1. Botulinum toxin and surgical intervention in children and adolescents with cerebral palsy: who, when and why do we treat?

Introduction: This audit aimed to increase understanding of the long-term outcomes of evidence-based medical and surgical interventions to improve gross motor function in children and adolescents with Cerebral Palsy. Methods: Retrospective audit of a birth cohort (2000-2009) attending a tertiary service in Western Australia. Results: The cohort comprises 771 patients aged 8 to 17 years. Percentage of children receiving no Botulinum Toxin treatments in each Gross Motor Functional Classification System level was: I: 40%, II: 26%, III: 33%, IV: 28% and V: 46%. Of the total cohort, 53% of children received 4 or less Botulinum Toxin treatments and 3.7% received more than 20 treatments. Statistically significant difference in the rate of use of Botulinum Toxin pre and post-surgery (p < 0.001) was documented. Children levels IV and V had 5 times the odds of surgery compared to children levels I-III (Odds Ratio 5.2, 95% Confidence Interval 3.5 to 7.8, p < 0.001). For 578 (75%) of participants the last recorded level was the same as the first. Conclusion: This audit documents medical intervention by age and Gross Motor Functional Classification System level in a large cohort of children with cerebral palsy over time and confirms stability of the level in the majority. IMPLICATIONS FOR REHABILITATION The information from this audit may be of use in discussions with families regarding the timing and use of Botulinum toxin and surgical intervention for motor function in children and adolescents with Cerebral Palsy. Long term use of Botulinum Toxin within an integrated evidence-based clinical program is not associated with loss of gross motor function in the long term as evidenced by the maintenance of Gross Motor Functional Classification System stability.

PMID: 31415723

2. Sarcopenia, Cerebral Palsy, and Botulinum Toxin Type A.

PMID: 31415277

3. Intrathecal baclofen therapy for treatment of spasticity in infants and small children under 6 years of age.
Hagemann C, Schmitt I, Lischetzki G, Kunkel P.
PURPOSE: The aim of this study is to prove the efficacy and safety of intrathecal baclofen therapy in infants and children below 6 years of age by retrospective analysis of our pediatric cohort of 135 primary pump implantations. METHODS: Between 2007 and 2018, 17 patients with pump implantations were below 6 years of age. Data were acquired retrospectively with a follow-up of 12 months to 11 years regarding complications. RESULTS: The youngest infant was 11 months at implantation with a bodyweight of 6.4 kg, and 63 cm length. Surgical complications were comparable to published literature and mainly involved the catheter (2 catheter dislocations and 1 catheter transection) and one pump infection resulting in 4 revision surgeries in 3 patients. One baclofen-related apnea during titration and an overdose after refill were treated conservatively. In a growing child, catheter slides are common and related to growth, scoliosis, spine surgery, and surgical failure. CONCLUSION: Intrathecal baclofen therapy in infants and small children is as safe and effective as published for older pediatric patients; therefore, intrathecal baclofen can be considered in all infants as long as an 8-cm incision fits into the triangle of the anterior superior iliac spine, costal margin of the 10th rib, and navel. We suggest the utilization of subfascial surgical technique for implantation pump and catheter. Titration of intrathecal baclofen should be performed slowly to avoid bradycardia in infants. This is a retrospective study (level of evidence 4).

PMID: 31399764


Cerebral palsy (CP) is the most common motor disability in children. Much of the previous research on CP has focused on reducing the severity of brain injuries, while very few researchers investigate the cause and amelioration of motor symptoms. This research focus has had an impact on the choice of animal models. Many of the commonly used animal models do not display a prominent CP-like motor phenotype. In general, rodent models show anatomically severe injuries in the central nervous system (CNS) in response to insults associated with CP including hypoxia, ischemia and neuroinflammation. Unfortunately, most rodent models do not display a prominent motor phenotype that includes the hallmarks of spasticity (muscle stiffness and hyperreflexia) and weakness. In order to study motor dysfunction related to developmental injuries, a larger animal model is needed such as rabbit, pig or non-human primate. In this work we describe and compare various animal models of CP and their potential for translation to the human condition.

