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Professor Nadia Badawi AM
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Interventions and Management

AlHakeem N, Ouellette EA, Travascio F, Asfour S.


BACKGROUND: Spasticity of the upper extremity often occurs after injury to the upper motor neurons (UMN). This condition can greatly interfere with the hand positioning in space and the functional use of the arm, affecting many daily living activities including walking. As gait and balance involve the coordination of all segments of the body, the control of upper limbs movement is necessary for smooth motion and stability. The purpose of this study was to assess the effects of surgical interventions on upper extremity spasticity to gait patterns in three spastic patients, as a way to assess the effect on patient's mobility. METHODS: Three patients with an anoxic brain injury, upper extremity spasticity, and an altered gait participated in this study. A specific treatment plan based on the patient was tailored by the orthopedic hand surgeon to help release the contractures and spastic muscles. Three-dimensional gait analysis was performed before surgery, 3, 6, and 12 months postoperatively. During each experimental session, the patient walked at a self-selected pace in a straight line across four force plates embedded into the floor (Kistler®). Motion data were acquired using Vicon® Motion Capturing System. Spatiotemporal measurements as well as bilateral kinematics of the hip, knee and ankle were studied. The results from matched non-disabled controls were included as reference. RESULTS: Overtime, clinical assessment displayed recovery in hand functions and restored sensation in the fingers. Gait analysis results demonstrated overall improvements in spatiotemporal parameters, specifically in cadence and walking speed. Improvements in kinematics of the lower limbs were also evident. CONCLUSION: The results of this study indicated that, within a timeframe of one year, gait patterns improved in all patients. These observations suggest that, over time, upper limb surgery has the potential to improve the biomechanics of gait in spastic patients.

PMID: 32154240

2. Technical aspects of surgical correction of spinal deformities in cerebral palsy.
Mineiro J, Yazici M.


PURPOSE: Cerebral palsy (CP) is a disorder arising from a non-progressive lesion in the developing immature brain with an encephalopathy, that results in various levels of motor and sensory dysfunction. Motor disability of these children can be assessed by the Gross Motor Function Classification System in five levels, and depending on their motor functional capability, the most severely affected children fall into levels IV and V. Children in groups IV and V present a full spectrum of musculoskeletal deformities, among which, scoliosis is the most frequently found spinal deformity that most often requires
surgical treatment. However, these are procedures that are usually technically demanding, requiring experienced surgical teams and a multidisciplinary approach. METHODS: In order to overcome some of the technical pitfalls that may complicate these complex surgical procedures, the authors have gathered together different tips and tricks that may help surgeons performing surgical correction of spinal deformities in CP children. CONCLUSION: Although for these children surgery is a major undertaking, with the multidisciplinary approach and advances of technology, anaesthesia and optimization of pre- and postoperative care, complications are manageable in most cases, improving not only the outcome of surgery but also the patient's quality of life and satisfaction of parents and caretakers.

PMID: 32165979

Ilharreborde B, de Saint Etienne A, Presedo A, Simon AL.


PURPOSE: Spinal sagittal alignment restoration has been associated with improved functional outcomes and with reduced complications rates. Several limitations exist for radiological analysis in cerebral palsy (CP) patients. The goal of this study was to summarize the existing literature and report the important considerations to evaluate in a CP patient undergoing spinal surgery. METHODS: A retrospective radiological analysis was performed, including non-ambulant CP children with progressive scoliosis. Full-spine sitting radiographs performed pre-and postoperatively were required to measure spino-pelvic sagittal parameters. RESULT: A total of 23 non-ambulating CP patients were included, mean age 16.0 years (standard error of the mean 0.5). Two distinct groups of patients were identified. Group 1 (61%) were patients with less trunk control (lumbar lordosis (LL) < 50°), retroverted and vertical pelvis (mean sacral slope (SS) 11.4° and pelvic tilt (PT) 38.1°) and anterior imbalance (mean sagittal vertical axis (SVA) 5.9 cm) and Group 2 (39%) were patients with better trunk control (LL > 60°, antverted and horizontal pelvis (mean SS 49.3°, PT 9.7°) and posterior imbalance (mean SVA 5.8 cm). Postoperative measures showed significant impact of surgery with a PT reduction of 19° (p = 0.007), a mean SS increase of 15° (p = 0.04) and a LL gained of 10° (p = 0.2). CONCLUSION: Sagittal spino-pelvic alignment in non-ambulating CP patients remains difficult to assess. The current literature is poor but our radiological study was able to define two distinct groups among Gross Motor Function Classification System (GMFCS) level V patients, based on the quality of their trunk control. All possible factors that may influence head and trunk posture should be systematically considered and optimized. LEVEL OF EVIDENCE: Level IV.

PMID: 32165977

4. Spine deformities in patients with cerebral palsy; the role of the pelvis.
Hasler C, Brunner R, Grundshein A, Ovadia D.


INTRODUCTION: Progressive neuromuscular spinal deformities with pelvic obliquity and loss of sitting balance are typical features of severely affected patients with cerebral palsy. The pelvis represents the key bone between the spine and the lower extremity when it comes to deciding whether and when to operate and if spine or hip surgery first is beneficial. The pelvis can be looked at as the lowest vertebra and as the rooftop of the lower extremities. BIOMECHANICAL CONSIDERATIONS: To allow for a normal spinal shape, the pelvis needs to be horizontal in the frontal plane and mildly anterior tilted in the sagittal plane, less for sitting and more for standing. Any abnormal pelvic position requires spinal compensation and challenges the equilibrium control of the individual. Both anatomical neighbourhoods - the spine and the hip joints - have to be considered when spinal deformities, hip instability and contractures evolve, in conservative therapy (bracing, physiotherapy, seating in the wheelchair) and when surgical interventions are weighed out against each other. SURGICAL CONSIDERATIONS: Multiple anatomical factors such as sagittal profile and pelvic orientation, pelvic transverse plane asymmetries and lumbosacral malformations have to be considered in case the pelvis is instrumented with sacral and iliac screws. Rotational deformities and asymmetries of the pelvic bones make the safe insertion of long screws challenging. Advantages of primary pelvic fixation include correction of pelvic obliquity, especially considering the lever arm of the whole spinal construct. The risk of revision surgery due to progression of distal curves is also reduced. Disadvantages of pelvic fixation include the complexity of the additional intervention, which may result in longer operating times, increased risk of blood loss, infection and hardware malpositioning.

