shared using the apps in remote coaching, and message content. Parents were interviewed about the acceptability of the intervention and trial design. Interviews were transcribed and analyzed using inductive thematic analysis. OUTCOMES & RESULTS: Nine children were recruited over 16 weeks. All fitted the inclusion criteria. Four families withdrew from the study. Five families completed the intervention. No family submitted the target number of video recordings for outcome measurement. Interrater agreement was moderate for child communication (K = 0.46) and vocalization (K = 0.60) and high for The Responsive Augmentative and Alternative Communication Style scale (RAACS) (rs = 0.96). Parents who completed the intervention reported positive experiences of the programme and remote coaching via the apps. Therapist messages via the app contained comments on parent and child behaviour and requests for parental reflection/action; parental messages contained reflections on children's communication. CONCLUSIONS & IMPLICATIONS: The intervention and study design demanded high levels of parental involvement and was not suitable for all families. Recording shorter periods of interaction via mobile phones or using alternative methods of data collection may increase feasibility of outcome measurement.

PMID: 30851010

Kavlak E, Tekin F.
BACKGROUND AND OBJECTIVE: Cerebral palsied children may have difficulties in acting as senders and/or receivers in the communication process. The aim of this study is examining that which maternal and child-related factors affect the communication skills of cerebral palsied children. METHODS: 188 cerebral palsy diagnosed children ages between 2-18 years were assessed by Communication Function Classification System for communication skills. Maternal factors such as occupation, and educational status, and child-related factors such as gender, time of birth, clinical type of cerebral palsy, origin period of cerebral palsy; also daily living activities and gross motor functions of children were included in the assessment for examining how they affect the communication skills of cerebral palsied children. RESULTS: Lower maternal age, higher gross motor function level, ataxic type and hemiparetic involvement (p< 0.05); educational status, maternal unemployment, female gender, and premature birth (p > 0.05) affected positively on the communication skills. There were no effects of delivery method (p > 0.05). CONCLUSIONS: Communication skills of cerebral palsied children are affected by maternal age, educational status, occupation, and child's gender, birth term, origin period of cerebral palsy, clinical type of cerebral palsy, extremity involvement, motor development level and gross motor function. But the method of delivery has no effect on the communication functions of cerebral palsied children.

PMID: 30856125


Millions of people worldwide are afflicted with paralysis from a disruption of neural pathways between the brain and the muscles. Because their cortical architecture is often preserved, these patients are able to plan movements despite an inability to execute them. In such people, brain machine interfaces have great potential to restore lost function through neuroprosthetic devices, circumventing dysfunctional corticospinal circuitry. These devices have typically derived control signals from the motor cortex (M1) which provides information highly correlated with desired movement trajectories. However, sensorimotor control simultaneously engages multiple cognitive processes such as intent, state estimation, decision making, and the integration of multisensory feedback. As such, cortical association regions upstream of M1 such as the posterior parietal cortex (PPC) that are involved in higher order behaviors such as planning and learning, rather than in encoding movement itself, may enable enhanced, cognitive control of neuroprosthetics, termed cognitive neural prosthetics (CNPs). We illustrate in this review, through a small sampling, the cognitive functions encoded in the PPC and discuss their neural representation in the context of their relevance to motor neuroprosthetics. We aim to highlight through examples a role for cortical signals from the PPC in developing CNPs, and to inspire future avenues for exploration in their research and development.

PMID: 30872993


Cerebral palsy (CP) is a neurodevelopmental disorder that results in functional motor impairment and disability in children. CP is characterized by neural injury though many children do not exhibit brain lesions or damage. Advanced structural MRI measures may be more sensitively related to clinical outcomes in this population. Magnetic resonance elastography (MRE) measures the viscoelastic mechanical properties of brain tissue, which vary extensively between normal and disease states, and we hypothesized that the viscoelasticity of brain tissue is reduced in children with CP. Using a global region-of-interest-based analysis, we found that the stiffness of the cerebral gray matter in children with CP is significantly lower than in typically developing (TD) children, while the damping ratio of gray matter is significantly higher in CP. A voxel-wise analysis confirmed this finding, and additionally found stiffness and damping ratio differences between groups in regions of white matter. These results indicate that there is a difference in brain tissue health in children with CP that is quantifiable through stiffness and damping ratio measured with MRE. Understanding brain tissue mechanics in the pediatric CP population may aid in the diagnosis and evaluation of CP.