PMID: 31411933

5. Improved Clinical and Functional Outcomes in Crouch Gait Following Minimally Invasive Hamstring Lengthening and Serial Casting in Children With Cerebral Palsy. Long JT, Cobb L, Garcia MC, McCarthy JJ.


BACKGROUND: Serial extension casting represents a novel solution for addressing residual knee flexion contractures following hamstring lengthening in children with cerebral palsy. The purpose of this study was to investigate postoperative changes in patients following hamstring lengthening with a serial casting protocol. METHODS: Measures from preoperative and postoperative gait analyses were reviewed retrospectively for 19 patients with cerebral palsy who underwent hamstring lengthening followed by serial extension casting. Postoperative changes in clinical, functional, and kinematic parameters were assessed using paired parametric methods. RESULTS: Improvements were measured in popliteal angle, knee contracture, peak stance phase knee extension, sagittal plane range of motion of the knee during walking, Gait Deviation Index, and pediatric outcomes data collection instrument Global score. Nearly 80% of the cohort (15/19 patients) demonstrated a significant or moderate response to the intervention, whereas 20% demonstrated no improvement. Of note, significantly increased anterior pelvic tilt was also observed. CONCLUSIONS: Hamstring lengthening combined with a serial casting protocol was associated with significant postoperative improvements in a range of clinical (eg, knee contracture), functional (eg, pediatric outcomes data collection instrument Global), and kinematic (eg, knee extension in stance) parameters. Improvements following this minimally invasive surgery were comparable to outcomes from procedures with higher complication rates. LEVEL OF EVIDENCE: This is a Level III Therapeutic Study (retrospective study investigating the results of a treatment).

PMID: 31403992
Lee SJ, Jin D, Kang SH, Gaebler-Spira D, Zhang LQ.

Although various treatment methods have been investigated to reduce spasticity and in-toeing gait in children with cerebral palsy (CP), methods to concurrently reduce an in-toeing gait and associated ankle/knee stiffness and spasticity according to a child's specific needs are lacking. This study aimed to develop a training program to improve walking function and transverse-plane (pivoting) neuromuscular control and reduce spasticity and in-toeing gait deviations. METHODS: Eight children with diplegic CP and in-toeing gait participated in this 6-week combined robotic ankle and/or knee intelligent stretching and pivoting neuromuscular control training program (Subject-specific Stretching and Pivoting Off-axis Neuromuscular control Training, SS-POINT). The effects of SS-POINT were evaluated using neuromechanical, functional, and clinical outcome measures and compared to those for eight children with CP and in-toeing gait who participated in pivoting neuromuscular control training (POINT) alone in a previous study. RESULTS: After the SS-POINT program, subjects with CP showed reduced knee stiffness and in-toeing angle, and improvements in both joint and leg functions in terms of ankle and knee active range of motion, ankle dorsiflexor strength, proprioception, walking speed, balance, and minimum pivoting angle. Furthermore, improvements in proprioceptive acuity and minimum pivoting angle after the SS-POINT were greater than those after the POINT.
CONCLUSION: The SS-POINT approach can be used as a subject-specific training program for improving leg and walking functions and reducing in-toeing during gait. SIGNIFICANCE: This approach can serve as an individualized intervention at the joint and walking levels to maximize intervention effects by adjusting training targets, sequences, and intensities to meet the individual needs of children with CP.

PMID: 31403432


Foot drop is one of the most common secondary conditions associated with hemiplegia post stroke and cerebral palsy (CP) in children, and is characterized by the inability to lift the foot (dorsiflexion) about the ankle. This investigation focuses on children and adolescents diagnosed with brain injury and aims to evaluate the orthotic and therapeutic effects due to continuous use of a foot drop stimulator (FDS). Seven children (10 ± 3.89 years) with foot drop and hemiplegia secondary to brain injury (stroke or CP) were evaluated at baseline and after 3 months of FDS usage during community ambulation. Primary outcome measures included using mechanistic (joint kinematics, toe displacement, temporal-spatial asymmetry), and functional gait parameters (speed, step length, time) to evaluate the orthotic and therapeutic effects. There was a significant correlation between spatial asymmetry and speed without FDS at 3 months (r = 0.76, p < 0.05, df = 5) and no correlation between temporal asymmetry and speed for all conditions. The results show orthotic effects including significant increase in toe displacement (p < 0.025 N = 7) during the swing phase of gait while using the FDS. A positive correlation exists between toe displacement and speed (with FDS at 3 months: r = 0.62, p > 0.05, without FDS at 3 months: r = 0.44, p > 0.05). The results indicate an orthotic effect of increased dorsiflexion and toe displacement during swing with the use of the FDS in children with hemiplegia. Further, the study suggests that there could be a potential long-term effect of increased dorsiflexion during swing with continuous use of FDS.