PMID: 32165976
Brunner R.


INCIDENCE AND CAUSE: Cerebral palsy (CP) is characterized by poor motor control. The more severe the affection is, the more patients are prone to deformities. Patients with Gross Motor Function Classification System level V run an up to 90% risk for spinal deformities. These are caused by poor trunk control under load. Although trunk tone is impossible to assess it seems to be low in the majority of patients, leading to collapse under gravity. The constant malposition results in growth asymmetry which leads to fixation and deterioration of the deformity. BRACE TREATMENT: Brace treatment has a poor reputation in respect to the final outcome. Conventional braces as constructed for other spinal deformities are indeed difficult for patients with CP as they cannot change position in the brace and do not tolerate pressure on the belly for reflux problems. Respecting these points improves the tolerance of braces but still the time of use is far from the necessary when the trunk is upright. Nevertheless, they can help to postpone surgery for scoliosis, but they are very inefficient for sagittal plane deformities. AIM OF TREATMENT: The lack of trunk control further leads to an impairment of head control and upper extremity function. Providing stability improves these problems. Braces are superior to seating shells for both treating deformity and providing stability as they remain close to the skin and follow movements. Supports on seating shells in contrast are too far away for controlling the deformity and provide stability only if the patient doesn't move forward.

PMID: 32165975

Hasler C, Brunner R.


PMID: 32165974

7. Reliability, standard error of measurement, and minimal detectable change of the star excursion balance test in children with cerebral palsy.
Kim DH, An DH, Yoo WG.


PURPOSE: The present study aimed to estimate the reliability, standard error of measurement (SEM), and minimum detectable change (MDC) of the star excursion balance test (SEBT) in children with cerebral palsy (CP). METHODS: Eight children with CP (four boys and four girls, sixteen legs) participated in this study. Each child carried out the SEBT and was assessed by two examiners. To determine intra-rater reliability, the intra-class correlation coefficient (ICC) model (3, 3) was calculated. To determine the inter-rater reliability, the ICC model (2, 3) was computed. RESULTS: In terms of the intra-rater reliability of the SEBT, the ICC varied from 0.98 to 0.99 and the total ICC score was 0.99 (p< 0.001). For the inter-rater reliability, the ICC varied from 0.98 to 1.00 and the total ICC score was 0.99 (p< 0.001). The SEBT had an SEM of 2.63 and an MDC of 7.31. CONCLUSION: The SEBT is not only reliable with a small SEM, but is also a simple and cheap assessment of dynamic balance in children with CP.

PMID: 32144971

Schitter AM, Fleckenstein J, Frei P, Taeymans J, Kurpiers N, Radlinger L.
BACKGROUND: WATSU (portmanteau word: water and shiatsu) is a form of passive hydrotherapy in chest-deep thermoneutral water (35°C = 95°F = 308.15 K). It combines elements of myofascial stretching, joint mobilization, massage, and shiatsu and is reported to be used to address physical and mental issues. The objective of this systematic review (PROSPERO Registration No. CRD42016029347) and the meta-analyses was to assess the applications, indications, and the effects of WATSU to form a basis for further studies. METHODS: A search for "WATSU OR watershiatsu OR (water AND shiatsu)" was conducted without any restrictions in 32 databases. Peer reviewed original articles addressing WATSU as a stand-alone hydrotherapy were assessed for risk of bias. Quantitative data of effects on pain, physical function, and mental issues were processed in random model meta-analyses with subgroup analyses by study design. Effect sizes were expressed as Hedges's g (± 95% confidence intervals). RESULTS: Of 1,906 unique citations, 27 articles regardless of study design were assessed for risk of bias. WATSU has been applied to individuals of all ages. Indications covered acute (e.g. pregnancy-related low back pain) and chronic conditions (e.g. cerebral palsy) with beneficial effects of WATSU regarding e.g. relaxation or sleep quality. Meta-analyses suggest beneficial effect sizes of WATSU on pain (overall Hedges's g = -0.71, 95% CI = -0.91 to -0.51), physical function (overall Hedges's g = -0.76, 95% CI = -1.08 to -0.44), and mental issues (overall Hedges's g = -0.68, 95% CI = -1.02 to -0.35). CONCLUSION: Various applications, indications and beneficial effects of WATSU were identified. The grade of this evidence is estimated to be low to moderate at the best. To strengthen the findings of this study, high-quality RCTs are needed.

PMID: 32168328

9. Cerebral palsy with dislocated hip and scoliosis: what to deal with first?
Helenius IJ, Viehweger E, Castelein RM.

PURPOSE: Hip dislocation and scoliosis are common in children with cerebral palsy (CP). Hip dislocation develops in 15% and 20% of children with CP, mainly between three and six years of age and especially in the spastic and dyskinetic subtypes. The risk of scoliosis increases with age and increasing disability as expressed by the Gross Motor Function Score. METHODS: A hip surveillance programme and early surgical treatment have been shown to reduce the hip dislocation, but it remains unclear if a similar programme could reduce the need for neuromuscular scoliosis. When hip dislocation and neuromuscular scoliosis are co-existent, there appears to be no clear guidelines as to which of these deformities should be addressed first: hip or spine. RESULTS: Hip dislocation or windswept deformity may cause pelvic obliquity and initiate scoliosis, while neuromuscular scoliosis itself leads to pelvic obliquity and may increase the risk of hip dislocation especially on the high side. It remains unclear if treating imminent hip dislocation can prevent development of scoliosis and vice versa, but they may present at the same time for surgery. Current expert opinion suggests that when hip dislocation and scoliosis present at the same time, scoliosis associated pelvic obliquity should be corrected before hip reconstruction. If the patient is not presenting with pelvic obliquity the more symptomatic condition should be addressed first. CONCLUSION: Early identification of hip displacement and neuromuscular scoliosis appears to be important for better surgical outcomes.

