# Science Infos Paralysie Cérébrale



### N°23- MAI 2016

### ENQUÊTE LANCEE LE 23 MAI 2016 :

QUESTIONNAIRE DISPONIBLE EN LIGNE



La Fondation Motrice lance l'enquête nationale (ESPACE\*) pour évaluer les soins reçus, besoins perçus, les priorités et améliorations attendues en Rééducation Motrice par les personnes atteintes de Paralysie Cérébrale et leur famille.

Pour toute question concernant l'enquête, merci de vous adresser à : espace@kappasante.com

#### SOUS L'ÉGIDE DES SOCIÉTÉS SAVANTES :



\*ESPACE : Enquête Satisfaction PAralysie CErébrale

La rééducation motrice, notamment la kinésithérapie, constitue l'un des piliers de la prise en charge des personnes atteintes de Paralysie Cérébrale (PC)/Infirmité Motrice Cérébrale (IMC).. A ce jour il n'existe pas en France de référentiels ou recommandations de bonne pratique auxquelles se référer. Peu d'informations sont disponibles sur le point de vue des personnes concernées et de leur famille. Néanmoins, il semble exister une difficulté à avoir un accès aux soins de façon équitable sur le territoire (perception de disparités territoriales dans les prises en charge).

Pour formaliser le ressenti des familles, La Fondation Motrice en partenariat avec les associations représentants les familles (Fédération Française des Associations des Infirmes Moteurs Cérébraux-FFAIMC), avec la participation des associations, Hémiparésie et Envoludia, **a lancé le 23 mai 2016** l'enquête nationale ESPACE destinée à recueillir la satisfaction et le ressenti des personnes atteintes de PC et de leur famille.

Le questionnaire, préparé sur la base du retour d'expérience de personnes atteintes de PC et de leur famille, a été élaboré avec l'aide d'un comité d'étude pluridisciplinaire regroupant des personnes concernées, associations de familles, médecins (neurologie, médecine physique et de réadaptation, épidémiologiste) et masseurs kinésithérapeutes spécialisés.

Cette enquête a été élaborée avec la participation active et le soutien de la Société Francophone d'Etudes et de Recherche sur les Handicaps de l'enfant (sferhe) ; Société Française de Médecine Physique et de Réadaptation (SOFMER) ; Cercle de Documentation et d'Information pour la rééducation des Infirmes Moteurs Cérébraux (CDI).

Les résultats pourront mener les promoteurs à proposer à un groupe multidisciplinaire de réflexion sur l'élaboration de bonnes pratiques en rééducation motrice en lien avec la Haute Autorité de Santé. Par ailleurs, ils feront émerger des projets répondant aux attentes des personnes atteintes et leur famille.

#### Pour en savoir plus et/ou pour participer à l'enquête (confidentielle et anonyme)

Science Infos Paralysie Cérébrale, mai 2016, FONDATION MOTRICE 67 rue Vergniaud 75013 Paris - tel +33 1 1 45 54 03 03 contact: Christine Doumergue cdoumergue@lafondationmotrice.org

### Sommaire

## Manifestations et congrès Publications scientifiques

Cerebral palsy.

### Epidémiologie

Bangladesh Cerebral Palsy Register (BCPR): a pilot study to develop a national cerebral palsy (CP) register with surveillance of children for CP. 8

#### **4** • Prévalence - Incidence

Birth Prevalence of Cerebral Palsy: A Population-Based Study. 9 Prevalence of Cerebral Palsy among 8-Year-Old Children in 2010 and Preliminary Evidence of Trends in Its Relationship to Low Birthweight. 9

#### **4** • Facteurs de risque – Causes

Asphyxia, Neurologic Morbidity, and Perinatal Mortality in Early-Term and Postterm Birth. 10 Appearance of neurodevelopmental disorders in children delivered post-term: a cross-section study. 10 Early onset preeclampsia and cerebral palsy: a double hit model? 10 Increased nuchal translucency thickness and the risk of neurodevelopmental disorders. 11 Neonatal brain injury as a consequence of insufficient cerebral oxygenation. 11 The neurological outcomes of cerebellar injury in premature infants. 12 Weight Status in the First 2 Years of Life and Neurodevelopmental Impairment in Extremely Low Gestational Age

Newborns. 12

#### **4**•Génétique

Genetic factors may underlie many cerebral palsy cases: New research implicates genetic variations, not lack of oxygen at birth, in disorder. 13

### Lésions Prévention des lesions 13

#### •Données fondamentales

Hyaluronidase and Hyaluronan Oligosaccharides Promote Neurological Recovery after Intraventricular Hemorrhage.

13

Human Amnion Epithelial Cells Modulate Ventilation-Induced White Matter Pathology in Preterm Lambs. 14

In vivo quantification of intraventricular hemorrhage in a neonatal piglet model using an EEG-layout based electrical impedance tomography array. 14

Stromal Cell-Derived Factor-1 $\alpha$  Plays a Crucial Role Based on Neuroprotective Role in Neonatal Brain Injury in Rats.

15

Rodent Hypoxia-Ischemia Models for Cerebral Palsy Research: A Systematic Review. 15 Systemic dendrimer-drug treatment of ischemia-induced neonatal white matter injury. 16

#### •Données cliniques

Brain structural and microstructural alterations associated with cerebral palsy and motor impairments in adolescents born extremely preterm and/or extremely low birthweight. 16

Does Contralesional Hand Function After Neonatal Stroke Only Depend on Lesion Characteristics? 17 Effects and Safety of Magnesium Sulfate on Neuroprotection: A Meta-analysis Based on PRISMA Guidelines. 17 Effect of Early Prophylactic High-Dose Recombinant Human Erythropoietin in Very Preterm Infants on

Neurodevelopmental Outcome at 2 Years: A Randomized Clinical Trial. 18

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Fetal and Neonatal Effects of N-Acetylcysteine When Used for Neuroprotection in Maternal Chorioamnionitis. 18 Grey matter injury patterns in cerebral palsy: associations between structural involvement on MRI and clinical outcomes. 19

Magnesium sulphate and perinatal mortality and morbidity in very-low-birthweight infants born between 24 and 32 weeks of gestation in Japan. 19

MR angiography findings in infants with neonatal arterial ischemic stroke in the middle cerebral artery territory: A prospective study using circle of Willis MR angiography. 20

Neurodevelopmental Outcomes of Extremely Preterm Infants Randomized to Stress Dose Hydrocortisone. 20 Proximity of magnesium exposure to delivery and neonatal outcomes. 21

The clinical outcomes of deep gray matter injury in children with cerebral palsy in relation with brain magnetic resonance imaging. 21

The ProVIDe study: the impact of protein intravenous nutrition on development in extremely low birthweight babies. 22

### Motricite – Mobilité - Posture

Muscle strength does not explain standing ability in children with bilateral spastic cerebral palsy: a cross sectional descriptive study. 22

Muscle synergies and complexity of neuromuscular control during gait in cerebral palsy. 23 Plantar flexor muscle weakness may cause stiff-knee gait. 23

Proximal Femoral Growth Modification: Effect of Screw, Plate, and Drill on Asymmetric Growth of the Hip. 24 Relationship between stature and tibial length for children with moderate-to-severe cerebral palsy. 24

The mechanics of activated semitendinosus are not representative of the pathological knee joint condition of children with cerebral palsy. 25

The impact of walking devices on kinematics in patients with spastic bilateral cerebral palsy. 25 Validation of an activity monitor for children who are partly or completely wheelchair-dependent. 25

### Traitement - Rééducation motrice et cognitive

#### Pharmacologie Efficacite Tolérance

Clinical Trial of Erythropoietin in Young Children With Cerebral Palsy. 26

Management of drooling in children with cerebral palsy: A French survey. 26

Intrathecal baclofen pumps do not accelerate progression of scoliosis in quadriplegic spastic cerebral palsy. 27 Intrathecal Baclofen Therapy in a Child With Severe Scoliosis: Report of 2 Cases. 27

Muscle histopathology in children with spastic cerebral palsy receiving botulinum toxin type A. 27

Safety profile of incobotulinum toxin A [Xeomin(\*)] in gastrocnemious muscles injections in children with cerebral palsy: Randomized double-blind clinical trial. 28

Treatment with botulinum toxin in children with cerebral palsy: a qualitative study of parents' experiences. 28

#### **Chirurgie**

Factors Contributing to Satisfaction with Changes in Physical Function after Orthopedic Surgery for Musculoskeletal Dysfunction in Patients with Cerebral Palsy. 29

Long-term outcomes after multilevel surgery including rectus femoris, hamstring and gastrocnemius procedures in children with cerebral palsy. 29

Long-term Outcomes of Triple Arthrodesis in Cerebral Palsy Patients. 30

Perioperative complications of orthopedic surgery for lower extremity in patients with cerebral palsy. 30 Salvage procedures for the painful chronically dislocated hip in cerebral palsy. 31

#### **Réadaptation fonctionnelle**

A Perceptual Motor Intervention Improves Play Behavior in Children with Moderate to Severe Cerebral Palsy. 31 A Prospective Case-Control Study of Radial Extracorporeal Shock Wave Therapy for Spastic Plantar Flexor Muscles in

Very Young Children With Cerebral Palsy. 32

Comparison of Rehabilitation Outcomes for Long Term Neurological Conditions: A Cohort Analysis of the Australian Rehabilitation Outcomes Centre Dataset for Adults of Working Age. 32

Development of an EMG-ACC-Based Upper Limb Rehabilitation Training System. 33

External Mechanical Work and Pendular Energy Transduction of Overground and Treadmill Walking in Adolescents with Unilateral Cerebral Palsy. 33

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FAST CP: protocol of a randomised controlled trial of the efficacy of a 12-week combined Functional Anaerobic and Strength Training programme on muscle properties and mechanical gait deficiencies in adolescents and young adults with spastic-type cerebral palsy. 33

Improvements in bimanual hand function after baby-CIMT in two-year old children with unilateral cerebral palsy: A retrospective study. 34

Increased Adaptation Rates and Reduction in Trial-by-Trial Variability in Subjects with Cerebral Palsy Following a Multi-session Locomotor Adaptation Training. 34

Reliability and Validity of Objective Measures of Physical Activity in Youth With Cerebral Palsy Who Are Ambulatory. 35

Single blind randomised controlled trial of GAME (Goals - Activity – Motor Enrichment) in infants at high risk of cerebral palsy. 35

The Effects of a Home-Based Connective Tissue Targeting Therapy on Hip Development in Children With Cerebral Palsy: Six Case Reports. 36

WalkAide Efficacy on Gait and Energy Expenditure in Children with Hemiplegic Cerebral Palsy: A Randomized Controlled Trial. 36

#### **Orthèse**

An individual approach for optimizing ankle-foot orthoses to improve mobility in children with spastic cerebral palsy walking with excessive knee flexion. 37

Minimising impairment: Protocol for a multicentre randomised controlled trial of upper limb orthoses for children with cerebral palsy. 37

#### Stimulation cérébrale - Stimulation neurosensorielle

Transcranial Direct-Current Stimulation Can Enhance Motor Learning in Children. 38

Transcranial direct current stimulation combined with integrative speech therapy in a child with cerebral palsy: A case report. 38

Transcranial direct current stimulation in children and adolescents: a comprehensive review. 38

#### Réalité virtuelle - Jeux video

Efficacy of neurodevelopmental treatment combined with the Nintendo(®) Wii in patients with cerebral palsy. 39

#### Thérapie cellulaire Médecine regénérative

Human Umbilical Cord Blood Cells Ameliorate Motor Deficits in Rabbits in a Cerebral Palsy Model. 39 How to make an oligodendrocyte. 40 Platelet-Rich Plasma in a Patient with Cerebral Palsy. 40

#### **Autres methodes**

Dog-assisted therapies and activities in rehabilitation of children with cerebral palsy and physical and mental disabilities. 40

Effects of Hippotherapy on Psychosocial Aspects in Children With Cerebral Palsy and Their Caregivers: A Pilot Study.

