1. How does the interaction of presumed timing, location and extent of the underlying brain lesion relate to upper limb function in children with unilateral cerebral palsy?


BACKGROUND: Upper limb (UL) function in children with unilateral cerebral palsy (CP) vary largely depending on presumed timing, location and extent of brain lesions. These factors might exhibit a complex interaction and the combined prognostic value warrants further investigation. This study aimed to map lesion location and extent and assessed whether these differ according to presumed lesion timing and to determine the impact of structural brain damage on UL function within different lesion timing groups. MATERIALS AND METHODS: Seventy-three children with unilateral CP (mean age 10 years 2 months) were classified according to lesion timing: malformations (N = 2), periventricular white matter (PWM, N = 42) and cortical and deep grey matter (CDGM, N = 29) lesions. Neuroanatomical damage was scored using a semi-quantitative MRI scale. UL function was assessed at body function and activity level. RESULTS: CDGM lesions were more pronounced compared to PWM lesions (p = 0.0003). Neuroanatomical scores were correlated with a higher degree to UL function in the CDGM group (rs = 0.39 to rs = 0.84) compared to the PWM group (rs = -0.42 to rs = -0.61). Regression analysis found lesion location and extent to explain 75% and 65% (p < 0.02) respectively, of the variance in AHA performance in the CDGM group, but only 24% and 12% (p < 0.03) in the PWM group. CONCLUSIONS: In the CDGM group, lesion location and extent seems to impact more on UL function compared to the PWM group. In children with PWM lesions, other factors like corticospinal tract (re)organization and structural connectivity may play an additional role.

PMID: 28606752

2. Duruöz Hand Index: Is it valid and reliable in children with unilateral cerebral palsy?

Sanal-Top C, Karadag-Saygi E, Saçaklıdr R, Duruöz MT.


PURPOSE: To investigate the validity and reliability of the Duruöz Hand Index (DHI) in patients with unilateral cerebral palsy (CP). METHODS: Assessments of patients (n = 23) were performed using the Modified Ashworth Scale (MAS), the Manual Ability Classification System (MACS), the grip and pinch strength tests, and DHI. Following the data collection, retest of DHI was administered telephonically within a 2-week period. RESULTS: Test-retest reliability and internal consistency of DHI were found to be excellent with a Cronbach's alpha value of 0.93 and an intraclass correlation coefficient value of 0.94. The correlation between the DHI and MACS was detected significantly high (r = 0.840, p = 0.010). The DHI
also correlated with grip and pinch strength in the affected side (r = -0.459, p < 0.050; r = -0.509, p < 0.050).

CONCLUSIONS: DHI is a valid and reliable questionnaire for patients with unilateral CP.

PMID: 28604242

3. Exercise interventions for cerebral palsy.

Ryan JM, Cassidy EE, Noorduyn SG, O'Connell NE.