PMID: 32165978

10. Comparison between semitendinosus transfer to distal femur and medial hamstrings surgical lengthening for treatment of flexed knee gait in cerebral palsy.
de Morais Filho MC, Fujino MH, Blumetti FC, Dos Santos CA, Kawamura CM, Ramos BCA, Lopes JAF.

PURPOSE: Hamstrings surgical lengthening (HSL) has been frequently used for the treatment of flexed knee gait in cerebral palsy; however, recurrence of knee flexion deformity (KFD) and increase of anterior pelvic tilt (APT) were reported in a long-term follow-up. RESEARCH QUESTION: The aim of this study was to compare semitendinosus transfer to distal femur (STTX) and semitendinosus surgical lengthening (STL) regarding the reduction of KFD and the increase of APT after flexed knee gait treatment. METHODS: One hundred and eleven patients were evaluated and they were divided into two groups according to surgical procedures at knees: group A (65 patients/130 knees), including patients who received medial HSL as part of multilevel approach; group B (46 patients/92 knees), represented by patients who underwent orthopedic surgery...
including an STTX instead of STL. RESULTS: Fixed knee flexion deformity (FKFD) decreased only in group B (from 6.79° to 2.96°, p < 0.001) after intervention. In kinematics, APT increased from 16.38° to 19.03° in group A (p = 0.003), while group B also increased from 15.26° to 20.59° (p < 0.001). The minimum knee flexion in stance phase (MKFS) reduced from 25.34° to 21.65° (p = 0.016) in group A and from 31° to 19.57° (p < 0.001) in group B. In the comparison between groups A and B, the increase of APT (p = 0.028) and reduction of FKFD (p < 0.001), popliteal angle (p = 0.001), bilateral popliteal angle (p = 0.003) and MKFS (p = 0.006) were higher after STTX than STL. CONCLUSION: In the present study, patients who received STTX exhibited more improvement of knee extension at clinical examination and during gait than those who underwent to STL; however, STTX was not effective to prevent the increase of APT after flexed knee gait treatment.

PMID: 32167417


PURPOSE: Surgical procedures, such as medial hamstring lengthening (MHL) and femoral derotational osteotomy (FDO), can improve the gait of children with cerebral palsy (CP); however, substantial variation exists in the factors that influence the decision to perform surgery. The purpose of this study was to use expert surgeon opinion through a Delphi technique to establish consensus for indications in ambulatory children with CP. METHODS: A 15-member panel, all established experts with at least nine years' experience in the surgical management of children with CP, was created (mean of 20.81 years' experience). All panel members also had expertise of the use of movement analysis for the assessment of gait disorders in children with CP. The group initially focused on two of the most commonly performed procedures, MHL and FDO, in an attempt to gain consensus (> 80%). This was obtained through a standardized, iterative Delphi process. RESULTS: For MHL, a total of 59 questions were surveyed: 41 indication questions and 18 outcome questions, for which there was consensus on ten indication questions and seven outcomes. For FDO, a total of 55 questions were surveyed: 43 indication questions and 12 outcome questions, for which there was consensus on 29 indication questions and eight outcomes. CONCLUSION: This study is the first to use an expert panel to identify best-practice indications for common surgical procedures of children with CP. The results from this study will allow for more informed evaluation of practice and form the basis for future improvement efforts to standardize surgical recommendations internationally. LEVEL OF EVIDENCE: Level IV.

PMID: 32165981

Bonnefoy-Mazure A, De Coulon G, Lascombes P, Armand S.


PURPOSE: Assessment of surgical treatments on gait in patients with bilateral cerebral palsy (CP) is often performed in short-term studies. The purpose of this study was to analyze the influence of single-event multilevel surgery (SEMLS) on long-term evolution of gait using gait deviation index (GDI) and walking speed. METHODS: In all, 28 patients with bilateral CP (Gross Motor Function Classification System I to III) with two clinical gait analyses (CGA) were included (mean age: 9.0 years (sd 2.9) at the first CGA, 19.6 years (sd 4.1) at the last, all of them at skeletal maturity). GDI, walking speed and their changes were calculated. Statistical analysis was performed to observe differences between baseline and follow-up CGA. Pearson's correlations were conducted to evaluate the associations between GDI and walking speed changes with: GDI at baseline and walking speed at baseline. GDI and walking speed evolution have been analyzed for two groups of patients: with and without SEMLS. RESULTS: Regardless of the treatment, GDI was significantly higher at follow-up CGA (baseline: 73.1 (sd 13.1) versus follow-up: 80.1 (sd 13.2); p = 0.014). Significant negative correlations were found between GDI change and GDI at baseline (r = -0.52; p = 0.004) and between walking speed change and walking speed at the baseline (r = -0.70; p < 0.001). Regarding the group of patients with or without SEMLS, only significant improvement of GDI was found for patients with SEMLS (at baseline: 69.0 (sd 12.1) versus follow-up: 77.8 (sd 11.2); p < 0.05). CONCLUSION: Analysis at skeletal maturity showed a gait quality maintained for patients without SEMLS and an improvement for patients with SEMLS. LEVEL OF EVIDENCE: Level III.
Bonnefoy-Mazure A, De Coulon G, Armand S.


Aim: To explore how patients with cerebral palsy (CP) perceive their gait and evaluate associations between subjective gait perception and: objective gait parameters, endurance, pain, and fatigue. Method: Sixty-two patients (21 females and 41 males; mean [SD] age 20y [5y 1mo], range 15-29y) performed a clinical gait analysis. Self-selected walking speed, Gait Profile Score, and Gait Variable Score were calculated. Subjective gait perception was assessed with a visual analogue scale using the question: 'On a scale from 0 (worst) to 10 (optimal), how would you describe your walking today?'. A 6-minute walk test (6MWT) measured endurance; the 36-Item Short Form Health Survey (SF-36) evaluated quality of life. T-tests, Pearson correlations, and univariate and multiple linear regression models were used to compare and find associations between the data.