41

Horseback riding therapy in addition to conventional rehabilitation program decreases spasticity in children with cerebral palsy: A small sample study. 41

Laser acupuncture as an adjunctive therapy for spastic cerebral palsy in children. 42

### Langage -parole-communication

Mastery motivation in children with complex communication needs: longitudinal data analysis. 42

### Sommeil

Children with cerebral palsy: why are they awake at night? A pilot study. 42

### Autres Troubles / Troubles concomitants

#### **4** •Troubles musculosquelettiques, des tissue conjonctifs et osseux

Osteopenic features of the hip joint in patients with cerebral palsy: a hospital-based study. 43 Site-Specific Bone Mineral Density Is Unaltered Despite Differences in Fat-Free Soft Tissue Mass Between Affected and Nonaffected Sides in Hemiplegic Paralympic Athletes with Cerebral Palsy: Preliminary Findings. 43 The effect of anticonvulsant use on bone mineral density in non-ambulatory children with cerebral palsy. 44

#### **4** • Métabolisme

The Effect of Handcycle Ergometer Exercise on Glucose Tolerance in Ambulatory and Non-Ambulatory Adolescents.

44

#### •Troubles respiratoires

Factors Associated with Respiratory Illness in Children and Young Adults with Cerebral Palsy. 45 Tracheostomy in neurologically compromised paediatric patients: role of starplasty. 45

#### **4** • Nutrition – Troubles nutritionnels 45

Body mass index in ambulatory children with cerebral palsy: A cohort study. 45 Effects of maternal low-protein diet on parameters of locomotor activity in a rat model of cerebral palsy. 46 Effects of Neuromuscular Electrical Stimulation on the Masticatory Muscles and Physiologic Sleep Variables in Adults

with Cerebral Palsy: A Novel Therapeutic Approach. 46

Nutritional and health benefits of semi-elemental diets: A comprehensive summary of the literature. 47

#### **•**Troubles digestifs 47

Medical and surgical management of neurogenic bowel. 47

#### **4** •Sphère bucco-dentaire – Troubles de la déglutition

A telehealth approach to conducting clinical swallowing evaluations in children with cerebral palsy. 48 [Submandibular gland resection for the management of sialorrhea in paediatric patients with cerebral palsy and unresponsive to type A botullinum toxin. Pilot study].[48

#### **•**Troubles urinaires

Urodynamic Findings in Adults with Moderate to Severe Cerebral Palsy. 49

#### •Troubles de la vision

Ophthalmic profile and systemic features of pediatric facial nerve palsy. 49

#### **•**Troubles de l'audition

Comparison of Auditory Perception in Cochlear Implanted Children with and without Additional Disabilities. 50

### Qualité de vie et rapport au monde

Burden of Neurological Conditions in Canada. 50

#### Vie quotidienne

Activities and participation of children with cerebral palsy: parent perspectives. 51 Development of work participation in young adults with cerebral palsy: a longitudinal study. 51 Physical Accessibility of Routine Prenatal Care for Women with Mobility Disability. 52 Upper limb function in everyday life of children with cerebral palsy: description and review of parent report

measures. 52

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### **4** • Activité physique - Sport

Cardiovascular diseases in Paralympic athletes. 53

Comparison of Patterns of Physical Activity and Sedentary Behavior Between Children With Cerebral Palsy and Children With Typical Development. 53

Health-related physical fitness of ambulatory adolescents and young adults with spastic cerebral palsy. 54

Metabolic, cardiorespiratory, and neuromuscular fitness performance in children with cerebral palsy: A comparison with healthy youth. 54

### **4** •Vieillissement - Passage à l'âge adulte

Extrapolating published survival curves to obtain evidence-based estimates of life expectancy in cerebral palsy. 55

#### **4** • Prise en charge et Accompagnement

Musculoskeletal system pain and related factors in mothers of children with cerebral palsy. 55

## Manifestations et congrès

#### Juin 2016

#### International Conference on Cerebral Palsy and Other Childhood-onset Disabilities

#### Joint meeting

5Th International Conference of Cerebral palsy (ICPC° 28th Annual Meeting of the European Academy of Childhood Disability (EACD) 1 st Biennal meeting od the International Alliance of Academies od Childhood Disability (IAACD) 1-4 Juin 2016 Suède, Stockholm <u>http://eacd2016.org/</u>

#### Septembre 2016

#### **6th International Conference on Clinical Neonatology**

22-24 Spetembre 2016 Turin, Italie https://www.eiseverywhere.com/ehome/105597/234360/

#### 21ème Congrès du GEIMOC

24 Septembre 2016 Liège, BELGIQUE

#### Octobre 2016

#### 31ème Congrès de la Société française de Médecine Physique et de réadpatation (SOFMER)

13-15 Octobre 2016 Saint Etienne, France http://saint-etienne.sofmer2016.com/

## **Publications scientifiques**

#### Cerebral palsy.

Graham HK, Rosenbaum P, Paneth N, Dan B, Lin JP, Damiano DL, Becher JG, Gaebler-Spira D, Colver A, Reddihough DS, Crompton KE, Lieber RL.

Nat Rev Dis Primers. 2016 Jan 7;2:15082. doi: 10.1038/nrdp.2015.82.

Cerebral palsy is the most common cause of childhood-onset, lifelong physical disability in most countries, affecting about 1 in 500 neonates with an estimated prevalence of 17 million people worldwide. Cerebral palsy is not a disease entity in the traditional sense but a clinical description of children who share features of a non-progressive brain injury or lesion acquired during the antenatal, perinatal or early postnatal period. The clinical manifestations of cerebral palsy vary greatly in the type of movement disorder, the degree of functional ability and limitation and the affected parts of the body. There is currently no cure, but progress is being made in both the prevention and the amelioration of the brain injury. For example, administration of magnesium sulfate during premature labour and cooling of high-risk infants can reduce the rate and severity of cerebral palsy. Although the disorder affects individuals throughout their lifetime, most cerebral palsy research efforts and management strategies currently focus on the needs of children. Clinical management of children with cerebral palsy is directed towards maximizing function and participation in activities and minimizing the effects of the factors that can make the condition worse, such as epilepsy, feeding challenges, hip dislocation and scoliosis. These management strategies include enhancing neurological function during early development; managing medical co-morbidities, weakness and hypertonia; using rehabilitation technologies to enhance motor function; and preventing secondary musculoskeletal problems. Meeting the needs of people with cerebral palsy in resource-poor settings is particularly challenging. PMID: 27188686 [PubMed - in process]

### Epidémiologie

Bangladesh Cerebral Palsy Register (BCPR): a pilot study to develop a national cerebral palsy (CP) register with surveillance of children for CP.

Khandaker G, Smithers-Sheedy H, Islam J, Alam M, Jung J, Novak I, Booy R, Jones C, Badawi N, Muhit M. BMC Neurol. 2015 Sep 25;15:173. doi: 10.1186/s12883-015-0427-9.

BACKGROUND: The causes and pathogenesis of cerebral palsy (CP) are all poorly understood, particularly in low- and middle-income countries (LMIC). There are gaps in knowledge about CP in Bangladesh, especially in the spheres of epidemiological research, intervention and service utilization. In high-income countries CP registers have made substantial contributions to our understanding of CP. In this paper, we describe a pilot study protocol to develop, implement, and evaluate a CP population register in Bangladesh (i.e., Bangladesh Cerebral Palsy Register - BCPR) to facilitate studies on prevalence, severity, aetiology, associated impairments and risk factors for CP.

METHODS/DESIGN: The BCPR will utilise a modified version of the Australian Cerebral Palsy Register (ACPR) on a secured web-based platform hosted by the Cerebral Palsy Alliance Research Institute, Australia. A standard BCPR record form (i.e., data collection form) has been developed in consultation with local and international experts. Using this form, the BPCR will capture information about maternal health, birth history and the nature of disability in all children with CP aged <18 years. The pilot will be conducted in the Shahjadpur sub-district of Sirajgonj district in the northern part of Bangladesh. There are 296 villages in Shahjadpur, a total population of 561,076 (child population ~ 226,114), an estimated 70,998 households and 12,117 live births per annum. Children with CP will be identified by using the community based Key Informants Method (KIM). Data from the completed BPCR record together with details of assessment by a research physician will be entered into an online data repository.

DISCUSSION: Once implemented, BCPR will be, to the best of our knowledge, the first formalised CP register from a LMIC. Establishment of the BCPR will enable estimates of prevalence; facilitate clinical surveillance and promote research to improve the care of individuals with CP in Bangladesh.

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### **4**Prévalence-Incidence

#### Birth Prevalence of Cerebral Palsy: A Population-Based Study.

Van Naarden Braun K(1), Doernberg N(2), Schieve L(2), Christensen D(2), Goodman A(2), Yeargin-Allsopp M(2). *Pediatrics. 2016 Jan;137(1). doi: 10.1542/peds.2015-2872. Epub 2015 Dec 9.* 

OBJECTIVE: Population-based data in the United States on trends in cerebral palsy (CP) birth prevalence are limited. The objective of this study was to examine trends in the birth prevalence of congenital spastic CP by birth weight, gestational age, and race/ethnicity in a heterogeneous US metropolitan area.

METHODS: Children with CP were identified by a population-based surveillance system for developmental disabilities (DDs). Children with CP were included if they were born in metropolitan Atlanta, Georgia, from 1985 to 2002, resided there at age 8 years, and did not have a postneonatal etiology (n = 766). Birth weight, gestational age, and race/ethnicity subanalyses were restricted to children with spastic CP (n = 640). Trends were examined by CP subtype, gender, race/ethnicity, co-occurring DDs, birth weight, and gestational age.

RESULTS: Birth prevalence of spastic CP per 1000 1-year survivors was stable from 1985 to 2002 (1.9 in 1985 to 1.8 in 2002; 0.3% annual average prevalence; 95% confidence interval [CI] -1.1 to 1.8). Whereas no significant trends were observed by gender, subtype, birth weight, or gestational age overall, CP prevalence with co-occurring moderate to severe intellectual disability significantly decreased (-2.6% [95% CI -4.3 to -0.8]). Racial disparities persisted over time between non-Hispanic black and non-Hispanic white children (prevalence ratio 1.8 [95% CI 1.5 to 2.1]). Different patterns emerged for non-Hispanic white and non-Hispanic black children by birth weight and gestational age.