PMID: 30870734
29. Associations between preterm infant stress, epigenetic alteration, telomere length and neurodevelopmental outcomes: A systematic review.
Casavant SG, Cong X, Moore J, Starkweather A.


BACKGROUND: Every year, an estimated 15 million babies are born preterm (<37 weeks' gestational age [GA]) globally. These preterm infants are exposed to repeated stressful and often painful procedures as part of routine life-saving care within the neonatal intensive care unit (NICU). Preterm birth continues to be a major health issue associated with increased risk of neurodevelopmental and behavioral disorders such as cerebral palsy, cognitive impairment, autism spectrum disorders and psychiatric disease. OBJECTIVE: This paper identifies epigenetic alterations and incidence of telomere erosion that have been studied in preterm infants while in the NICU and as a long-term outcome measure. Better understanding of epigenetic alterations and telomere erosion might aid in early detection and prevention/alleviation of the negative effects of cumulative painful/stressful experiences in this population. METHODS: The Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) standards were used to guide this review. Systematic searches of databases included PubMed, CINAHL, SCOPUS and PsychInfo. RESULTS: Twenty-one studies were included, appraised and then synthesized into a narrative summary. DISCUSSION: Several putative epigenetic markers were identified although there was a paucity of studies related to telomere length. The interaction of disease entity combined with therapeutic interventions intended to treat may inadvertently increase infant allostatic load or ability to adapt to stress. Future research should include not only human studies but leverage newly available large data sets to conduct additional analysis.

PMID: 30870624


AIM: To clarify the frequency of occurrence of uterine rupture and its prognosis, a nationwide survey was performed. METHODS: Cases of uterine rupture recorded for a period of 5 years were included. RESULTS: There were 152 cases of uterine rupture with an incidence rate of 0.015%. The scarred uterine rupture cases were found to have a significantly earlier occurrence of uterine ruptures in comparison to the unscarred cases: unscarred 39.0 weeks, cesarean section 37.0 weeks, myomectomy 32 weeks and adenomyomectomy 30-32 weeks. And it became apparent that the frequency of hysterectomy, cerebral palsy and neonatal death were higher in the cases of uterine rupture during labor than before delivery. Among the cases of scarred uterine rupture, neonatal prognosis was poorer in cases of pregnancy after myomectomy or adenomyomectomy in comparison with postcesarean section cases. CONCLUSION: This survey revealed the current incidence of uterine rupture in Japan.

PMID: 30854725

Lewis MA.


This case study will focus on the "evolution" of finding meaningful occupation for a young man, Patrick, 27, in spite of his multiple disabilities within his rural home environment in Stowe, Vermont. The purpose of this case study is to show others (family members, therapists, support service individuals and adult agencies) that it is possible to be self-employed despite life's challenges, and that with some creativity (and funding), there is a niche of meaningful occupation available for all!

PMID: 30856141
Prevention and Cure

32. Apolipoprotein E allelic variants and cerebral palsy.
Gümuş E, Aras BD, Çilingir O, Yarar C, Çarman KB, Laçiner-Gürlevik S, Koçak O, Artan S.