Results: Overall mean (SD) subjective gait perception was 7.5 (1.8) and was significantly higher for patients in Gross Motor Function Classification System (GMFCS) level I (7.9 [1.5]) than for patients in GMFCS levels II and III (5.9 [2.0]). Positive correlations were found between subjective gait perception and gait scores, walking speed, 6MWT distance, and SF-36 score. Only walking speed was a significant predictor of subjective gait perception. Interpretation: Subjective gait perception was influenced by GMFCS level and linked partially with the walking speed. The gait quality did not explain subjective gait perception. It is important to combine subjective and objective gait scores to develop personalized therapeutic goals. What this paper adds: Subjective gait perception is influenced by the physical impairment levels of patients with cerebral palsy. Subjective gait perception and objective gait scores are associated. Walking speed is the only predictor of gait perception.

PMID: 32162342

Zarkou A, Lee SCK, Prosser LA, Jeka JJ.


Sensory dysfunction is prevalent in cerebral palsy (CP). Evidence suggests that sensory deficits can contribute to manual ability impairments in children with CP, yet it is still unclear how they contribute to balance and motor performance. Therefore, the objective of this study was to investigate the relationship between lower extremity (LE) somatosensation and functional performance in children with CP. Ten participants with spastic diplegia (Gross Motor Function Classification Scale: I-III) and who were able to stand independently completed the study. Threshold of light touch pressure, two-point discriminatory ability of the plantar side of the foot, duration of cutaneous vibration sensation, and error in the joint position sense of the ankle were assessed to quantify somatosensory function. The balance was tested by the Balance Evaluation System Test (BESTest) and postural sway measures during a standing task. Motor performance was evaluated by using a battery of clinical assessments: (1) Gross Motor Function Measure (GMFM-66-IS) to test gross motor ability; (2) spatiotemporal gait characteristics (velocity, step length) to evaluate walking ability; (3) Timed Up and Go (TUG) and 6 Min Walk (6MWT) tests to assess functional mobility; and (4) an isokinetic dynamometer was used to test the Maximum Volitional Isometric Contraction (MVIC) of the plantar flexor muscles. The results showed that the light touch pressure measure was strongly associated only with the 6MWT. Vibration and two-point discrimination were strongly related to balance performance. Further, the vibration sensation of the first metatarsal head demonstrated a significantly strong relationship with motor performance as measured by GMFM-66-IS, spatiotemporal gait parameters, TUG, and ankle plantar flexors strength test. The joint position sense of the ankle was only related to one subdomain of the BESTest (Postural Responses). This study provides preliminary evidence that LE sensory deficits can possibly contribute to the pronounced balance and motor impairments in CP. The findings emphasize the importance of developing a thorough LE sensory test battery that can guide traditional treatment protocols toward a more holistic therapeutic approach by combining both motor and sensory rehabilitative strategies to improve motor function in CP.

PMID: 32161527

15. Ankle foot orthoses in cerebral palsy: Effects of ankle stiffness on trunk kinematics, gait stability and energy cost of
walking.
Meyns P, Kerkum YL, Brehm MA, Becher JG, Buizer AI, Harlaar J.


In children with cerebral palsy (CP), rigid ventral shell ankle-foot orthoses (vAFOs) are often prescribed to reduce excessive knee flexion in stance and lower the energy cost of walking (ECW). However, how vAFOs affect ECW is a complex issue, as vAFOs may have an impact on lower limb biomechanics, upper body movements, and balance. Besides, the vAFO's biomechanical effect have been shown to be dependent on its stiffness around the ankle joint. We examined whether vAFO stiffness influences trunk movements and gait stability in CP, and whether there is a relationship between these factors and ECW. Fifteen children with spastic CP were prescribed vAFOs. Stiffness was varied into a rigid, stiff and flexible configuration. At baseline (shoes-only) and for each vAFO stiffness configuration, 3D-gait analyses and ECW-tests were performed. From the gait analyses, we derived trunk tilt, lateroflexion, and rotation range of motion (RoM) and the mediolateral and anteroposterior Margins of Stability (MoS) and their variability as measures of gait stability. With the ECW-test we determined the netEC. We found that wearing vAFOs significantly increased trunk lateroflexion (Wald $\chi^2 = 33.7$, $p < 0.001$), rotation RoM (Wald $\chi^2 = 20.5$, $p < 0.001$) and mediolateral gait instability (Wald $\chi^2 = 10.4$, $p = 0.016$). The extent of these effects partly depended on the stiffness of the vAFO. Significant relations between trunk movements, gait stability and ECW were found $r = 0.57-0.81$, $p < 0.05$), which indicates that trunk movements and gait stability should be taken into account when prescribing vAFOs to improve gait in children with CP walking with excessive knee flexion.

PMID: 32147412

Xu D, Wu YN, Gaebler-Spira D, Gao F, Clegg NJ, Delgado MR, Zhang LQ.


AIM: To assess the neural and non-neural contributions to spasticity in the impaired ankle of children with cerebral palsy (CP).

METHOD: Instrumented tapping of the Achilles tendon was done isometrically to minimize non-neural contributions and elicit neural contributions. Robot-controlled ankle stretching was done at various velocities, including slow stretching, with minimized neural contributions. Spasticity was assessed as having neural (phasic and tonic stretch reflex torque, tendon reflex gain, contraction rate, and half relaxation rate) and non-neural origin (elastic stiffness and viscous damping) in 17 children with CP (six females and 11 males; mean age [SD] 10y 8mo [3y 11mo], range 4y-18y) and 17 typically developing children (six females and 11 males; mean age [SD] 12y 7mo [2y 9mo], range 7y-18y). All torques were normalized to weight×height.

RESULTS: Children with CP showed increased phasic and tonic stretch reflex torque (p=0.004 and p=0.001 respectively), tendon reflex gain (p=0.02), contraction rate (p=0.038), half relaxation rate (p=0.02), elastic stiffness (p=0.01), and viscous damping (p=0.01) compared to typically developing children. INTERPRETATION: Controlled stretching and instrumented tendon tapping allow the systematic quantification of various neural and non-neural changes in CP, which can be used to guide impairment-specific treatment. WHAT THIS PAPER ADDS: Ankle spasticity is associated with increased phasic and tonic stretch reflexes, tendon reflex gain, and contraction and half relaxation rates. Ankle spasticity is also associated with increased elastic stiffness and viscous damping.