CONCLUSIONS: Given improvements in neonatal survival, evidence of stability of CP prevalence is encouraging. Yet lack of overall decreases supports continued monitoring of trends and increased research and prevention efforts. Racial/ethnic disparities, in particular, warrant further study.

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#### Free PMC Article

PMCID: PMC4703497 PMID: 26659459 [PubMed - indexed for MEDLINE]

## Prevalence of Cerebral Palsy among 8-Year-Old Children in 2010 and Preliminary Evidence of Trends in Its Relationship to Low Birthweight.

Durkin MS, Benedict RE, Christensen D, Dubois LA, Fitzgerald RT, Kirby RS, Maenner MJ, Van Naarden Braun K, Wingate MS, Yeargin-Allsopp M.

Paediatr Perinat Epidemiol. 2016 May 23. doi: 10.1111/ppe.12299. [Epub ahead of print]

BACKGROUND: The public health objective for cerebral palsy (CP) in the United States is to reduce the percentage of children with CP who were born low birthweight (LBW, <2500 g) by 10% between 2006 and 2020. This study reports the prevalence of CP in a constant surveillance area for the years 2006, 2008, and 2010 and describes initial progress towards the CP public health objective.

METHODS: Data on children with CP at age 8 years were ascertained by the Autism and Developmental Disabilities Monitoring (ADDM) Network, a population-based surveillance system that monitored CP in four areas of the United States.

RESULTS: CP prevalence in 2010 was 2.9 per 1000 [95% confidence interval (Cl) 2.6, 3.2], down from 3.5 (95% Cl 3.2, 3.9) in the same surveillance area in 2006. Among CP cases with no documented postneonatal aetiology, 49.1% (95% Cl 42.9, 55.2) were born LBW in 2010 compared with 54.3% (95% Cl 48.4, 60.1) in 2006. In 2010, 28.1% (95% Cl 22.9, 30.4) were born very low birthweight (VLBW, <1500 g) compared with 35.4% (95% Cl 30.0, 41.2) in 2006. The relative risks for associations between CP and both LBW and VLBW also declined, though not significantly, during the study period.

CONCLUSIONS: Declines in the associations between CP and LBW categories may have contributed to declines during the study period in both the prevalence of CP and the percentage of children with CP who were born LBW or VLBW. Ongoing monitoring of these trends is warranted.

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PMID: 27215680 [PubMed - as supplied by publisher]

### **4**Facteurs de risque – Causes

#### Asphyxia, Neurologic Morbidity, and Perinatal Mortality in Early-Term and Postterm Birth.

Seikku L, Gissler M, Andersson S, Rahkonen P, Stefanovic V, Tikkanen M, Paavonen J, Rahkonen L. *Pediatrics. 2016 May 27. pii: e20153334. [Epub ahead of print]* 

BACKGROUND AND OBJECTIVES: Neonatal outcomes vary by gestational age. We evaluated the association of earlyterm, full-term, and postterm birth with asphyxia, neurologic morbidity, and perinatal mortality.

METHODS: Our register-based study used retrospective data on 214 465 early-term (37(+0)-38(+6) gestational weeks), 859 827 full-term (39(+0)-41(+6)), and 55 189 postterm ( $\geq$ 42(+0)) live-born singletons during 1989-2008 in Finland. Asphyxia parameters were umbilical cord pH and Apgar score at 1 and 5 minutes. Neurologic morbidity outcome measures were cerebral palsy (CP), epilepsy, intellectual disability, and sensorineural defects diagnosed by the age of 4 years. Newborns with major congenital anomalies were excluded from perinatal deaths.

RESULTS: Multivariate analysis showed that, compared with full-term pregnancies, early-term birth increased the risk for low Apgar score (<4) at 1 and 5 minutes (odds ratio 1.03, 95% confidence interval 1.03-1.04 and 1.24, 1.04-1.49, respectively), CP (1.40, 1.27-1.55), epilepsy (1.14, 1.06-1.23), intellectual disability (1.39, 1.27-1.53), sensorineural defects (1.24, 1.17-1.31), and perinatal mortality (2.40, 2.14-2.69), but risk for low umbilical artery pH  $\leq$ 7.10 was decreased (0.83, 0.79-0.87). Postterm birth increased the risk for low Apgar score (<4) at 1 minute (1.26, 1.26-1.26) and 5 minutes (1.80, 1.43-2.34), low umbilical artery pH  $\leq$ 7.10 (1.26, 1.19-1.34), and intellectual disability (1.19, 1.00-1.43), whereas risks for CP (1.03, 0.84-1.26), epilepsy (1.00, 0.87-1.15), sensorineural defects (0.96, 0.86-1.07), and perinatal mortality (0.91, 0.69-1.22) were not increased.

CONCLUSIONS: Early-term birth was associated with low Apgar score, increased neurologic morbidity, and perinatal mortality. Asphyxia and intellectual disability were more common among postterm births, but general neurologic morbidity and perinatal mortality were not increased.

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PMID: 27235446 [PubMed - as supplied by publisher]

#### Appearance of neurodevelopmental disorders in children delivered post-term: a cross-section study.

Vukojevic M, Trninic I, Dodaj A, Malenica M, Barisic T, Stojic S. Mater Sociomed. 2016 Apr;28(2):99-103. doi: 10.5455/msm.2016.28.99-103. Epub 2016 Mar 25.

GOAL: To analyze the appearance of neurodevelopmental disorders in children delivered post-term and to find out whether prolonged pregnancy may be a cause of such disorders in a selected group participants.

PATIENTS AND METHODS: This study included a cohort of 34 children born post-term suffering from neurodevelopmental disorders who were treated at the Service for psycho-physiological and speaking disorders in Mostar, Bosnia and Herzegovina during an 18-year period.

RESULTS: There were 59.4% of male and 40.6% female patients (P=0.002). The most common neurodevelopmental disorder in the sample was intellectual disability (38.2%), followed by epilepsy (26.4%), delayed psychomotor development (14.7%), and cerebral palsy (11.7%) (P<0.001). The correlation between mothers' parity and post-term delivery was found (P=0.016).

CONCLUSION: Post-term delivery may be the cause of neurodevelopmental disorders. The most common disorder among them were intellectual difficulties.

Free PMC Article

PMCID: PMC4851530 PMID: 27147913 [PubMed]

#### Early onset preeclampsia and cerebral palsy: a double hit model?

Science Infos Paralysie Cérébrale, mai 2016, FONDATION MOTRICE 67 rue Vergniaud 75013 Paris - tel +33 1 10 45 54 03 03 contact: Christine Doumergue cdoumergue@lafondationmotrice.org

Mor O, Stavsky M, Yitshak-Sade M, Mastrolia SA, Beer-Weisel R, Rafaeli-Yehudai T, Besser L, Hamou B, Mazor M, Erez O

Am J Obstet Gynecol. 2016 Jan;214(1):105.e1-9. doi: 10.1016/j.ajog.2015.08.020. Epub 2015 Aug 15.

BACKGROUND: Cerebral palsy (CP) is a late sequel of pregnancy, and the role of preeclampsia is debatable.

OBJECTIVE: The aims of this study were to determine the association between

preeclampsia and cerebral palsy and to determine the risk factors for the development of cerebral palsy in these patients.

STUDY DESIGN: A retrospective population-based cohort study was designed that included 229,192 singleton pregnancies. The study population was divided into 2 groups: (1) patients with preeclampsia (n = 9749) and (2) normotensive gestations (n = 219,443). Generalized Estimating Equation multiple logistic regression models were performed to study the associations among preeclampsia, small for gestational age, gestational age at delivery, and the risk factors for the development of cerebral palsy in neonates of women with preeclampsia.

RESULTS: The rate of cerebral palsy was double in patients with preeclampsia than in the normotensive group (0.2% vs 0.1%; P = .015); early onset preeclampsia and small for gestational age were independent risk factors for the subsequent development of cerebral palsy (odds ratio, 8.639 [95% confidence interval, 4.269-17.480]; odds ratio, 2.737 [95% confidence interval, 1.937-3.868], respectively). A second model was conducted to determine the risk factors for the development of cerebral palsy in women with preeclampsia. Birth asphyxia, complications of prematurity, and neonatal infectious morbidity, but not small for gestational age or gestational age at delivery, were independent risk factors for the development of cerebral palsy.

CONCLUSION: In a comparison with normal pregnant women, the rate of cerebral palsy is double among patients with preeclampsia, especially those with early-onset disease. Early-onset preeclampsia is an independent risk factor for cerebral palsy. Among women with preeclampsia, the presence of neonatal infectious morbidity, birth asphyxia, and complications of prematurity are independent risk factors for the development of cerebral palsy, which further supports the role of a multi-hit model in the pathogenesis of this syndrome.

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PMID: 26283455 [PubMed - indexed for MEDLINE]

Increased nuchal translucency thickness and the risk of neurodevelopmental disorders. Hellmuth SG, Pedersen LH, Miltoft CB, Petersen OB, Kjaergaard S, Ekelund C, Tabor A. *Ultrasound Obstet Gynecol. 2016 May 17. doi: 10.1002/uog.15961. [Epub ahead of print]* 

OBJECTIVE: To investigate the association between fetal nuchal translucency thickness (NT) and neurodevelopmental disorders in euploid children.

METHOD: This study included 222,505 euploid children, with a routine first trimester screening. They were divided into three groups by NT thickness: NT <95(th) percentile, 217,103 (97.6%), NT 95(th) -99(th) percentile, 4,760 (2.1%), and NT >99(th) percentile 642 (0.3%). All children were followed-up through national patient registries at a mean age of 4.4 years. We obtained information on diagnoses of mental retardation, autism spectrum disorders (ASDs), cerebral palsy, epilepsy and febrile seizures.

RESULTS: There was no excess risk of neurodevelopmental disorders among euploid children with a NT between 95(th) and 99(th) percentile. For children with a NT >99(th) percentile, there were increased risks of mental retardation (OR=6.16, 95% confidence interval (CI): 1.51-25.0, 2 cases) and ASDs (OR=2.48, 95%CI: 1.02-5.99, 5 cases) compared to children with a NT<95(th) percentile (110 and 686 cases of mental retardation and ASDs, respectively). There was no detected increased risk of cerebral palsy (OR= 1.91, 95%CI: 0.61-5.95, 3 cases), epilepsy (OR= 1.51, 95%CI: 0.63-3.66, 5 cases) or febrile seizures (OR=0.72, 95%CI: 0.44-1.16, 17 cases) among children with a NT >99(th) percentile.

CONCLUSION: In a large unselected cohort, euploid fetuses with a NT 95(th) -99(th) percentile had no increased risk of neurodevelopmental disorders. Among euploid fetuses with a NT >99(th) percentile there was an increased risk of mental retardation and ASDs but the absolute risk was low (<1%) and thus reassuring.