BACKGROUND: Cerebral palsy (CP) is a neurodevelopmental disorder resulting from an injury to the developing brain. It is the most common form of childhood disability with prevalence rates of between 1.5 and 3.8 per 1000 births reported worldwide. The primary impairments associated with CP include reduced muscle strength and reduced cardiorespiratory fitness, resulting in difficulties performing activities such as dressing, walking and negotiating stairs. Exercise is defined as a planned, structured and repetitive activity that aims to improve fitness, and it is a commonly used intervention for people with CP. Aerobic and resistance training may improve activity (i.e. the ability to execute a task) and participation (i.e. involvement in a life situation) through their impact on the primary impairments of CP. However, to date, there has been no comprehensive review of exercise interventions for people with CP. OBJECTIVES: To assess the effects of exercise interventions in people with CP, primarily in terms of activity, participation and quality of life. Secondary outcomes assessed body functions and body structures. Comparators of interest were no treatment, usual care or an alternative type of exercise intervention. SEARCH METHODS: In June 2016 we searched CENTRAL, MEDLINE, Embase, nine other databases and four trials registers. SELECTION CRITERIA: We included randomised controlled trials (RCTs) and quasi-RCTs of children, adolescents and adults with CP. We included studies of aerobic exercise, resistance training, and 'mixed training' (a combination of at least two of aerobic exercise, resistance training and anaerobic training). DATA COLLECTION AND ANALYSIS: Two review authors independently screened titles, abstracts and potentially relevant full-text reports for eligibility; extracted all relevant data and conducted 'Risk of bias' and GRADE assessments. MAIN RESULTS: We included 29 trials (926 participants); 27 included children and adolescents up to the age of 19 years, three included adolescents and young adults (10 to 22 years), and one included adults over 20 years. Males constituted 53% of the sample. Five trials were conducted in the USA; four in Australia; two in Egypt, Korea, Saudi Arabia, Taiwan, the Netherlands, and the UK; three in Greece; and one apiece in India, Italy, Norway, and South Africa. Twenty-six trials included people with spastic CP only; three trials included children and adolescents with spastic and other types of CP. Twenty-one trials included people who were able to walk with or without assistive devices, four trials also included people who used wheeled mobility devices in most settings, and one trial included people who used wheeled mobility devices only. Three trials did not report the functional ability of participants. Only two trials reported participants' manual ability. Eight studies compared aerobic exercise to usual care, while 15 compared resistance training and 4 compared mixed training to usual care or no treatment. Two trials compared aerobic exercise to resistance training. We judged all trials to be at high risk of bias overall. We found low-quality evidence that aerobic exercise improves gross motor function in the short term (standardised mean difference (SMD) 0.53, 95% confidence interval (CI) 0.02 to 1.04, N = 65, 3 studies) and intermediate term (mean difference (MD) 12.96%, 95% CI 0.52% to 25.40%, N = 12, 1 study). Aerobic exercise does not improve gait speed in the short term (MD 0.09 m/s, 95% CI -0.11 m/s to 0.28 m/s, N = 82, 4 studies, very low-quality evidence) or intermediate term (MD -0.17 m/s, 95% CI -0.59 m/s to 0.24 m/s, N = 12, 1 study, low-quality evidence). No trial assessed participation or quality of life following aerobic exercise. We found low-quality evidence that resistance training does not improve gross motor function (SMD 0.12, 95% CI -0.19 to 0.43, N = 164, 7 studies), gait speed (MD 0.03 m/s, 95% CI -0.02 m/s to 0.07 m/s, N = 185, 8 studies), participation (SMD 0.34, 95% CI -0.01 to 0.70, N = 127, 2 studies) or parent-reported quality of life (MD 12.70, 95% CI -5.63 to 31.03, n = 12, 1 study) in the short term. There is also low-quality evidence that resistance training does not improve gait speed (MD -0.03 m/s, 95% CI -0.17 m/s to 0.11 m/s, N = 84, 3 studies), gross motor function (SMD 0.13, 95% CI -0.30 to 0.55, N = 85, 3 studies) or participation (MD 0.37, 95% CI -6.61 to 7.35, N = 36, 1 study) in the intermediate term. We found low-quality evidence that mixed training does not improve gross motor function (SMD 0.02, 95% CI -0.29 to 0.33, N = 163, 4 studies) or gait speed (MD 0.10 m/s, -0.07 m/s to 0.27 m/s, N = 58, 1 study) but does improve participation (MD 0.40, 95% CI 0.13 to 0.67, N = 65, 1 study) in the short-term. There is no difference between resistance training and aerobic exercise in terms of the effect on gross motor function in the short term (SMD 0.02, 95% CI -0.50 to 0.55, N = 56, 2 studies, low-quality evidence). Thirteen trials did not report adverse events, seven reported no adverse events, and nine reported non-serious adverse events. AUTHORS' CONCLUSIONS: The quality of evidence for all conclusions is low to very low. As included trials have small sample sizes, heterogeneity may be underestimated, resulting in considerable uncertainty relating to effect estimates. For children with CP, there is evidence that aerobic exercise may result in a small improvement in gross motor function, though it does not improve gait speed. There is evidence that resistance training does not improve gait speed, gross motor function, participation or quality of life among children with CP. Based on the evidence available, exercise appears to be safe for people with CP; only 55% of trials, however, reported adverse events or stated that they monitored adverse events. There is a need for large, high-quality, well-reported RCTs that assess the effectiveness of exercise in terms of activity and participation, before drawing any firm conclusions on the effectiveness of exercise for people with CP. Research is also required to determine if current exercise guidelines for the general population are effective and feasible for people with CP.

PMID: 28602046

Abdolrahmani A, Sakita H, Yonetsu R, Iwata A.