PMID: 31417338

Whitney DG.

BACKGROUND: Adults with cerebral palsy (CP) have increased risk for skeletal fragility and osteoarthritis. However, racial differences in these outcomes have not been examined. Such knowledge could improve patient-specific clinical care for the prevention and management of these conditions. The purpose of this study was to determine if there were racial differences in the prevalence of osteoporosis, all-cause fracture, and osteoarthritis among young and middle-aged adults with CP.
METHODS: Data from 2016 were extracted from a random 20% sample of the Medicare fee-for-service database. International Classification of Diseases, Tenth Revision, Clinical Modification codes were used to identify 18-64 year olds with CP, as well
as osteoporosis, all-cause fracture, osteoarthritis, and neurodevelopmental and noncommunicable disease comorbidities.

RESULTS: Of the 16,488 adults with CP, 13,334 were White, 2477 were Black, and 677 were Hispanic. The age-standardized prevalence of osteoporosis (women: 12.9%, 9.0%, 8.3%, respectively; men: 9.2%, 4.8%, 7.9%, respectively) and fracture (women: 7.4%, 4.2%, 9.9%; men: 6.0%, 2.3%, 6.0%) was lower for Black adults with CP compared to White adults with CP (all p < 0.005). No racial differences were observed for age-standardized prevalence of osteoarthritis (women: 13.6%, 14.4%, 9.6%; men: 10.7%, 10.4%, 12.7%). The racial differences between Black and White adults with CP remained even after adjusting for age, U.S. region, neurodevelopmental comorbidities, and several noncommunicable diseases for osteoporosis (women: OR = 0.66, 99.5% CI = 0.48-0.91; men: OR = 0.51, 99.5% CI = 0.35-0.75) and fracture (women: OR = 0.57, 99.5% CI = 0.37-0.89; men: OR = 0.39, 99.5% CI = 0.23-0.68). CONCLUSIONS: Study findings suggest racial differences in skeletal fragility among young and middle-aged adults with CP, with White women and men with CP having greater risk compared to Black women and men with CP. This study found no evidence of racial differences in the prevalence of osteoarthritis.

PMID: 31417943


INTRODUCTION: To investigate the effectiveness of electromyography (EMG) biofeedback therapy in improving motor dysfunction among children with cerebral palsy (CP). METHODS AND ANALYSIS: The following databases will be searched: PubMed, EMBASE, ScienceDirect, the Cochrane Library, China National Knowledge infrastructure (CNKI), Technology Periodical Database (VIP), WanFang Data and China Biology Medicine (CBM) from inception to June 2019. All relevant randomized controlled trials (RCTs) utilizing EMG biofeedback therapy for CP will be included. The main outcome is the Gross Motor Function Measure (GMFM). Additional outcomes such as the Modified Ashworth Scale (MAS), Integral Electromyogram (iEMG), Composite Spasticity Scale (CSS), passive range of motion (PROM) or other related outcomes will be included, adverse effects of EMG biofeedback therapy and comparators will also be included. Two reviewers will screen studies, extract data and assess quality independently. Review Manager 5.3 will be used to assess the risk of bias, data synthesis, and subgroup analysis. ETHICS AND DISSEMINATION: This systematic review does not require formal ethical approval because all data will be analyzed anonymously. Results will provide a general overview and evidence concerning the effectiveness and safety of EMG biofeedback therapy for children with CP. The findings of this systematic review will be disseminated through peer-reviewed publications or conference presentations.

PMID: 31415383

10. [Specifics of spa-based rehabilitation of severe forms of spastic mobility loss of patients with cerebral palsy].

Osmanov EA, Larina NV, Vlasenko SV, Golubova TF, Marusich II, Kushmir GM.