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PMID: 27183961 [PubMed - as supplied by publisher]

#### Neonatal brain injury as a consequence of insufficient cerebral oxygenation.

Placha K, Luptakova D, Baciak L, Ujhazy E, Juranek I.

*Neuro Endocrinol Lett. 2016 May 15;37(2):79-96. [Epub ahead of print]* Science Infos Paralysie Cérébrale, mai 2016, FONDATION MOTRICE 67 rue Vergniaud 75013 Paris - tel +33 1 11 45 54 03 03 contact: Christine Doumergue cdoumergue@lafondationmotrice.org Neonatal brain hypoxic-ischemic injury represents a serious health care and socio-economical problem since it is one of the most common causes of mortality and morbidity of newborns. Neonatal hypoxic-ischemic encephalopathy is often associated with signs of perinatal asphyxia, with an incidence of about 2-4 per 1,000 live births and mortality rate up to 20%. In about one half of survivors, cerebral hypoxic-ischemic insult may result in more or less pronounced neuro-psychological sequelae of immediate or delayed nature, such as seizures, cerebral palsy or behavioural and learning disabilities, including attention-deficit hyperactivity disorder. Hypoxic-ischemic injury develops as a consequence of transient or permanent restriction of blood supply to the brain. Severity of hypoxicischemic encephalopathy varies depending on the intensity and duration of hypoxia-ischemia, on the type and size of the brain region affected, and on the maturity of the foetal/neonatal brain. Though a primary cause of hypoxicischemic injury is lack of oxygen in the neonatal brain, underlying mechanisms of subsequent events that are critical for developing hypoxic-ischemic encephalopathy are less understood. Their understanding is however necessary for elaborating effective management for newborns that underwent cerebral hypoxic-ischemic insult and thus are at risk of a negative outcome. The present paper summarizes current knowledge on cerebral hypoxic-ischemic injury of the neonate, fundamental processes involved in etiopathogenesis, with a special focus on cellular and molecular mechanisms and particular attention on certain controversial aspects of oxidative stress involvement. PMID: 27179569 [PubMed - as supplied by publisher]

#### The neurological outcomes of cerebellar injury in premature infants.

Kobayashi S, Wakusawa K, Inui T, Tanaka S, Kobayashi Y, Onuma A, Haginoya K. Brain Dev. 2015 Oct;37(9):858-63. doi: 10.1016/j.braindev.2015.01.009. Epub 2015 Feb 13.

AIM: Cerebellar injury is a characteristic injury associated with preterm infants. However, the impact of cerebellar injury on the development of preterm infants is unclear.

METHOD: We reviewed magnetic resonance image studies of preterm infants with cerebral palsy retrospectively and evaluated the developmental outcomes.

RESULTS: Cerebellar injury was recognized in 9 (2.4%) of 381 patients with cerebral palsy who were born preterm. The median gestational age was 26 (range 23-32) weeks and the median birth weight was 938 (range 492-1450) g. Seven of the nine patients had severe symmetric injuries to the inferior cerebellar hemispheres, resulting in a pancake-like appearance of the residual upper cerebellum. Supratentorial lesions were also recognized: periventricular leukomalacia in seven; atrophy of the basal ganglia in two; and intraventricular hemorrhage in two. Importantly, the motor dysfunction was related to the reduction in the white matter volume and severity of basal ganglia atrophy, but not to the cerebellar injury. Four of the nine patients could walk without limitations despite extensive cerebellar disruption. Only four patients could speak meaningful words during the study and only one spoke two-word sentences.

INTERPRETATION: The patients with cerebellar injury might have a communication handicap, rather than altered motor function. Prematurity-related cerebellar complications require more attention in terms of cognitive and speech function, in addition to neuromotor development.

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PMID: 25684440 [PubMed - indexed for MEDLINE]

## Weight Status in the First 2 Years of Life and Neurodevelopmental Impairment in Extremely Low Gestational Age Newborns.

Belfort MB, Kuban KC, O'Shea TM, Allred EN, Ehrenkranz RA, Engelke SC, Leviton A; Extremely Low Gestational Age Newborn (ELGAN) Study Investigators; Extremely Low Gestational Age Newborn ELGAN Study Investigators. *J Pediatr. 2016 Jan;168:30-5.e2. doi: 10.1016/j.jpeds.2015.09.036. Epub 2015 Oct 21.* 

OBJECTIVE: To examine the extent to which weight gain and weight status in the first 2 years of life relate to the risk of neurodevelopmental impairment in extremely preterm infants.

STUDY DESIGN: In a cohort of 1070 infants born between 23 and 27 weeks' gestation, we examined weight gain from 7-28 days of life (in quartiles) and weight z-score at 12 and 24 months corrected age (in 4 categories: <-2;  $\geq$ -2, <-1;  $\geq$ 1, <1; and  $\geq$ 1) in relation to these adverse neurodevelopmental outcomes: Bayley-II mental development index <55, Bayley-II psychomotor development index <55, cerebral palsy, Gross Motor Function Classification System  $\geq$ 1

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(cannot walk without assistance), microcephaly. We adjusted for confounders in logistic regression, stratified by sex, and performed separate analyses including the entire sample, and excluding children unable to walk without assistance (motor impairment).

RESULTS: Weight gain in the lowest quartile from 7-28 days was not associated with higher risk of adverse outcomes. Children with a 12-month weight z-score <-2 were at increased risk for all adverse outcomes in girls, and for microcephaly and Gross Motor Function Classification System  $\geq 1$  in boys. However, excluding children with motor impairment attenuated all associations except that of weight z-score <-2 with microcephaly in girls. Similarly, most associations of low weight z-score at 24 months with adverse outcomes were attenuated with exclusion of children with motor impairment.

CONCLUSION: Excluding children who have gross motor impairment appears to eliminate the association of low weight status with neurodevelopmental impairments at 2 years in extremely preterm infants.

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PMID: 26470687 [PubMed - indexed for MEDLINE]

### **4**Génétique

Genetic factors may underlie many cerebral palsy cases: New research implicates genetic variations, not lack of oxygen at birth, in disorder.

[No authors listed] *Am J Med Genet A. 2015 Sep;167A(9):vii-viii. doi: 10.1002/ajmg.a.37283.* PMID: 26288074 [PubMed - indexed for MEDLINE]

### Lésions Prévention des lesions

### Données fondamentales

## Hyaluronidase and Hyaluronan Oligosaccharides Promote Neurological Recovery after Intraventricular Hemorrhage.

Vinukonda G, Dohare P, Arshad A, Zia MT, Panda S, Korumilli R, Kayton R, Hascall VC, Lauer ME, Ballabh P. J Neurosci. 2016 Jan 20;36(3):872-89. doi: 10.1523/JNEUROSCI.3297-15.2016.

Intraventricular hemorrhage (IVH) in premature infants results in inflammation, arrested oligodendrocyte progenitor cell (OPC) maturation, and reduced myelination of the white matter. Hyaluronan (HA) inhibits OPC maturation and complexes with the heavy chain (HC) of glycoprotein inter- $\alpha$ -inhibitor to form pathological HA (HC-HA complex), which exacerbates inflammation. Therefore, we hypothesized that IVH would result in accumulation of HA, and that either degradation of HA by hyaluronidase treatment or elimination of HCs from pathological HA by HA oligosaccharide administration would restore OPC maturation, myelination, and neurological function in survivors with IVH. To test these hypotheses, we used the preterm rabbit model of glycerol-induced IVH and analyzed autopsy samples from premature infants. We found that total HA levels were comparable in both preterm rabbit pups and human infants with and without IVH, but HA receptors--CD44, TLR2, TLR4--were elevated in the forebrain of both humans and rabbits with IVH. Hyaluronidase treatment of rabbits with IVH reduced CD44 and TLR4 expression, proinflammatory cytokine levels, and microglia infiltration. It also promoted OPC maturation, myelination, and neurological recovery. HC-HA and tumor necrosis factor-stimulated gene-6 were elevated in newborns with IVH; and depletion of HC-HA levels by HA oligosaccharide treatment reduced inflammation and enhanced myelination and neurological recovery in rabbits with IVH. Hence, hyaluronidase or HA oligosaccharide treatment represses inflammation, promotes OPC maturation, and restores myelination and neurological function in rabbits with IVH. These therapeutic strategies might improve the neurological outcome of premature infants with IVH. Significance statement: Approximately 12,000 premature infants develop IVH every year in the United States, and a large number of survivors with IVH develop cerebral palsy and cognitive deficits. The onset of IVH induces inflammation of the periventricular white matter, which results in arrested maturation of OPCs and myelination failure. HA is a major component of the extracellular matrix of the brain, which regulates inflammation through CD44 and TLR2/4 Science Infos Paralysie Cérébrale, mai 2016, FONDATION MOTRICE 67 rue Vergniaud 75013 Paris - tel +33 1 13 45 54 03 03 contact: Christine Doumergue cdoumergue@lafondationmotrice.org

receptors. Here, we show two mechanism-based strategies that effectively enhanced myelination and neurological recovery in preterm rabbit model of IVH. First, degrading HA by hyaluronidase treatment reduced CD44 and TLR4 expression, proinflammatory cytokines, and microglial infiltration, as well as promoted oligodendrocyte maturation and myelination. Second, intraventricular injection of HA oligosaccharide reduced inflammation and enhanced myelination, conceivably by depleting HC-HA levels.

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PMCID: PMC4719021 [Available on 2016-07-20]

PMID: 26791217 [PubMed - indexed for MEDLINE]

#### Human Amnion Epithelial Cells Modulate Ventilation-Induced White Matter Pathology in Preterm Lambs.

Barton SK(1), Melville JM, Tolcos M, Polglase GR, McDougall AR, Azhan A, Crossley KJ, Jenkin G, Moss TJ. Dev Neurosci. 2015;37(4-5):338-48. doi: 10.1159/000371415. Epub 2015 Feb 25.

BACKGROUND: Preterm infants can be inadvertently exposed to high tidal volumes (VT) during resuscitation in the delivery room due to limitations of available equipment. High VT ventilation of preterm lambs produces cerebral white matter (WM) pathology similar to that observed in preterm infants who develop cerebral palsy. We hypothesized that human amnion epithelial cells (hAECs), which have anti-inflammatory and regenerative properties, would reduce ventilation-induced WM pathology in neonatal late preterm lamb brains.

METHODS: Two groups of lambs (0.85 gestation) were used, as follows: (1) ventilated lambs (Vent; n = 8) were ventilated using a protocol that induces injury (VT targeting 15 ml/kg for 15 min, with no positive end-expiratory pressure) and were then maintained for another 105 min, and (2) ventilated + hAECs lambs (Vent+hAECs; n = 7) were similarly ventilated but received intravenous and intratracheal administration of  $9 \times 10(7)$  hAECs ( $18 \times 10(7)$  hAECs total) at birth. Oxygenation and ventilation parameters were monitored in real time; cerebral oxygenation was measured using near-infrared spectroscopy. qPCR (quantitative real-time PCR) and immunohistochemistry were used to assess inflammation, vascular leakage and astrogliosis in both the periventricular and subcortical WM of the frontal and parietal lobes. An unventilated control group (UVC; n = 5) was also used for qPCR analysis of gene expression. Two-way repeated measures ANOVA was used to compare physiological data. Student's t test and one-way ANOVA were used for immunohistological and qPCR data comparisons, respectively.