Gümuş E, Aras BD, Cilingir O, Yarar C, Carman KB, Lacinier-Gurlievik S, Kocak O, Artan S. Apolipoprotein E allelic variants and cerebral palsy. Turk J Pediatr 2018; 60: 361-371. Cerebral palsy (CP) is the most frequent cause of mobility restriction and posture disturbance in childhood. Against the complexity in disease etiology, genetic factors, including Apolipoprotein E allelic distribution in this patient population, are worthy targets for investigation. ApoE is a lipoprotein of central nervous system encoded by ApoE gene with its 3 main co-dominant alleles, 2, 3 and 4. We aimed to evaluate the allelic frequencies of ApoE gene and its association with coexisting clinical entities such as vision and hearing impairment, cognitive problems, seizures and MRI findings in a pediatric patient population native to middle Anatolian region. Seventy-eight children with CP and 60 healthy controls were genotyped. Genotypic variations along with coexisting clinical conditions and CP-related medical findings were compared between the patient and control groups. The Denver Developmental Screening Test for all, the Wechsler Intelligence Scale for Children-IV (short form WISC-IV; Turkish version) for the patients > 6y and the Stanford-Binet Intelligence Scale (SB-5) for those who aged 2-6 years old were employed to evaluate cognitive and mental abilities of the patients. ApoE 2 and 4 alleles were more frequent in the patient group (p < 0.05), whereas ApoE 3 allele was more frequent in the healthy controls. ApoE 2/4 genotype has been determined 29% in the case group, but none in healthy control group. In the patient group with apolipoprotein 4 or 2 alleles, the rate of emergency cesarean section was found being significantly higher than the group with 3 allele. Brain MRI findings were not significantly different among ApoE allelic variants within the patient group. Our data show that the ApoE alleles may be effective in the development of cerebral palsy and may be associated with some clinical manifestations in those patients.

PMID: 30859759

33. Intrathecal Autologous Bone Marrow-Derived Hematopoietic Stem Cell Therapy in Neurological Diseases.


BACKGROUND: Cellular transplantation is a promising treatment strategy for neurological diseases. OBJECTIVE: To report the results of intrathecal hematopoietic stem cell therapy in different neurological diseases in the past 6 years in a single center. METHODS: From October 2011 to September 2018, 220 patients with various neurological diseases were transplanted intrathecally by their bone marrow stem cells. To have a longer follow up, we only reported the first 80 patients, transplanted up to July 2015-10 patients had spinal cord injuries and paralysis, 12 had advanced Parkinson's disease, 28 had cerebral palsy, 7 had hypoxic brain damage, 2 had autism, 4 had multiple sclerosis, 5 had progressive cerebellar atrophy, and 12 had other neurological diseases. The patients were admitted to the Bone Marrow Transplant Unit. On the first day, 50-200 (median 100) mL bone marrow was aspirated from the patients' posterior iliac crests, mixed with 120 mL culture media (RPMI), and 12 mL heparin. The samples were then transferred to immunology lab in cold box. Mononuclear cells (MNCs) were separated by a Ficoll-Hypaque gradient, washed, and suspended in ringers. Cell viability was assessed with trypan blue viability test. Transplantation was performed 3-4 hours after bone marrow collection. 5-10 mL of the cerebrospinal fluids were aspirated and about 20 mL MNCs (containing stem cells) in ringers were injected intrathecally (IT). The patients were laid down on their back for 4-5 hours. The median number of MNCs was 4×10^7 (range 1-450×10^7). The median viability of the cells was 90% (range 60%-98%). The patients received intravenous ceftriaxone every 12 hours and were discharged from the hospital few days after autologous stem cell therapy. RESULTS: We noted clinical improvements in 9 of 12 patients with Parkinson's disease, 20 of 28 patients with cerebral palsy, 6 of 7 patients with hypoxic brain damage, 2 of 4 patients with multiple sclerosis, and 4 of 5 patients with cerebellar atrophy. The improvements were noted after 2-4 weeks of cell therapy. There were no improvements in patients with spinal cord injury and complete paralysis and those with autism. There were variable improvements in other patients treated. CONCLUSION: Most patients with advanced Parkinson's disease, cerebral palsy, hypoxic brain damage, progressive cerebellar atrophy, and kernicterus neuropathy reported clinical effects of this safe intervention resulting in better functioning and an increased quality of life.

PMID: 30863518