[Purpose] This pilot study examined the immediate effects of quick-seated trunk exercise on sit-to-stand movement in children with cerebral palsy. [Subjects and Methods] Five children with spastic cerebral palsy (hemiplegia, 3; diplegia, 2; age 6-17 years) performed five sessions of natural-seated trunk exercise at a self-selected speed (control). Following a 50-min rest period, five sessions of the quick-seated trunk exercise were conducted (experimental intervention) for each child. Each seated trunk exercise included 10 repetitions in the anterior-posterior and lateral directions. Sit-to-stand was assessed before and after each intervention using a motion analysis system. The total sit-to-stand task duration and sagittal, angular movements of the trunk, hip, knee, and ankle were calculated. [Results] There was a significant difference in the total duration of the sit-to-stand movement before and after natural-seated trunk exercise (2.40 ± 0.67 s vs. 2.24 ± 0.44 s) as well as quick seated trunk exercise (2.41 ± 0.54 s vs. 2.06 ± 0.45 s). However, the sit-to-stand duration increased after natural-seated trunk exercise in one participant while that after quick-seated trunk exercise decreased in all participants. [Conclusion] Performing a trunk exercise in a seated position resulted in immediate improvement of the temporal sit-to-stand parameters in children with spastic cerebral palsy.

PMID: 28603369

5. The association between the maximum step length test and the walking efficiency in children with cerebral palsy.

Kimoto M, Okada K, Sakamoto H, Kondou T.


[Purpose] To improve walking efficiency could be useful for reducing fatigue and extending possible period of walking in children with cerebral palsy (CP). For this purpose, current study compared conventional parameters of gross motor performance, step length, and cadence in the evaluation of walking efficiency in children with CP. [Subjects and Methods] Thirty-one children with CP (21 boys, 10 girls; mean age, 12.3 ± 2.7 years) participated. Parameters of gross motor performance, including the maximum step length (MSL), maximum side step length, step number, lateral step up number, and single leg standing time, were measured in both dominant and non-dominant sides. Spatio-temporal parameters of walking, including speed, step length, and cadence, were calculated. Total heart beat index (THBI), a parameter of walking efficiency, was also calculated from heartbeats and walking distance in 10 minutes of walking. To analyze the relationships between these parameters and the THBI, the coefficients of determination were calculated using stepwise analysis. [Results] The MSL of the dominant side best accounted for the THBI (R²=0.759). [Conclusion] The MSL of the dominant side was the best explanatory parameter for walking efficiency in children with CP.

PMID: 28603353


Daunter AK, Kratz AL, Hurvitz EA.


AIM: Selective dorsal rhizotomy (SDR) is a surgical treatment for spasticity in children with cerebral palsy (CP). Studies suggest long-lasting effects of SDR on spasticity; long-term effects on symptoms and function are not clear. This study tested whether adults with CP (average 22y after SDR) report less pain, fatigue, and functional decline than a retrospectively assessed non-surgical comparison group. METHOD: This was a case-control study. Eighty-eight adults with CP (mean age 27y; SDR=38 male/female/missing=20/16/2; non-surgical [comparison]=50, male/female=19/31) recruited from a tertiary care center and the community completed a battery of self-reported outcome measures. Regression models were used to test whether SDR status predicted pain, fatigue, functional change, and hours of assistance (controlling for Gross Motor Function Classification System level). RESULTS: SDR status did not significantly predict pain interference (p=0.965), pain intensity (p=0.512), or fatigue (p=0.404). SDR related to lower decline in gross motor functioning (p=0.010) and approximately 6 fewer hours of daily assistance than for those in the comparison group (p=0.001). INTERPRETATION: Adults with CP who had...
SDR in childhood reported less gross motor decline and fewer daily assistance needs than non-surgically treated peers, suggesting the functional impact of SDR persists long after surgery.

PMID: 28617943

7. Spasticity Measurement Based on Tonic Stretch Reflex Threshold in Children with Cerebral Palsy Using the PediAnklebot.

Germanotta M, Taborri J, Rossi S, Frascarelli F, Palermo E, Cappa P, Castelli E, Petrarca M.