RESULTS: Respiratory parameters were not different between groups. Interleukin (IL)-6 mRNA levels in subcortical WM were lower in the Vent+hAECs group than the Vent group (p = 0.028). IL-1 $\beta$  and IL-6 mRNA levels in periventricular WM were higher in the Vent+hAECs group than the Vent group (p = 0.007 and p = 0.001, respectively). The density of Iba-1-positive microglia was lower in the subcortical WM of the parietal lobes (p = 0.010) in the Vent+hAECs group but not in the periventricular WM. The number of vessels in the WM of the parietal lobe exhibiting protein extravasation was lower (p = 0.046) in the Vent+hAECs group. Claudin-1 mRNA levels were higher in the periventricular WM (p = 0.005). The density of GFAP-positive astrocytes was not different between groups.

CONCLUSIONS: Administration of hAECs at the time of birth alters the effects of injurious ventilation on the preterm neonatal brain. Further studies are required to understand the regional differences in the effects of hAECs on ventilation-induced WM pathology and their net effect on the developing brain.

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PMID: 25720586 [PubMed - indexed for MEDLINE]

## In vivo quantification of intraventricular hemorrhage in a neonatal piglet model using an EEG-layout based electrical impedance tomography array.

Tang T, Weiss MD, Borum P, Turovets S, Tucker D, Sadleir R. *Physiol Meas. 2016 May 20;37(6):751-764.* [Epub ahead of print]

Intraventricular hemorrhage (IVH) is a common occurrence in the days immediately after premature birth. It has been correlated with outcomes such as periventricular leukomalacia (PVL), cerebral palsy and developmental delay. The causes and evolution of IVH are unclear; it has been associated with fluctuations in blood pressure, damage to the subventricular zone and seizures. At present, ultrasound is the most commonly used method for detection of IVH, but is used retrospectively. Without the presence of adequate therapies to avert IVH, the use of a continuous monitoring technique may be somewhat moot. While treatments to mitigate the damage caused by IVH are still under development, the principal benefit of a continuous monitoring technique will be in investigations into the etiology of IVH, and its associations with periventricular injury and blood pressure fluctuations. Electrical impedance Science Infos Paralysie Cérébrale, mai 2016, FONDATION MOTRICE 67 rue Vergniaud 75013 Paris - tel +33 1 14 45 54 03 03 contact: Christine Doumergue cdoumergue@lafondationmotrice.org

tomography (EIT) is potentially of use in this context as accumulating blood displaces higher conductivity cerebrospinal fluid (CSF) in the ventricles. We devised an electrode array and EIT measurement strategy that performed well in detection of simulated ventricular blood in computer models and phantom studies. In this study we describe results of pilot in vivo experiments on neonatal piglets, and show that EIT has high sensitivity and specificity to small quantities of blood (<1 ml) introduced into the ventricle. EIT images were processed to an index representing the quantity of accumulated blood (the 'quantity index', QI). We found that QI values were linearly related to fluid quantity, and that the slope of the curve was consistent between measurements on different ubjects. Linear discriminant analysis showed a false positive rate of 0%, and receiver operator characteristic analysis found area under curve values greater than 0.98 to administered volumes between 0.5, and 2.0 ml. We believe our study indicates that this method may be well suited to quantitative monitoring of IVH in newborns, simultaneously or interleaved with electroencephalograph assessments.

PMID: 27206102 [PubMed - as supplied by publisher]

## Stromal Cell-Derived Factor-1 $\alpha$ Plays a Crucial Role Based on Neuroprotective Role in Neonatal Brain Injury in Rats.

Mori M, Matsubara K, Matsubara Y, Uchikura Y, Hashimoto H, Fujioka T, Matsumoto T. Int J Mol Sci. 2015 Aug 5;16(8):18018-32. doi: 10.3390/ijms160818018.

Owing to progress in perinatal medicine, the survival of preterm newborns has markedly increased. However, the incidence of cerebral palsy has risen in association with increased preterm birth. Cerebral palsy is largely caused by cerebral hypoxic ischemia (HI), for which there are no effective medical treatments. We evaluated the effects of stromal cell-derived factor- $1\alpha$  (SDF- $1\alpha$ ) on neonatal brain damage in rats. Left common carotid (LCC) arteries of seven-day-old Wistar rat pups were ligated, and animals were exposed to hypoxic gas to cause cerebral HI. Behavioral tests revealed that the memory and spatial perception abilities were disturbed in HI animals, and that SDF- $1\alpha$  treatment improved these cognitive functions. Motor coordination was also impaired after HI but was unimproved by SDF- $1\alpha$  treatment. SDF- $1\alpha$  reduced intracranial inflammation and induced cerebral remyelination, as indicated by the immunohistochemistry results. These data suggest that SDF- $1\alpha$  specifically influences spatial perception abilities in neonatal HI encephalopathy.

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PMCID: PMC4581233 PMID: 26251894 [PubMed - indexed for MEDLINE]

#### Rodent Hypoxia-Ischemia Models for Cerebral Palsy Research: A Systematic Review.

Rumajogee P, Bregman T, Miller SP, Yager JY, Fehlings MG. Front Neurol. 2016 Apr 25;7:57. doi: 10.3389/fneur.2016.00057. eCollection 2016.

Cerebral palsy (CP) is a complex multifactorial disorder, affecting approximately 2.5-3/1000 live term births, and up to 22/1000 prematurely born babies. CP results from injury to the developing brain incurred before, during, or after birth. The most common form of this condition, spastic CP, is primarily associated with injury to the cerebral cortex and subcortical white matter as well as the deep gray matter. The major etiological factors of spastic CP are hypoxia/ischemia (HI), occurring during the last third of pregnancy and around birth age. In addition, inflammation has been found to be an important factor contributing to brain injury, especially in term infants. Other factors, including genetics, are gaining importance. The classic Rice-Vannucci HI model (in which 7-day-old rat pups undergo unilateral ligation of the common carotid artery followed by exposure to 8% oxygen hypoxic air) is a model of neonatal stroke that has greatly contributed to CP research. In this model, brain damage resembles that observed in severe CP cases. This model, and its numerous adaptations, allows one to finely tune the injury parameters to mimic, and therefore study, many of the pathophysiological processes and conditions observed in human patients. Investigators can recreate the HI and inflammation, which cause brain damage and subsequent motor and cognitive deficits. This model further enables the examination of potential approaches to achieve neural repair and regeneration. In the present review, we compare and discuss the advantages, limitations, and the translational value for CP research of HI models of perinatal brain injury.

Free PMC Article PMCID: PMC4843764 PMID: 27199883 [PubMed]

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#### Systemic dendrimer-drug treatment of ischemia-induced neonatal white matter injury.

Nance E, Porambo M, Zhang F, Mishra MK, Buelow M, Getzenberg R, Johnston M, Kannan RM, Fatemi A, Kannan S J Control Release. 2015 Sep 28;214:112-20. doi: 10.1016/j.jconrel.2015.07.009. Epub 2015 Jul 13.

Extreme prematurity is a major risk factor for perinatal and neonatal brain injury, and can lead to white matter injury that is a precursor for a number of neurological diseases, including cerebral palsy (CP) and autism. Neuroinflammation, mediated by activated microglia and astrocytes, is implicated in the pathogenesis of neonatal brain injury. Therefore, targeted drug delivery to attenuate neuroinflammation may greatly improve therapeutic outcomes in models of perinatal white matter injury. In this work, we use a mouse model of ischemia-induced neonatal white matter injury to study the biodistribution of generation 4, hydroxyl-functionalized polyamidoamine dendrimers. Following systemic administration of the Cy5-labeled dendrimer (D-Cy5), we demonstrate dendrimer uptake in cells involved in ischemic injury, and in ongoing inflammation, leading to secondary injury. The sub-acute response to injury is driven by astrocytes. Within five days of injury, microglial proliferation and migration occurs, along with limited differentiation of oligodendrocytes and oligodendrocyte death. From one day to five days after injury, a shift in dendrimer co-localization occurred. Initially, dendrimer predominantly co-localized with astrocytes, with a subsequent shift towards microglia. Co-localization with oligodendrocytes reduced over the same time period, demonstrating a region-specific uptake based on the progression of the injury. We further show that systemic administration of a single dose of dendrimer-N-acetyl cysteine conjugate (D-NAC) at either sub-acute or delayed time points after injury results in sustained attenuation of the 'detrimental' pro-inflammatory response up to 9days after injury, while not impacting the 'favorable' anti-inflammatory response. The D-NAC therapy also led to improvement in myelination, suggesting reduced white matter injury. Demonstration of treatment efficacy at later time points in the postnatal period provides a greater understanding of how microglial activation and chronic inflammation can be targeted to treat neonatal brain injury. Importantly, it may also provide a longer therapeutic window.

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### **4**Données cliniques

Brain structural and microstructural alterations associated with cerebral palsy and motor impairments in adolescents born extremely preterm and/or extremely low birthweight.

Kelly CE, Chan L, Burnett AC, Lee KJ, Connelly A, Anderson PJ, Doyle LW, Cheong JL, Thompson DK; Victorian Infant Collaborative Study Group.

*Dev Med Child Neurol. 2015 Dec;57(12):1168-75. doi: 10.1111/dmcn.12854. Epub 2015 Jul 21. Comment in Dev Med Child Neurol. 2015 Dec;57(12):1090-1.* 

AIM: To elucidate neurobiological changes underlying motor impairments in adolescents born extremely preterm (gestation <28wks) and/or with extremely low birthweight (ELBW, <1000g), our aims were the following: (1) to compare corticospinal tract (CST) microstructure and primary motor cortex (M1) volume, area, and thickness between extremely preterm/ELBW adolescents and a comparison group with normal birthweight (>2499g); (2) to compare CST microstructure and M1 volume, area, and thickness between extremely preterm/ELBW adolescents with cerebral palsy (CP), motor impairment without CP, and no motor impairment; and (3) to investigate associations between CST microstructure and M1 measures.

METHOD: This study used diffusion and structural magnetic resonance imaging to examine the CST and M1 in a geographical cohort of 191 extremely preterm/ELBW adolescents (mean age 18y 2.4mo [SD 9.6mo]; 87 males, 104 females) and 141 adolescents in the comparison group (mean age 18y 1.2mo [SD 9.6mo]; 59 males, 82 females).

RESULTS: Extremely preterm/ELBW adolescents had higher CST axial, radial, and mean diffusivities and lower M1 thickness than the comparison group. Extremely preterm/ELBW adolescents with CP had higher CST diffusivities than non-motor-impaired extremely preterm/ELBW adolescents. CST diffusivities correlated with M1 volume and area.