PMID: 32147834

17. Effectiveness of treadmill training on gait function in children with cerebral palsy: meta-analysis.
Han YG, Yun CK.


The purpose of this review was to analysis the effects of treadmill training on gait function in children with cerebral palsy. Cumulative Index to Nursing and Allied Health Literature (CINAHL), Cochrane Central Register of Controlled Trials, Physiotherapy Evidence Database (PEDro), PubMed and Web of Science were searched. Investigating the effects of treadmill training on gait endurance, gait speed, limb support time, cadence, and step length in children with cerebral palsy. Similar outcomes were pooled by calculating the standardized mean difference. Of the eight studies, 179 participants were included.
The average PEDro score was 6.25/10. The results of the sensitivity test for bias evaluation using the duval and tweedie's trim and fill method showed low publication bias. The test regarding the effect of treadmill training on overall gait function yielded a moderate effect size of 0.53, which was a statistically significant effect as its confidence interval did not include. The overall effect size of gait endurance was 0.85. The overall effect size of gait speed and limb support time were 0.52 and 0.73. The overall effect size of cadence and step length were 0.14 and 0.21, indicating a nonsignificant improvement. These findings suggested that treadmill training on cerebral palsy was effective for gait endurance, gait speed and limb support time than cadence and step length.

PMID: 32161730

Short MR, Damiano DL, Kim Y, Bulea TC.

Children with unilateral cerebral palsy (CP) walk independently although with an asymmetrical, more poorly coordinated pattern compared to their peers. While gait biomechanics in unilateral CP and their alteration from those without CP have been well documented, cortical mechanisms underlying gait remain inadequately understood. To the best of our knowledge, this is the first study utilizing electroencephalography (EEG) during treadmill gait in older children with and without CP. Lower limb surface electromyographic (EMG) data were collected and muscle synergy analyses performed to quantify motor output. Our primary goal was to evaluate the relationships between cortical and muscle activation within and across groups and hemispheres to provide novel insights into neural control of gait and how it may be disrupted by an early unilateral brain injury. Participants included 9 children with unilateral CP, mean age 16.0 ± 2.7 years, and 12 with typical development (TD), mean age 14.8 ± 3.0 years. EEG data were collected during a standing baseline and treadmill walking at self-selected speed. EMG of 16 lower limb muscles were also collected bilaterally and synchronized with EEG. No significant group differences were found in synergy number or structure across groups. Six cortical clusters were identified as having gait-related activation and all contained participants from both CP and TD groups; however, the percent of individuals per group appearing in different clusters varied. Notably, the cluster least represented in CP was the non-dominant motor region. Both groups showed mu-band ERD in the motor clusters during gait although sustained beta-band ERD was not evident in TD. The CP group showed greater cortical activation than TD during walking as measured by mu- and beta-ERD in the dominant and non-dominant motor and parietal regions and elevated low gamma-activity in the frontal and parietal areas, a unique finding in CP. CP showed greater bilateral motor EEG-EMG coherence in the gamma-band with the hallucis longus compared to TD. In summary, individuals with CP display increased cortical activation during gait possibly relating to differences in distal motor control of the more affected side. Strategies that iteratively reduce cortical activation while improving selective motor control are needed in CP.

PMID: 32153376

19. Stretching Interventions in Children With Cerebral Palsy: Why Are They Ineffective in Improving Muscle Function and How Can We Better Their Outcome?
Kalkman BM, Bar-On L, O'Brien TD, Maganaris CN.

Hyper-resistance at the joint is one of the most common symptoms in children with cerebral palsy (CP). Alterations to the structure and mechanical properties of the musculoskeletal system, such as a decreased muscle length and an increased joint stiffness are typically managed conservatively, by means of physiotherapy involving stretching exercises. However, the effectiveness of stretching-based interventions for improving function is poor. This may be due to the behavior of a spastic muscle during stretch, which is poorly understood. The main aim of this paper is to provide a mechanistic explanation as to why the effectiveness of stretching is limited in children with CP and consider clinically relevant means by which this shortcoming can be tackled. To do this, we review the current literature regarding muscle and tendon plasticity in response to stretching in children with CP. First, we discuss how muscle and tendon interact based on their morphology and mechanical properties to provide a certain range of motion at the joint. We then consider the effect of traditional stretching exercises on these muscle and tendon properties. Finally, we examine possible strategies to increase the effectiveness of stretching therapies and we highlight areas of further research that have the potential to improve the outcome of non-invasive interventions in children with cerebral palsy.
Bekkers S, Pruijn IMJ, Van Hulst K, Delsing CP, Erasmus CE, Scheffer ART, Van Den Hoogen FJA.


AIM: To assess: (1) the effect on drooling of bilateral submandibular duct ligation as surgical therapy after the administration of submandibular botulinum neurotoxin A (BoNT-A) for excessive drooling and (2) the predictive value of treatment success with BoNT-A on treatment success after bilateral submandibular duct ligation. METHOD: This was a within-participant retrospective observational study in which 29 children with severe drooling (15 males, 14 females) received BoNT-A treatment at a mean age of 9 years 6 months (SD 2y 5mo), followed by bilateral submandibular duct ligation at a mean age of 10 years 11 months (SD 2y 4mo). Fifteen children were diagnosed with cerebral palsy (CP), with 12 children classified in Gross Motor Function Classification System levels IV and V. The 14 children without CP had non-progressive developmental disorders. The primary drooling severity outcomes were the Visual Analogue Scale (VAS; subjective assessment) and drooling quotient (objective assessment). Measurements were taken before each intervention and again at 8 and 32 weeks. RESULTS: The VAS was significantly lower after bilateral submandibular duct ligation at follow-up compared to BoNT-A treatment (mean difference -33, p≤0.001; 95% confidence interval [CI]= -43.3 to -22.9). The mean drooling quotient did not significantly differ between BoNT-A treatment and bilateral submandibular duct ligation at follow-up (3.3, p=0.457; 95% CI= -4.35 to 9.62) or between 8 and 32 weeks (4.7, p=0.188; 95% CI= -2.31 to 11.65). INTERPRETATION: BoNT-A treatment and bilateral submandibular duct ligation are both effective treatment modalities for drooling. At 32-week follow-up, subjective drooling severity after bilateral submandibular duct ligation was significantly lower compared to previous BoNT-A injections in participants. However, treatment success with BoNT-A is no precursor to achieving success with bilateral submandibular duct ligation.