Nowadays, objective measures are becoming prominent in spasticity assessment, to overcome limitations of clinical scales. Among others, Tonic Stretch Reflex Threshold (TSRT) showed promising results. Previous studies demonstrated the validity and reliability of TSRT in spasticity assessment at elbow and ankle joints in adults. Purposes of the present study were to assess: (i) the feasibility of measuring TSRT to evaluate spasticity at the ankle joint in children with Cerebral Palsy (CP), and (ii) the correlation between objective measures and clinical scores. A mechatronic device, the pediAnklebot, was used to impose 50 passive stretches to the ankle of 10 children with CP and 3 healthy children, to elicit muscles response at 5 different velocities. Surface electromyography, angles, and angular velocities were recorded to compute dynamic stretch reflex threshold; TSRT was computed with a linear regression through angles and angular velocities. TSRTs for the most affected side of children with CP resulted into the biomechanical range (95.7 ± 12.9° and 86.7 ± 17.4° for Medial and Lateral Gastrocnemius, and 75.9 ± 12.5° for Tibialis Anterior). In three patients, the stretch reflex was not elicited in the less affected side. TSRTs were outside the biomechanical range in healthy children. However, no correlation was found between clinical scores and TSRT values. Here, we demonstrated the capability of TSRT to discriminate between spastic and non-spastic muscles, while no significant outcomes were found for the dorsiflexor muscle.

PMID: 28611612


Johans SJ, Swong KN, Hofler RC, Anderson DE.


Dystonia is a movement disorder characterized by involuntary muscle contractions, which cause twisting movements or abnormal postures. Deep brain stimulation has been used to improve the quality of life for secondary dystonia caused by cerebral palsy. Despite being a viable treatment option for childhood dystonic cerebral palsy, deep brain stimulation is associated with a high rate of infection in children. The authors present a small series of patients with dystonic cerebral palsy who underwent a stepwise approach for bilateral globus pallidus interna deep brain stimulation placement in order to decrease the rate of infection. Four children with dystonic cerebral palsy who underwent a total of 13 surgical procedures (electrode and battery placement) were identified via a retrospective review. There were zero postoperative infections. Using a multistaged surgical plan for pediatric patients with dystonic cerebral palsy undergoing deep brain stimulation may help to reduce the risk of infection.

PMID: 28604158

9. Posture-Dependent Dysphagia After Botulinum Toxin Type A Injection at Sternocleidomastoid in a Patient With Athetoid Cerebral Palsy.

Chang WK, Kim K, Seo HG, Leigh JH, Bang MS.


Cervical dystonia is a common issue in patients with athetoid cerebral palsy. Botulinum toxin injection to dystonic cervical muscles is a well-recognized treatment option, but it is known to be associated with dysphagia. Previously reported cases of dysphagia after botulinum toxin injection to the sternocleidomastoid muscle were related to the regional spread of toxin to the
pharyngeal muscles. We report a unique case of posture-dependent dysphagia due to preactivation of the suprathyroid and infrahyoid muscles to compensate for impaired head stabilization by the weakened sternocleidomastoid muscle while swallowing. This case suggests a possible mechanism of dysphagia in patients with athetoid cerebral palsy.

PMID: 28604409


Wood R.


[No abstract available]

PMID: 28612955

Prevention and Cure


Rizk M, Aziz J, Shorr R, Allan DS.


BACKGROUND: Cell-based therapy using umbilical cord blood is used increasingly for novel applications. To balance heightened public expectations and ensure appropriateness of emerging cell-based treatment choices, regular evidence-based assessment of novel cord blood-derived therapies is needed. METHODS AND RESULTS: We performed a systematic search of the literature and identified 57 studies (814 patients) for analysis. 16 studies (353 patients) included control groups for comparison. The most commonly reported novel indication for therapy was neurological diseases (25 studies, 476 patients), including studies of cerebral palsy (12 studies, 276 patients). Other indications included diabetes mellitus (9 studies, 149 patients), cardiac and vascular diseases (7 studies, 24 patients) and hepatic diseases (4 studies, 106 patients). Most studies administered total nucleated cells, mononuclear cells or CD34-selected cells (31 studies, 513 patients) while 20 studies described the use of cord blood-derived mesenchymal stromal cells. The majority of reports (46 studies, 627 patients) described cellular products obtained from allogeneic sources while 11 studies (187 patients) used autologous products. We identified 3 indications where multiple prospective controlled studies have been published (4/4 reported clinical benefit in cerebral palsy, 1/3 studies reported benefit for cirrhosis, and 1/3 studies reported biochemical response in type 1 diabetes), although heterogeneity between studies precluded meaningful pooled analysis of results. CONCLUSIONS: We anticipate a more clear understanding of the clinical benefit for specific indications once more controlled studies are reported. Patients should continue to be enrolled on registered clinical trials for novel therapies. Blood establishments, transplant centres, and regulatory bodies need to prepare for greater clinical demand.