INTERPRETATION: Extremely preterm/ELBW adolescents have altered CST microstructure, which is associated with CP. Furthermore, the results elucidate how CST and M1 alterations interrelate to potentially influence motor function in extremely preterm/ELBW adolescents.

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© 2015 Mac Keith Press. PMID: 26195287 [PubMed - indexed for MEDLINE]

#### Does Contralesional Hand Function After Neonatal Stroke Only Depend on Lesion Characteristics?

Dinomais M, Hertz-Pannier L, Groeschel S, Delion M, Husson B, Kossorotoff M, Renaud C, Chabrier S, The Tich SN; AVCnn Study Group.

Stroke. 2016 May 10. pii: STROKEAHA.116.013545. [Epub ahead of print]

BACKGROUND AND PURPOSE: In children having suffered from neonatal arterial ischemic stroke, the relationship between contralesional hand performance and structural changes in brain areas remote from the infarct site was examined.

METHODS: Using voxel-based morphometry, we correlated contralesional gross manual dexterity assessed by the box and block test and whole-brain gray and white-matter volume changes on high-resolution magnetic resonance imaging in 37 7-year-old post-neonatal arterial ischemic stroke children. We also compared the volume of the identified structures with magnetic resonance imaging data of 10 typically developing age-matched children.

RESULTS: Areas showing the highest positive correlation with the box and block test scores were ipsilesional mediodorsal thalamus, contralesional cerebellar lobule VIIa Crus I, and ipsilesional corticospinal tract at the level of superior corona radiata, the posterior limb of the internal capsule, and the cerebral peduncle and the ipsilesional body of corpus callosum. When compared with typically developing age-matched children, post-neonatal arterial ischemic stroke children with severe contralesional hand motor deficit exhibited significant volume reductions in these structures (except the cerebellum), whereas no differences were found with those with good manual dexterity. No negative correlation was found between box and block test scores and brain areas.

CONCLUSIONS: Contralesional hand performance after neonatal arterial ischemic stroke is correlated with atrophy in brain areas directly or functionally connected but anatomically remote from the infarct. Our study suggests a role of the cerebellar lobule VIIa Crus I and mediodorsal thalamus in manual dexterity.

CLINICAL TRIAL REGISTRATION: URL: https://clinicaltrials.gov. Unique identifier: NCT02511249.

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PMID: 27165960 [PubMed - as supplied by publisher]

### Effects and Safety of Magnesium Sulfate on Neuroprotection: A Meta-analysis Based on PRISMA Guidelines.

Zeng X, Xue Y, Tian Q, Sun R, An R.

Medicine (Baltimore). 2016 Jan;95(1):e2451. doi: 10.1097/MD.0000000002451.

To evaluate the evidence of effects and safety of magnesium sulfate on neuroprotection for preterm infants who had exposure in uteri. We searched electronic databases and bibliographies of relevant papers to identify studies comparing magnesium sulfate (MgSO4) with placebo or other treatments in patients at high risk of preterm labor and reporting effects and safety of MgSO4 for antenatal infants. Then, we did this meta-analysis based on PRISMA guideline. The primary outcomes included fatal death, cerebral palsy (CP), intraventricular hemorrhage, and periventricular leukomalacia. Secondary outcomes included various neonatal and maternal outcomes. Ten studies including 6 randomized controlled trials and 5 cohort studies, and involving 18,655 preterm infants were analyzed. For the rate of moderate to severe CP, MgSO4 showed the ability to reduce the risk and achieved statistically significant difference (odd ratio [OR] 0.61, 95% confidence interval [CI] 0.42-0.89, P = 0.01). The comparison of mortality rate between the MgSO4 group and the placebo group only presented small difference clinically, but reached no statistical significance (OR 0.92, 95% CI 0.77-1.11, P = 0.39). Summarily, the analysis of adverse effects on babies showed no margin (P > 0.05). Yet for mothers, MgSO4 exhibited obvious side-effects, such as respiratory depression, nausea and so forth, but there exited great heterogeneity. MgSO4 administered to women at high risk of preterm labor could reduce the risk of moderate to severe CP, without obvious adverse effects on babies. Although there exit many unfavorable effects on mothers, yet they may be lessened through reduction of the dose of MgSO4 and could be tolerable for mothers. So MgSO4 is both beneficial and safety to be used as a neuroprotective agent for premature infants before a valid alternative was discovered.

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PMCID: PMC4706271

PMID: 26735551 [PubMed - indexed for MEDLINE]

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## Effect of Early Prophylactic High-Dose Recombinant Human Erythropoietin in Very Preterm Infants on Neurodevelopmental Outcome at 2 Years: A Randomized Clinical Trial.

Natalucci g, Latal B, Koller B, Rüegger C, Sick B, Held L, Bucher HU, Fauchère JC; Swiss EPO Neuroprotection Trial Group. Collaborators: Zeilinger G, Pasquier S, Capone A, Bührer C, Glanzmann R, Schulzke

S, Weber P, Scharrer B, Bär W, Keller E, Killer C, Pfister R, Karin K, Hüppi PS, Borradori-Tolsa C, Bucher HU, Fauchère JC, Koller B, Wellmann S, Hagmann C, Latal B, Natalucci G.

JAMA. 2016 May 17;315(19):2079-85. doi: 10.1001/jama.2016.5504.

IMPORTANCE: Very preterm infants are at risk of developing encephalopathy of prematurity and long-term neurodevelopmental delay. Erythropoietin treatment is neuroprotective in animal experimental and human clinical studies.

OBJECTIVE: To determine whether prophylactic early high-dose recombinant human erythropoietin (rhEPO) in preterm infants improves neurodevelopmental outcome at 2 years' corrected age.

DESIGN, SETTING, AND PARTICIPANTS: Preterm infants born between 26 weeks 0 days' and 31 weeks 6 days' gestation were enrolled in a randomized, double-blind, placebo-controlled, multicenter trial in Switzerland between 2005 and 2012. Neurodevelopmental assessments at age 2 years were completed in 2014.

INTERVENTIONS: Participants were randomly assigned to receive either rhEPO (3000 IU/kg) or placebo (isotonic saline, 0.9%) intravenously within 3 hours, at 12 to 18 hours, and at 36 to 42 hours after birth.

MAIN OUTCOMES AND MEASURES: Primary outcome was cognitive development assessed with the Mental Development Index (MDI; norm, 100 [SD, 15]; higher values indicate better function) of the Bayley Scales of Infant Development, second edition (BSID-II) at 2 years corrected age. The minimal clinically important difference between groups was 5 points (0.3 SD). Secondary outcomes were motor development (assessed with the Psychomotor Development Index), cerebral palsy, hearing or visual impairment, and anthropometric growth parameters.

RESULTS: Among 448 preterm infants randomized (mean gestational age, 29.0 [range, 26.0-30.9] weeks; 264 [59%] female; mean birth weight, 1210 [range, 490-2290] g), 228 were randomized to rhEPO and 220 to placebo. Neurodevelopmental outcome data were available for 365 (81%) at a mean age of 23.6 months. In an intention-to-treat analysis, mean MDI was not statistically significantly different between the rhEPO group (93.5 [SD, 16.0] [95% CI, 91.2 to 95.8]) and the placebo group (94.5 [SD, 17.8] [95% CI, 90.8 to 98.5]) (difference, -1.0 [95%

Cl, -4.5 to 2.5]; P = .56). No differences were found between groups in the secondary outcomes.

CONCLUSIONS AND RELEVANCE: Among very preterm infants who received prophylactic early high-dose rhEPO for neuroprotection, compared with infants who received placebo, there were no statistically significant differences in neurodevelopmental outcomes at 2 years. Follow-up for cognitive and physical problems that may not become evident until later in life is required.

TRIAL REGISTRATION: clinicaltrials.gov Identifier: NCT00413946.

PMID: 27187300 [PubMed - in process]

#### Fetal and Neonatal Effects of N-Acetylcysteine When Used for Neuroprotection in Maternal Chorioamnionitis.

Jenkins DD, Wiest DB, Mulvihill DM, Hlavacek AM, Majstoravich SJ, Brown TR, Taylor JJ, Buckley JR, Turner RP, Rollins LG, Bentzley JP, Hope KE, Barbour AB, Lowe DW, Martin RH, Chang EY.

J Pediatr. 2016 Jan;168:67-76.e6. doi: 10.1016/j.jpeds.2015.09.076. Epub 2015 Nov 3.

OBJECTIVE: To evaluate the clinical safety of antenatal and postnatal N-acetylcysteine (NAC) as a neuroprotective agent in maternal chorioamnionitis in a randomized, controlled, double-blinded trial.

STUDY DESIGN: Twenty-two mothers >24 weeks gestation presenting within 4 hours of diagnosis of clinical chorioamnionitis were randomized with their 24 infants to NAC or saline treatment. Antenatal NAC (100 mg/kg/dose) or saline was given intravenously every 6 hours until delivery. Postnatally, NAC (12.5-25 mg/kg/dose, n = 12) or saline (n = 12) was given every 12 hours for 5 doses. Doppler studies of fetal umbilical and fetal and infant cerebral blood flow, cranial ultrasounds, echocardiograms, cerebral oxygenation, electroencephalograms, and serum cytokines were evaluated before and after treatment, and 12, 24, and 48 hours after birth. Magnetic resonance spectroscopy and diffusion imaging were performed at term age equivalent. Development was followed for cerebral palsy or autism to 4 years of age.

RESULTS: Cardiovascular measures, cerebral blood flow velocity and vascular resistance, and cerebral oxygenation did not differ between treatment groups. Cerebrovascular coupling was disrupted in infants with chorioamnionitis treated with saline but preserved in infants treated with NAC, suggesting improved vascular regulation in the presence of neuroinflammation. Infants treated with NAC had higher serum anti-inflammatory interleukin-1 receptor Science Infos Paralysie Cérébrale, mai 2016, FONDATION MOTRICE 67 rue Vergniaud 75013 Paris - tel +33 1 18 45 54 03 03 contact: Christine Doumergue cdoumergue@lafondationmotrice.org

antagonist and lower proinflammatory vascular endothelial growth factor over time vs controls. No adverse events related to NAC administration were noted.

CONCLUSIONS: In this cohort of newborns exposed to chorioamnionitis, antenatal and postnatal NAC was safe, preserved cerebrovascular regulation, and increased an anti-inflammatory neuroprotective protein.

TRIAL REGISTRATION: ClinicalTrials.gov: NCT00724594.

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PMCID: PMC4698030 [Available on 2017-01-01] PMID: 26545726 [PubMed - indexed for MEDLINE]

Grey matter injury patterns in cerebral palsy: associations between structural involvement on MRI and clinical outcomes.

Reid SM, Dagia CD, Ditchfield MR, Reddihough DS Dev Med Child Neurol. 2015 Dec;57(12):1159-67. doi: 10.1111/dmcn.12800. Epub 2015 May 12. Comment in Dev Med Child Neurol. 2015 Dec;57(12):1089-90.