PMID: 32149393

21. Challenges Faced by Therapists Providing Services for Infants with or at Risk for Cerebral Palsy.
Gmmash AS, Effgen SK, Goldey K.


OBJECTIVE: The purpose of this study was to identify the challenges physical therapists (PTs) and occupational therapists (OTs) have in providing early intervention (EI) for infants with or at risk for cerebral palsy. METHODS: Therapists' responses to an open-ended question were collected via survey that was distributed to EI providers and analyzed using content analysis. RESULTS: The primary self-reported barriers to PT/OT EI services had 5 themes: (1) inadequate communication and collaboration, (2) challenges in coordination with family, (3) policy limitations, (4) meeting the child's individual medical needs, and (5) unequal access to resources. CONCLUSION: Respondents reported that barriers are complex and exist at the individual, family, team, and societal levels. Further research is needed to explore barriers and solutions at each of these levels, from meeting a child's individual medical needs to improving interprofessional communication to increasing equitable access to resources.

PMID: 32150028

22. Brain-computer interfaces for communication.
Vansteensel MJ, Jarosiewicz B.


Locked-in syndrome (LIS) is characterized by an inability to move or speak in the presence of intact cognition and can be
caused by brainstem trauma or neuromuscular disease. Quality of life (QoL) in LIS is strongly impaired by the inability to communicate, which cannot always be remedied by traditional augmentative and alternative communication (AAC) solutions if residual muscle activity is insufficient to control the AAC device. Brain-computer interfaces (BCIs) may offer a solution by employing the person's neural signals instead of relying on muscle activity. Here, we review the latest communication BCI research using noninvasive signal acquisition approaches (electroencephalography, functional magnetic resonance imaging, functional near-infrared spectroscopy) and subdural and intracortical implanted electrodes, and we discuss current efforts to translate research knowledge into usable BCI-enabled communication solutions that aim to improve the QoL of individuals with LIS.

PMID: 32164869


BCI (brain-computer interface) and functional electrical stimulation (FES) technologies have advanced significantly over the last several decades. Recent efforts have involved the integration of these technologies with the goal of restoring functional movement in paralyzed patients. Implantable BCIs have provided neural recordings with increased spatial resolution and have been combined with sophisticated neural decoding algorithms and increasingly capable FES systems to advance efforts toward this goal. This chapter reviews historical developments that have occurred as the exciting fields of BCI and FES have evolved and now overlapped to allow new breakthroughs in medicine, targeting restoration of movement and lost function in users with disabilities.

PMID: 32164861


BACKGROUND: The purpose of this study was to describe clinical presentation, epilepsy, EEG, extent and site of the underlying cerebral lesion with special reference towards aetiologic background factors in a population-based group of children with hemiplegic cerebral palsy. METHODS: Forty-seven children of school-age, fulfilling the SPCE (Surveillance of Cerebral palsy in Europe)-criteria of hemiplegic cerebral palsy, identified via the Swedish cerebral palsy register, were invited and asked to participate in the study. RESULTS: Fifteen boys and six girls participated. Of the sixteen children born at term, five had no risk factors for cerebral palsy. Two out of five preterm children presented additional risk factors. Debut of motor impairment was observed in the first year of life in sixteen children. Age at diagnosis varied from 2 months to 6 years. Epilepsy was common and associated with grey- and white matter injury. CONCLUSIONS: Recognizing the importance of risk factors for cerebral palsy, any child with these risk factors should be offered a check-up by a paediatrician or a paediatric neurologist. Thereby reducing diagnostic delay. Epilepsy is common in hemiplegic cerebral palsy and associated with grey- and white matter injury in this cohort.

PMID: 32164572

25. Treatment and new progress of neonatal hypoxic-ischemic brain damage. Yang L, Zhao H, Cui H.

Neonatal hypoxic ischemia (HI) results in different extents of brain damage, and immature brain tissue is particularly sensitive to the stimulation of HI. Hypoxic-ischemic brain damage (HIBD) is a common and serious nervous system disease in neonates, for both full-term infants and preterm infants, and is one of the main causes of neonatal death. The surviving infants are often associated with cerebral palsy, mental retardation, and other sequelae, which severely affect quality of life. For term infants, hypoxia and ischemia mainly affect gray matter, whereas in preterm infants, the white matter. However, up to now, inadequate standards and specific measures that can be used to treat hypoxic-ischemic brain injury are available. Recently, in addition to supportive therapy and symptomatic treatment, research on the treatment of hypoxic-ischemic brain injury has focused on the following aspects: hypothermia therapy, stem cell therapy, neuroprotective agents, ibuprofen, and combination therapy. In this review, we will summarize the treatment of HIBD and make suggestions for the future treatment direction.

PMID: 32167570


Neonatal hypoxic-ischaemic encephalopathy (HIE) is a serious condition: many survivors develop neurological impairments, including cerebral palsy and intellectual disability. Preclinical studies show that the systemic administration of umbilical cord blood cells (UCBCs) is beneficial for neonatal HIE. We conducted a single-arm clinical study to examine the feasibility and safety of intravenous infusion of autologous UCBCs for newborns with HIE. When a neonate was born with severe asphyxia, the UCB was collected, volume-reduced, and divided into three doses. The processed UCB was infused at 12-24, 36-48, and 60-72 hours after the birth. The designed enrolment was six newborns. All six newborns received UCBC therapy strictly adhering to the study protocol together with therapeutic hypothermia. The physiological parameters and peripheral blood parameters did not change much between pre- and post-infusion. There were no serious adverse events that might be related to cell therapy. At 30 days of age, the six infants survived without circulatory or respiratory support. At 18 months of age, neurofunctional development was normal without any impairment in four infants and delayed with cerebral palsy in two infants. This pilot study shows that autologous UCBC therapy is feasible and safe.