PMID: 28602892


We have analyzed concentrations of magnesium (Mg), calcium (Ca), copper (Cu), zinc (Zn) and iron (Fe) in hair of a group of 82 children with mental retardation, in which 9 patients suffered from epilepsy, 18 from the Down's syndrome and 55 from
cerebral palsy. Girls comprised little over 50% of the patients. In the group of boys with epilepsy, we found Mg, Ca, Cu and Fe deficiency, and normal level of Zn. In the group of girls with epilepsy, apart from low Fe concentration, a high level of Ca, Mg, Zn, and Cu was noted. For girls with the Down's syndrome, a high or normal level of Ca, Mg, Zn and Cu was found, whereas the Fe concentration varied and presented itself in a non-characteristic way. Both groups of children with cerebral palsy, i.e. boys and girls, displayed low Fe concentration in their hair; low Cu level was found in older patients as well. In this group of patients, we also noted high concentrations of Ca, Mg and Zn in girls and normal in boys. A high concentration of Ca in girls with cerebral palsy requires separate analysis. The obtained results could be useful as guidance in the direction and determination of the amount of possible patient nutritional supplementation.

PMID: 28612063


Dyskinetic cerebral palsy (CP) has long been associated with basal ganglia and thalamus lesions. Recent evidence further points at white matter (WM) damage. This study aims to identify altered WM pathways in dyskinetic CP from a standardized, connectome-based approach, and to assess structure-function relationship in WM pathways for clinical outcomes. Individual connectome maps of 25 subjects with dyskinetic CP and 24 healthy controls were obtained combining a structural parcellation scheme with whole-brain deterministic tractography. Graph theoretical metrics and the network-based statistic were applied to compare groups and to correlate WM state with motor and cognitive performance. Results showed a widespread reduction of WM volume in CP subjects compared to controls and a more localized decrease in degree (number of links per node) and fractional anisotropy (FA), comprising parieto-occipital regions and the hippocampus. However, supramarginal gyrus showed a significantly higher degree. At the network level, CP subjects showed a bilateral pathway with reduced FA, comprising sensorimotor, intraparietal and fronto-parietal connections. Gross and fine motor functions correlated with FA in a pathway comprising the sensorimotor system, but gross motor also correlated with prefrontal, temporal and occipital connections. Intelligence correlated with FA in a network with fronto-striatal and parieto-frontal connections, and visuoception was related to right occipital connections. These findings demonstrate a disruption in structural brain connectivity in dyskinetic CP, revealing general involvement of posterior brain regions with relative preservation of prefrontal areas. We identified pathways in which WM integrity is related to clinical features, including but not limited to the sensorimotor system. Hum Brain Mapp, 2017. © 2017 Wiley Periodicals, Inc.

PMID: 28608616


Objective  This study aims to analyze the etiology and perinatal outcome of nonimmune hydrops fetalis (NIHF) in Southern China. Methods  All cases with NIHF diagnosed antenatally from January 1, 2007 to December 31, 2014 were identified and analyzed. Results  Total 482 cases of NIHF were identified during the study period. The most common cause of NIHF was hemoglobin (Hb) Bart's disease (61.8%), followed by chromosomal abnormalities (13.5%), idiopathic etiology (13.1%), cardiac abnormalities (6.4%), and others (5.2%). After 20 weeks' gestation, a total of 408 cases of NIHF presented, including Hb Bart's disease (279 cases), cardiac abnormalities (27 cases), and infection (7 cases). NIHF caused by chromosomal abnormalities mainly presented between 15 and 19 weeks' gestation. Of the 482 cases, 459 cases elected termination of pregnancy. The remaining 23 cases elected to continue their pregnancy. Among them, 14 (60.9%) resulted in intrauterine fetal death, 6 had neonatal death, 3 infants survived to 1 year of age. Of the three infants, one has cerebral palsy, and the remaining two are normal. Conclusions  Hb Bart's disease is the most common cause of NIHF in Southern China. An effective prenatal screening and counseling program for thalassemia in this region may be the most effective way to lower the incidence NIHF.

PMID: 28611934