AIMS: In a population cohort of children with grey matter injury (GMI) and cerebral palsy (CP), we aimed to describe and classify magnetic resonance imaging characteristics specific to GMI, and to identify key structure-function associations that serve as a basis for rating GMI in clinically relevant ways.

METHOD: Symmetry, extent of cerebral injury, and pathological pattern for 54 children (37 males, 17 females) with CP and a predominant GMI pattern on chronic-phase magnetic resonance imaging were related to gross motor function, motor type and topography, epilepsy, intellectual disability, blindness, and deafness.

RESULTS: Relative to mild GMI where there was no pallidal abnormality, severe GMI, comprising pallidal abnormality alone or in conjunction with other deep nuclear and generalized cortical-subcortical involvement, was strongly associated with Gross Motor Function Classification System levels IV to V (OR 35.7 [95% CI 3.5, 368.8]). Involvement of the basal ganglia was associated with non-spastic/mixed motor types, but predominantly where cortical-subcortical grey and white matter involvement was not extensive. The prevalence of epilepsy was highest where there was diffuse cortical-subcortical involvement and white matter loss.

INTERPRETATION: Better understanding of structure-function relationships in CP and GMI, and how to rate the severity of GMI, will be helpful in the clinical context and also as a basis for investigation of causal pathways in CP. © 2015 Mac Keith Press.

PMID: 25970144 [PubMed - indexed for MEDLINE]

## Magnesium sulphate and perinatal mortality and morbidity in very-low-birthweight infants born between 24 and 32 weeks of gestation in Japan.

Ohhashi M, Yoshitomi T, Sumiyoshi K, Kawagoe Y, Satoh S, Sameshima H, Ikenoue T Eur J Obstet Gynecol Reprod Biol. 2016 Apr 11;201:140-145. doi: 10.1016/j.ejogrb.2016.03.048. [Epub ahead of print]

OBJECTIVE: Maternal exposure to magnesium sulphate has a neuroprotective effect in premature infants. This study aimed to examine this neuroprotective effect and the dose-response relationship in very-low-birthweight infants born between 24 and 32 weeks of gestation.

STUDY DESIGN: A retrospective cohort study compared the rates of mortality and brain damage between three groups: no magnesium sulphate, low-dose (<50g) magnesium sulphate and high-dose ( $\geq$ 50g) magnesium sulphate. RESULTS: Japanese maternal and neonatal databases were linked using six key parameters from 2003 to 2007. Of 298,514 deliveries, 9101 were very-low-birthweight infants. Among these, full matching was possible for 5562 infants. Of the fully-matched infants, 3763 were born between 24 and 32 weeks of gestation, and 1813 (48%) were followed-up beyond 18 months. A multivariate analysis of the data, including gestational age, sex, fetal growth restriction, antenatal steroids and low pH (<7.1), showed that the low-dose group had no beneficial effects in terms of a reduction in mortality or incidence of brain damage (cerebral palsy or mental retardation). The high-dose group showed a significantly higher mortality rate [odds ratio (OR) 1.9, 95% confidence interval (CI) 1.2-2.9]. A stratified subgroup analysis of infants born between 28 and 32 weeks of gestation showed that survivors in the low-dose group had significantly lower rates of cerebral palsy (OR 0.4, 95% CI 0.2-0.98) and brain damage (OR 0.2, 95% CI 0.1-0.9), while the high-dose group did not show any significant changes.

CONCLUSION: This study found that antepartum exposure to magnesium sulphate did not reduce the infant mortality rate or influence neurological outcomes. However, among infants born between 28 and 32 weeks of gestation, rates of cerebral palsy and brain damage were found to be significantly lower among survivors in the low-dose group.

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## MR angiography findings in infants with neonatal arterial ischemic stroke in the middle cerebral artery territory: A prospective study using circle of Willis MR angiography.

Husson B, Hertz-Pannier L, Adamsbaum C, Renaud C, Presles E, Dinomais M, Kossorotoff M, Landrieu P, Chabrier S Eur J Radiol. 2016 Jul;85(7):1329-35. doi: 10.1016/j.ejrad.2016.05.002. Epub 2016 May 6.

AIM: Neonatal arterial ischemic stroke (NAIS) results from a focal disruption of the blood flow in a cerebral artery by a not well understood mechanism. Our objective is to describe the acute MRangiography (MRA) findings in infants with an NAIS in the middle cerebral artery (MCA) territory and correlate them with early parenchymal infarcts and motor outcome.

METHODS: Among one hundred prospectively followed neonates with NAIS, we studied thirty-seven patients with an MCA infarct explored with circle of Willis MRA. MCA flow characteristics were documented, along with infarct location/extent and motor outcome at age 7 years.

RESULTS: Twenty-three (62%) of the children showed arterial changes, all ipsilateral to the NAIS, with occlusion in six, thrombus-type flow defect in nine, and unilateral increased flow in enlarged insular arteries in the remaining eight. There was a statistically significant correlation between parenchymal and arterial MR findings (p=0.0002). A normal MRA had a negative predictive value of 100% (95% CI: 76.8-100) in ruling out a main branch infarct. Patients with abnormal MRA tended to be at increased risk for cerebral palsy (OR=3.1). Occlusion was associated with a worse outcome (p=0.04).

INTERPRETATION: MRangiography shows arterial abnormalities suggesting that embolism is a frequent cause of NAIS.

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PMID: 27235881 [PubMed - in process]

#### Neurodevelopmental Outcomes of Extremely Preterm Infants Randomized to Stress Dose Hydrocortisone.

Parikh NA, Kennedy KA, Lasky RE, Tyson JE. PLoS One. 2015 Sep 16;10(9):e0137051. doi: 10.1371/journal.pone.0137051. eCollection 2015

OBJECTIVE: To compare the effects of stress dose hydrocortisone therapy with placebo on survival without neurodevelopmental impairments in high-risk preterm infants.

STUDY DESIGN: We recruited 64 extremely low birth weight (birth weight ≤1000 g) infants between the ages of 10 and 21 postnatal days who were ventilator-dependent and at high-risk for bronchopulmonary dysplasia. Infants were randomized to a tapering 7-day course of stress dose hydrocortisone or saline placebo. The primary outcome at follow-up was a composite of death, cognitive or language delay, cerebral palsy, severe hearing loss, or bilateral blindness at a corrected age of 18-22 months. Secondary outcomes included continued use of respiratory therapies and somatic growth.

RESULTS: Fifty-seven infants had adequate data for the primary outcome. Of the 28 infants randomized to hydrocortisone, 19 (68%) died or survived with impairment compared with 22 of the 29 infants (76%) assigned to placebo (relative risk: 0.83; 95% CI, 0.61 to 1.14). The rates of death for those in the hydrocortisone and placebo groups were 31% and 41%, respectively (P = 0.42). Randomization to hydrocortisone also did not significantly affect the frequency of supplemental oxygen use, positive airway pressure support, or need for respiratory medications.

CONCLUSIONS: In high-risk extremely low birth weight infants, stress dose hydrocortisone therapy after 10 days of age had no statistically significant effect on the incidence of death or neurodevelopmental impairment at 18-22

months. These results may inform the design and conduct of future clinical trials. TRIAL REGISTRATION: ClinicalTrials.gov NCT00167544.

Free PMC Article

PMCID: PMC4573756

PMID: 26376074 [PubMed - indexed for MEDLINE]

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#### Proximity of magnesium exposure to delivery and neonatal outcomes.

Turitz AL, Too GT, Gyamfi-Bannerman C

Am J Obstet Gynecol. 2016 May 10. pii: S0002-9378(16)30199-5. doi: 10.1016/j.ajog.2016.05.004. [Epub ahead of print]

BACKGROUND: In infants delivered preterm, magnesium sulfate reduces cerebral palsy in survivors. The benefit of magnesium given remote from delivery is unclear.

OBJECTIVE: Our objective is to evaluate the association of time from last exposure to magnesium with cerebral palsy. STUDY DESIGN: Secondary analysis of a multicenter trial evaluating magnesium for neuroprotection. For this study, we included women with live, non-anomalous, singleton gestations who received magnesium. Pregnancies with missing information at 2-year follow-up were excluded. Women were divided into 2 groups based on exposure timing: last infusion of magnesium <12 hours and last infusion of magnesium  $\geq$ 12 hours prior to delivery. The primary outcome was cerebral palsy of any severity at 2 years of life. Secondary outcomes were moderate/severe cerebral palsy and moderate/severe cerebral palsy or death. Chi-squared, Student's t-test, and Mann Whitney U were used for bivariate associations. We fit a multivariable logistic regression model to adjust for confounders.

RESULTS: 906 infants were analyzed. 568 were last exposed to magnesium <12 hours prior to delivery and 338 were last exposed  $\geq$ 12 hours. Cerebral palsy occurred in 28 offspring (3%), 2.3% of those last exposed <12 hours vs. 4.4% last exposed  $\geq$ 12 hours prior to delivery, p=0.07. On adjusted analyses, last exposure to magnesium <12 hours prior to delivery was associated with a significant reduction in cerebral palsy compared to last exposure  $\geq$ 12 hours (adjusted odds ratio 0.41, 95% confidence interval 0.18-0.91, p=0.03). There was no difference in secondary outcomes.

CONCLUSION: Exposure to magnesium proximal to delivery (<12 hours) is associated with reduced odds of cerebral palsy compared to more remote exposure. This highlights the importance of timing of magnesium for neuroprotection for women at risk of preterm delivery.

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PMID: 27177525 [PubMed - as supplied by publisher]

## The clinical outcomes of deep gray matter injury in children with cerebral palsy in relation with brain magnetic resonance imaging.

Choi JY, Choi YS, Rha D, Park ES.

Res Dev Disabil. 2016 May 20;55:218-225. doi: 10.1016/j.ridd.2016.04.010. [Epub ahead of print]

In the present study we investigated the nature and extent of clinical outcomes using various classifications and analyzed the relationship between brain magnetic resonance imaging (MRI) findings and the extent of clinical outcomes in children with cerebral palsy (CP) with deep gray matter injury. The deep gray matter injuries of 69 children were classified into hypoxic ischemic encephalopathy (HIE) and kernicterus patterns. HIE patterns were divided into four groups (I-IV) based on severity. Functional classification was investigated using the gross motor function classification system-expanded and revised, manual ability classification system, communication function classification system, and tests of cognitive function, and other associated problems. The severity of HIE pattern on brain MRI was strongly correlated with the severity of clinical outcomes in these various domains. Children with a kernicterus pattern showed a wide range of clinical outcomes in these areas. Children with severe HIE are at

high risk of intellectual disability (ID) or epilepsy and children with a kernicterus pattern are at risk of hearing impairment and/or ID. Grading severity of HIE pattern on brain MRI is useful for predicting overall outcomes. The

clinical outcomes of children with a kernicterus pattern range widely from mild to severe.WHAT THIS PAPER ADDS: Delineation of the clinical outcomes of children with deep gray matter injury, which are a common abnormal brain MRI finding in children with CP, is necessary. The present study provides clinical outcomes for various domains in children with deep gray matter injury on brain MRI. The deep gray matter injuries were divided into two major groups; HIE and kernicterus patterns. Our study showed that severity of HIE pattern on brain MRI was strongly

associated with the severity of impairments in gross motor function, manual ability, communication function, and cognition. These findings suggest that severity of HIE pattern can be useful for predicting the severity of impairments. Conversely, children with a kernicterus pattern showed a wide range of clinical outcomes in various domains. Children with severe HIE pattern are at high risk of ID or epilepsy and children with kernicterus pattern are at risk of hearing impairment or ID. The strength of our study was the assessment of clinical outcomes after 3 years of age using standardized classification systems in various domains in children with deep gray matter injury. Copyright © 2016 Elsevier Ltd. All rights reserved.