PMID: 32165664

27. Incidence Rate of Advanced Chronic Kidney Disease Among Privately Insured Adults with Neurodevelopmental Disabilities.
Whitney DG, Schmidt M, Bell S, Morgenstern H, Hirth RA.

PURPOSE: Due to complex medical profiles, adults with neurodevelopmental disabilities (NDDs) may have a heightened risk for early development of chronic kidney disease (CKD) and accelerated CKD progression to advanced stages and kidney failure. The purpose of this study was to estimate the incidence rate of advanced CKD for adults with NDDs and compare the incidence rate to adults without NDDs. PATIENTS AND METHODS: Data were used from the Optum Clinformatics® Data Mart to conduct this retrospective cohort study. The calendar year 2013 was used to identify eligible participants: individuals ≥18 years of age and without advanced CKD. Participants were followed from 01/01/2014 to advanced CKD, loss to follow-up, death, or end of the study period (12/31/2017), whichever came first. Diagnostic, procedure, and diagnosis-related group codes identified NDDs (intellectual disabilities, cerebral palsy, autism spectrum disorders), incident cases of advanced CKD (CKD stages 4+), diabetes, cardiovascular diseases, and hypertension present in the year 2013. Crude incidence rates (IR) of advanced CKD and IR ratios (IRR), comparing adults with vs without NDDs (with 95% CI) were estimated. Then, Cox regression estimated the hazard ratio (HR and 95% CI) for advanced CKD, comparing adults with NDDs to adults without NDDs while adjusting for covariates. RESULTS: Adults with NDDs (n=33,561) had greater crude IR of advanced CKD (IRR=1.32; 95% CI=1.24-1.42) compared to adults without NDDs (n=6.5M). The elevated rate of advanced CKD among adults with NDDs increased after adjusting for demographics (HR=2.19; 95% CI=2.04-2.34) and remained elevated with further adjustment for hypertension and diabetes (HR=2.01; 95% CI=1.87-2.15) plus cardiovascular disease (HR=1.84; 95% CI=1.72-1.97). Stratified analyses showed that the risk of advanced CKD was greater for all NDD subgroups. CONCLUSION: Study findings suggest that adults with NDDs have a greater risk of advanced CKD than do adults without NDDs, and that
difference is not explained by covariates used in our analysis.

PMID: 32161503

28. Quantifying bone marrow fat using standard T1-weighted magnetic resonance images in children with typical development and in children with cerebral palsy.
Zhang C, Slade JM, Miller F, Modlesky CM.

Excess bone marrow adiposity may have a negative effect on bone growth and development. The aim of this study was to determine whether a procedure using standard T1-weighted magnetic resonance images provides an accurate estimate of bone marrow fat in children with typical development and in children with mild spastic cerebral palsy (CP; n = 15/group; 4-11 y). Magnetic resonance imaging was used to acquire T1-weighted images. It was also used to acquire fat and water images using an iterative decomposition of water and fat with echo asymmetry and least-squares estimation (IDEAL) technique. Bone marrow fat volume and fat fraction in the middle-third of the tibia were determined using the standard T1-weighted images (BMFVT1 and BMFFT1, respectively) and the fat and water images (BMFVIDEAL and BMFFIDEAL, respectively). In both groups, BMFVT1 was highly correlated with (both r > 0.99, p < 0.001) and not different from (both p > 0.05) BMFVIDEAL. In both groups, BMFFT1 was moderately correlated with (both r = 0.71, p < 0.01) and not different from (both p > 0.05) BMFFIDEAL. There was no group difference in BMFVT1 or BMFVIDEAL (both p > 0.05). BMFFIDEAL was higher in children with CP (p < 0.05), but there was no group difference in BMFFT1 (p > 0.05). We conclude that a procedure using standard T1-weighted magnetic resonance images can produce estimates of bone marrow fat volume similar to estimates from the IDEAL technique in children. However, it is less sensitive to variation in the bone marrow fat fraction.

PMID: 32152339

29. Of Major Complication Types, Only Deep Infections after Spinal Fusion are Associated with Worse Health-Related Outcomes in Children with Cerebral Palsy.

STUDY DESIGN: Retrospective review. OBJECTIVE: To determine whether 1) major postoperative complications ("complications") are associated with 2-year improvements in Caregiver Priorities and Child Health Index of Life with Disabilities (CPCHILD) scores after scoliosis surgery, and 2) complications and preoperative characteristics predict 2-year improvements in CPCHILD Total score. SUMMARY OF BACKGROUND DATA: Spinal arthrodesis can halt the progression of spinal deformity in patients with cerebral palsy (CP)-related scoliosis. However, these patients are prone to postoperative complications. METHODS: Using a multicenter CP registry, we identified 222 patients aged ≤21 years who underwent spinal fusion from 2008-2015 and had ≥2-year follow-up. We compared CPCHILD score improvement between 71 patients who had ≥1 complication ("complications group") versus 151 who did not ("no-complications group"). Complications were deep infections, thromboembolic events, and cardiopulmonary, gastrointestinal, and neurologic complications. Multiple linear regression was used to identify predictors of 2-year postoperative CPCHILD score improvement. Alpha = 0.05. RESULTS: At 2-year follow-up, the complications group had similar mean improvement in CPCHILD score across all domains compared with the no-complications group (p > .05). When stratifying by complication type, deep infection was associated with less improvement in CPCHILD Comfort and Emotions (p = .02), Quality of Life (p < .01), and Total (p = .04) scores. When controlling for Gross Motor Function Classification System subcategory, age, and body mass index, only preoperative CPCHILD Total score and postoperative deep infection (F(4, 176) = 14 (p < 0.0001, R = 0.24) predicted 2-year improvement in CPCHILD Total score. Higher preoperative Total score and postoperative deep infection independently predicted less improvement in Total score. CONCLUSIONS: Postoperative deep infection and higher preoperative CPCHILD Total score independently predicted less improvement in CPCHILD Total score. Other major postoperative complications were not associated with differences in 2-year postoperative improvements in CPCHILD scores across all domains. LEVEL OF EVIDENCE: 3.

PMID: 32150131
30. Submuscular Placement of Baclofen Infusion Pumps: Case Series and Technique.