AIM: To study the efficacy of the developed approach to the specialized Spa-rehabilitation of motor disorders in patients with cerebral palsy (spastic diplegia). MATERIAL AND METHODS: The study included 86 patients (45 (52,33%) women and 41 (47.67%) men), aged from 10 to 16-years, mean 13,02±2,13. All patients were divided into 2 groups depending on the method of treatment. RESULTS AND CONCLUSION: Administration of minimally invasive procedures during the course of rehabilitation can effectively restore the volume of movements in the limbs, stimulate motor development in children and form a new stereotype of walking. An analysis of the follow-up data one year after the treatment confirms the efficacy of the proposed approach to rehabilitation.

PMID: 31407681


Diagnosis of cerebral palsy (CP) after perinatal stroke is often delayed beyond infancy, a period of rapid neuromotor development with heightened potential for rehabilitation. This study sought to assess whether the presence or absence of motor evoked potentials (MEPs) elicited by transcranial magnetic stimulation (TMS) could be an early biomarker of atypical development within the first year of life. In 10 infants with perinatal stroke, motor outcome was assessed with a standardized movement assessment. Single-pulse TMS was utilized to assess presence of MEPs. Younger infants (3-6 months CA, n = 5, 4/5 (80%)) were more likely to present with an MEP from the more-affected hemisphere (MAH) compared to older infants (7-12 months CA, n = 5, 0/5, (0%)) (p = 0.048). Atypical movement was demonstrated in the majority of infants with an absent MEP from the MAH (5/6, 83%) compared to those with a present MEP (1/4, 25%) (p = 0.191). We found that age influences the ability to elicit an MEP from the MAH, and motor outcome may be related to MAH MEP absence. Assessment of MEPs in conjunction with current practice of neuroimaging and motor assessments could promote early detection and intervention in infants at risk of CP.

PMID: 31412592

Garfinkle J, Li P, Boychuck Z, Bussières A, Majnemer A.

BACKGROUND: The early identification of cerebral palsy (CP) in the primary care context is often problematic and referral for diagnosis often delayed. This study aimed to identify clinical features associated with the early detection of CP that can be used by the primary care provider. METHODS: We performed a scoping review by searching six electronic databases. We included English language articles that addressed the diagnosis of CP and/or its differential diagnosis in children and ways of detecting CP before the diagnosis is established (i.e., early clinical signs of CP) via (1) questions on the patient's clinical history, (2) developmental screening and/or health questionnaires, or (3) physical or neurological examination. RESULTS: Included studies (n = 41; 27 overview studies and 14 original studies) were grouped into the three themes. Most of the overview articles relied on expert opinion, and all original studies included patients at high risk of developing CP. The most commonly identified features from each theme were early hand preference on clinical history, delayed or absent achievement of motor developmental milestones on developmental screening, and persistent primitive reflexes on neurological examination. CONCLUSIONS: Overall, the literature on the early observable clinical signs that should prompt referral for investigation of possible CP in the specific context of well-baby care surveillance was sparse and inconsistent. Further research should focus on evaluating the contribution of readily identifiable clinical features.

PMID: 31416726

13. Motor and Postural Patterns Concomitant with General Movements Are Associated with Cerebral Palsy at Term and Fidgety Age in Preterm Infants.
Ferrari F, Plessi C, Lucaccioni L, Bertoncelli N, Bedetti L, Ori L, Berardi A, Della Casa E, Iughetti L, D'Amico R.

General movements (GMs) in combination with neurological examination and magnetic resonance imaging at term age can accurately determine the risk of cerebral palsy. The present study aimed to assess whether 11 motor and postural patterns concomitant with GMs were associated with cerebral palsy. Video recordings performed after birth in 79 preterm infants were reviewed retrospectively. Thirty-seven infants developed cerebral palsy at 2 years corrected age and the remaining 42 showed typical development. GMs were assessed from preterm to fidgety age and GM trajectories were defined. The 11 motor and postural patterns were evaluated at each age and longitudinally, alone and in combination with GM trajectories. A logistic regression model was used to assess the association between GMs, concomitant motor and postural patterns, and cerebral palsy. We confirmed that high-risk GM trajectories were associated with cerebral palsy (odds ratio = 44.40, 95% confidence interval = 11.74-167.85). An association between concomitant motor and postural patterns and cerebral palsy was found for some of the patterns at term age and for all of them at fidgety age. Therefore, at term age, concomitant motor and postural patterns can support GMs for the early diagnosis of cerebral palsy.