BACKGROUND: Baclofen pumps provide treatment of symptoms of spasticity for disease processes such as cerebral palsy and traumatic brain injury. These devices provide continuous infusion or periodic dosing of intrathecal baclofen (ITB). Traditionally, these pumps have been placed subcutaneously. Subcutaneous device placement has been associated with infection and extrusion. Baclofen pumps are large and range from 8 to 10 cm in diameter and 4 to 8 cm in width. Patients requiring device placement typically have a paucity of subcutaneous tissue. Cachexia coupled with the size and bulk of these devices leads to increased protrusion and friction. Submuscular placement provides a well-vascularized pocket that directs the device inward eliminating protrusion and decreasing the potential for soft tissue breakdown. METHODS: A retrospective chart review of ITB pump placement in a submuscular plane by a single plastic surgeon at a major academic center in conjunction with a neurosurgeon was performed. Inclusion criteria were cases of primary placement or replacement of ITB pumps and spasticity requiring ITB. Major complications included infection, extrusion, and reoperation. RESULTS: Five patients during a 5-month period were treated with submuscular placement of ITB pumps. Average age of patients included was 18.4 years. Average preoperative body mass index was 18.8 kg/m², with values ranging from 15.8 to 20.1 kg/m². Medical histories of patients included diagnoses of cerebral palsy and traumatic brain injury causing spasticity. The most frequently cited reason for plastic surgical consultation preoperatively was cachexia. Two patients had previous baclofen pumps placed subcutaneously. Average follow-up was 6 months and ranged from 3 to 13 months. There were no major complications. One patient had a small seroma that spontaneously resolved. No patient had wound healing problems, and there were no extrusions of implanted devices or reoperations. CONCLUSION: Submuscular placement of baclofen pumps provides a well-vascularized and stable environment for device placement that minimizes the dangers of pump extrusion and infection. Patients who require treatment with ITB commonly have severe cachexia, which makes subcutaneous device placement high risk. Submuscular placement should be performed in all patients with body mass index less than 20 kg/m².

PMID: 32149846

Johansen M, Haslund-Thomsen H, Kristensen J, Thorgaard Skou S.


PURPOSE: To investigate intrarater and interrater reliability, agreement, and concurrent validity of a smartphone photography-based application compared with a universal goniometer in children with cerebral palsy. METHODS: Range of motion of hip abduction, popliteal angle, and ankle dorsiflexion was measured with a universal goniometer and a photography-based application in children with cerebral palsy, Gross Motor Function Classification System levels I to V. A 2-way random-effects intraclass correlation coefficients and Bland-Altman plots, standard error of measurement, and smallest detectable change were used for analyses. RESULTS: The application had good to excellent reliability and concurrent validity compared with a universal goniometer, while the large measurement error of both methods suggests that changes of 10° to 23° are needed to be certain that changes over time are not results of measurement error. CONCLUSIONS: A photography-based goniometer can be a reliable and valid tool when measuring range of motion in children with cerebral palsy.

PMID: 32150030

32. Wearable sensors to improve activities in individuals with cerebral palsy.
Dan J.


PMID: 32149396
33. Single group multisite safety trial of sibling cord blood cell infusion to children with cerebral palsy: study protocol and rationale.


INTRODUCTION: Cerebral palsy (CP) is the most common physical disability of childhood but has no cure. Stem cells have the potential to improve brain injury and are proposed as a therapy for CP. However, many questions remain unanswered about the most appropriate cell type, timing of infusions, dose required and associated risks. Therefore, human safety and efficacy trials are necessary to progress knowledge in the field. METHODS AND ANALYSIS: This is a single group study with sample size n=12 to investigate safety of single-dose intravenous 12/12 human leucocyte antigen-matched sibling cord blood cell infusion to children with CP aged 1-16 years without immune suppression. The study is similar to a 3+3 design, where the first two groups of participants have severe CP, and the final six participants include children with all motor severities. Children will be monitored for adverse events and the duration that donor cells are detected. Assessments at baseline, 3 and 12 months will investigate safety and preliminary evidence of change in gross motor, fine motor, cognitive and quality of life outcomes. ETHICS AND DISSEMINATION: Full approval was obtained from The Royal Children's Hospital Human Research Ethics Committee, and a clinical trial notification was accepted by Australia's Therapeutic Goods Administration. Participant guardian informed consent will be obtained before any study procedures. The main results of this study will be submitted for publication in a peer-reviewed journal. TRIAL REGISTRATION NUMBER: ACTRN12616000403437, NCT03087110.

PMID: 32152173

34. Therapeutic mechanism of cord blood mononuclear cells via the IL-8-mediated angiogenic pathway in neonatal hypoxic-ischaemic brain injury.
Cho KH, Choi JI, Kim JO, Jung JE, Kim DW, Kim M.


In a clinical trial of cerebral palsy, the level of plasma interleukin-8 (IL-8) was increased, correlated with motor improvement, after human umbilical cord blood mononuclear cell (hUCBC) infusion. This study aimed to elucidate the role of IL-8 in the therapeutic effects of hUCBCs in a mouse model of hypoxic-ischaemic brain injury (HI). In P7 HI mouse brains, hUCBC administration at day 7 after HI upregulated the gene expression of Cxcl2, the mouse IL-8 homologue and increased the expression of its receptor, CXCR2. hUCBC administration restored the sequential downstream signalling axis of p-p38/p-MAPKAPK2, NFκB, and angiogenic factors, which were downregulated by HI. An in vitro assay revealed the downregulation of the angiogenic pathway by CXCR2 knockdown and p38 inhibition. In vivo p38 inhibition prior to hUCBC administration in HI mouse brains produced identical results. Behavioural outcomes revealed a therapeutic effect (ps < 0.01) of hUCBC or IL-8 administration, which was correlated with decreases in infarct size and angiogenic findings in the striatum. In conclusion, the response of the host to hUCBC administration in mice upregulated Cxcl2, which led to the activation of the IL-8-mediated p-p38 signalling pathway. The upregulation of the downstream pathway and angiogenic growth factors via NFκB can be inferred to be the potential therapeutic mechanism of hUCBCs.

PMID: 32157146