PMID: 31398881
Beckers LWME, Stal RA, Smeets RJEM, Onghena P, Bastiaenen CHG.

Aim: To critically evaluate single-case design (SCD) studies performed within the population of children/adolescents with cerebral palsy (CP). Methods: A scoping review of SCD studies of children/adolescents with CP. Demographic, methodological, and statistical data were extracted. Articles were evaluated using the Risk of Bias in N-of-1 Trials (RoBiNT) Scale and the Consolidated Standards of Reporting Trials (CONSORT) extension for N-of-1 trials (CENT 2015). Comments regarding strengths and limitations were analyzed. Results: Studies investigated the effects of a wide range of interventions on various outcomes. Most SCD types were adopted in multiple studies. All studies used visual inspection rather than visual analysis, often complemented with basic statistical descriptors. Risk of bias was high, particularly concerning internal validity. Many CENT items were insufficiently reported. Several benefits and limitations of SCD were identified. Conclusions: The quality of evidence from results of SCD studies needs to be increased through risk of bias reduction.

PMID: 31411523

15. Evidence of Construct Validity for the Modified Mental Fatigue Scale When Used in Persons with Cerebral Palsy.
Bergqvist L, Öhrvall AM, Rönnbäck L, Johansson B, Himmelmann K, Peny-Dahlstrand M.

Introduction: Fatigue impacts negatively on everyday activities in individuals with cerebral palsy (CP). More knowledge is needed about how mental fatigue is manifested in this target group. The purpose of this study was to gather evidence about the validity of the modified Mental Fatigue Scale (m-MFS) in adults with CP. Methods: Mixed sequential exploratory design. The respondents were ten persons aged 22-56 with CP (MACS I-II). Results: The respondents perceived the m-MFS as easy to read and understand. Its structure was characterised as straightforward and the text of the rating options was deemed to assist identification with life situations. Very good agreement was seen between the respondents' and the instrument designers' intended meaning for the items in the m-MFS; the weighted kappa was 0.92. Conclusion: This study showed evidence of construct validity, based on response processes and content, for use of the modified MFS in adults with CP.

PMID: 31403825

Whitney DG, Kamdar NS, Ng S, Hurvitz EA, Peterson MD.

Purpose: Individuals with cerebral palsy (CP) are susceptible to early development of high-burden medical conditions, which may place a considerable strain on health care resources. However, little is known about the prevalence of high-burden medical conditions or health care resource utilization among adults with CP. The purpose of this study was to determine the prevalence of high-burden medical conditions and health care resource utilization and costs among adults with CP, as compared to adults without CP. Patients and methods: Cross-sectional data from the 2016 Optum Clininformatics® Data Mart, a de-identified nationwide claims database of beneficiaries from a single private payer in the US. ICD-10-CM diagnosis codes were used to identify all medical conditions among beneficiaries with and without CP who were between 18 and 64 years of age. Medical and outpatient pharmacy claims were used to identify annual all-cause health care resource utilization and health care costs as standardized reimbursement and patient out-of-pocket costs. Results: Adults with CP (n=5,555) had higher prevalence and odds of all medical conditions compared to adults without CP (OR=1.3-5.8; all P<0.05), except cancer (OR=1.1; 95% CI=0.9-1.3). Adults with CP had greater annual mean counts of all health care service types (eg, inpatient, emergency department) compared to adults without CP (all P<0.01). Adults with CP had higher unadjusted standardized reimbursement (mean difference=$16,288; cost ratio [CR]=2.6) and patient out-of-pocket (mean difference=$778; CR=1.7; 95% CI=1.6-1.7) costs. Conclusions: Adults with CP have a higher prevalence of high-burden medical conditions, health care resource utilization, and health care costs compared to adults without CP. Study findings suggest the need for earlier screening strategies and preventive medical services to quell the disease and economic burden attributable to adults with CP.

PMID: 31417318