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## The ProVIDe study: the impact of protein intravenous nutrition on development in extremely low birthweight babies.

Bloomfield FH, Crowther CA, Harding JE, Conlon CA, Jiang Y, Cormack BE. BMC Pediatr. 2015 Aug 26;15:100. doi: 10.1186/s12887-015-0411-y.

BACKGROUND: Preterm birth and very small size at birth have long-term effects on neurodevelopment and growth. A relatively small percentage of extremely low birthweight babies suffer from severe neurological disability; however, up to 50% experience some neurodevelopmental or learning disability in childhood. Current

international consensus is that increased protein intake in the neonatal period improves both neurodevelopment and growth, but the quantum of protein required is not known. This trial aims to assess whether providing an extra 1 to 2 g.kg(-1).d(-1) protein in the first 5 days after birth will improve neurodevelopmental outcomes and growth in extremely low birthweight babies.

METHODS/DESIGN: The ProVIDe study is a multicentre, two-arm, double-blind, parallel, randomised, controlled trial. In addition to standard intravenous nutrition, 430 babies with a birthweight of less than 1000 g who have an

umbilical arterial line in situ will be randomised in 1:1 ratio to receive either an amino acid solution (TrophAmine<sup>®</sup>) or placebo (saline) administered through the umbilical arterial catheter for the first 5 days. Exclusion criteria are

admission to neonatal intensive care more than 24 h after birth; multiple births of more than 2 babies; known chromosomal or genetic abnormality, or congenital disorder affecting growth; inborn error of metabolism, and in danger of imminent death.

PRIMARY OUTCOME: Survival free from neurodevelopmental disability at 2 years' corrected age, where neurodevelopmental disability is defined as cerebral palsy, blindness, deafness, developmental delay (standardised score more than 1 SD below the mean on the cognitive, language or motor subscales of the Bayley Scales of Infant Development Edition 3), or Gross Motor Function Classification System score  $\geq 1$ .

SECONDARY OUTCOMES: Growth, from birth to 36 weeks' corrected gestational age, at neonatal intensive care discharge and at 2 years' corrected age; body composition at 36 to 42 weeks' corrected postmenstrual age and at 2 years' corrected age; neonatal morbidity, including length of stay; nutritional intake.

DISCUSSION: This trial will provide the first direct evidence of the effects of giving preterm babies a higher intake of intravenous protein in the first week after birth on neurodevelopmental outcomes at 2 years corrected age.

TRIAL REGISTRATION: Australian New Zealand Clinical Trials Registry: ACTRN12612001084875.

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PMCID: PMC4549896 PMID: 26307566 [PubMed - indexed for MEDLINE]

### Motricite – Mobilité - Posture

## Muscle strength does not explain standing ability in children with bilateral spastic cerebral palsy: a cross sectional descriptive study.

Lidbeck C, Tedroff K, Bartonek Å.

BMC Neurol. 2015 Oct 8;15:188. doi: 10.1186/s12883-015-0441-y.

BACKGROUND: In bilateral cerebral palsy (CP) muscle strength is considered important for development of gross motor functions, but its influence on standing ability has not been explored. Our aims were to examine muscle strength with respect to the ability to stand with (SwS) or without (SwoS) hand support, asymmetrical weight bearing (WB), and whether the ability to produce strength was influenced by different seated conditions.

METHODS: In this cross sectional descriptive study standing posture was recorded with 3D motion analysis, and muscle strength was measured with a hand-held dynamometer, in 25 children with bilateral CP, GMFCS levels II-III, SwS (n = 14, median age 11.4 years), or SwoS, (n = 11, median age 11.4 years). Strength measurements were taken in the hip flexors, knee extensors, dorsiflexors and plantarflexors, in two seated conditions; a chair with arm- and backrests, and a stool.

RESULTS: Compared to SwoS, children SwS stood with a more flexed posture, but presented with equal strength in the hip flexors, dorsiflexors and plantarflexors, and with somewhat more strength in the knee extensors. Despite

asymmetric WB during standing, both limbs were equally strong in the two groups. No differences in strength were measured between the two seated conditions.

CONCLUSIONS: Despite challenges measuring muscle strength in CP, the lower limb muscle strength cannot be considered an explanatory factor for variations in standing in this group of children with bilateral CP. The findings rather strengthen our hypothesis that deficits in the sensory systems could be as determinant for standing as muscle weakness in children with bilateral spastic

Free PMC Article PMCID: PMC4599329

PMID: 26449859 [PubMed - indexed for MEDLINE]

#### Muscle synergies and complexity of neuromuscular control during gait in cerebral palsy.

Steele KM Rozumalski A, Schwartz MH.

Dev Med Child Neurol. 2015 Dec;57(12):1176-82. doi: 10.1111/dmcn.12826. Epub 2015 Jun 17. Comment in Dev Med Child Neurol. 2015 Dec;57(12):1091-2.

AIM: Individuals with cerebral palsy (CP) have impaired movement due to a brain injury near birth. Understanding how neuromuscular control is altered in CP can provide insight into pathological movement. We sought to determine if individuals with CP demonstrate reduced complexity of neuromuscular control during gait compared with unimpaired individuals and if changes in control are related to functional ability.

METHOD: Muscle synergies during gait were retrospectively analyzed for 633 individuals (age range 3.9-70y): 549 with CP (hemiplegia, n=122; diplegia, n=266; triplegia, n=73; quadriplegia, n=88) and 84 unimpaired individuals. Synergies were calculated using non-negative matrix factorization from surface electromyography collected during previous clinical gait analyses. Synergy complexity during gait was compared with diagnosis subtype, functional ability, and clinical examination measures.

RESULT: Fewer synergies were required to describe muscle activity during gait in individuals with CP compared with unimpaired individuals. Changes in synergies were related to functional impairment and clinical examination measures including selective motor control, strength, and spasticity.

INTERPRETATION: Individuals with CP use a simplified control strategy during gait compared with unimpaired individuals. These results were similar to synergies during walking among adult stroke survivors, suggesting similar neuromuscular control strategies between these clinical populations.

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PMCID: PMC4683117 [Available on 2016-12-01] PMID: 26084733 [PubMed - indexed for MEDLINE

#### Plantar flexor muscle weakness may cause stiff-knee gait.

Apti A, Akalan NE, Kuchimov S, Özdinçler AR, Temelli Y, Nene A. Gait Posture. 2016 May;46:201-7. doi: 10.1016/j.gaitpost.2016.03.010. Epub 2016 Mar 28.

AIM: The iterative simulation studies proclaim that plantar flexor (PF) muscle weakness is one of the contributors of stiff knee gait (SKG), although, whether isolated PF weakness generates SKG has not been validated in able-bodied people or individuals with neuromuscular disorders. The aim of the study was to investigate the effects of isolated PF muscle weakness on knee flexion velocity and SKG in healthy individuals.

METHOD: Twenty able-bodied young adults (23±3 years) participated in this study. Passive stretch (PS) protocol was applied until the PF muscle strength dropped 33.1% according to the hand-held dynamometric measurement. Seven additional age-matched able-bodies were compared with participants' to discriminate the influence of slow-walking. All participants underwent 3D gait analysis before and after the PS. Peak knee flexion angle, range of knee flexion between toe-off and peak knee flexion, total range of knee-flexion, and time of peak knee flexion in swing were selected to describe SKG pattern.

RESULTS: After PS, the reduction of plantar flexor muscle strength (33.14%) caused knee flexion velocity drop at toeoff (p=0.008) and developed SKG pattern by decreasing peak knee flexion (p=0.0001), range of knee flexion in early swing (p=0.006), and total knee flexion range (p=0.002). These parameters were significantly correlated with decreased PF velocity at toe-off (p=0.015, p=0.0001, p=0.005, respectively). The time of peak knee flexion was not significantly different between before and after stretch conditions (p=0.130).

CONCLUSIONS: These findings verified that plantar flexor weakness cause SKG pattern by completing three of SKG parameters. Any treatment protocol that weakens the plantar flexor muscle might impact the SKG pattern.

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#### Proximal Femoral Growth Modification: Effect of Screw, Plate, and Drill on Asymmetric Growth of the Hip.

d'Heurle A, McCarthy J, Klimaski D, Stringer K. J Pediatr Orthop. 2016 May 19. [Epub ahead of print]

BACKGROUND: Guided growth has long been used in the lower extremities but has not been applied to varus or valgus deformity in the hip, as may occur in children with cerebral palsy or developmental dysplasia of the hip. The purpose of this study was to determine if screw, plate, or drilling techniques decreased the femoral neck-shaft angle (NSA) and articular trochanteric disease (ATD), as well as describe growth plate structural changes with each method.

METHODS: Twelve 8-week-old lambs underwent proximal femoral hemiepiphysiodesis (IACUC approved) using either a screw (n=4), plate (n=4), or drilling procedure (n=4). Postoperative time was 6 months. Radiographs taken after limb harvest were used to measure NSA and ATD. Differences between treated and control sides were determined by 1-tailed paired t tests and Bonferroni ( $\alpha$ =0.05/3). Histology was obtained for 1 limb pair per group. Proximal femurs were cut in midcoronal plane and the longitudinal growth plates were examined for structural changes.

RESULTS: The mean NSA measured 7 degrees less than controls in this model using the screw technique, and this difference was statistically significant. Differences between the control and the treated groups did not reach statistical significance for either the plate or the drill group. Differences in ATD were not statistically significant, although there was a trend for larger ATD measurements using the screw technique. Histologically, physeal changes were observed on the operative sides in screw and plate specimens, but not drill specimens, compared with contralateral sham control. The screw specimen exhibited the most severe changes, with growth plate closure over half the section. The plate specimen showed focal loss of the physis across the section, but with no evidence of closure.

CONCLUSIONS: This study builds on previous work that indicates screw hemiepiphysiodesis can effectively alter the shape of the proximal femur, and result in a lower neck-shaft ankle (or lesser valgus). This study suggests that

implantation of a screw is likely to be more effective than a plate or drilling procedure in decreasing the NSA in skeletally immature hips.

CLINICAL SIGNIFICANCE: If further preclinical, and later clinical, studies demonstrate reproducible efficacy, guided growth of the proximal femur may eventually become a viable option for treatment or prevention of hip deformity in select patients.

PMID: 27203823 [PubMed - as supplied by publisher]

#### Relationship between stature and tibial length for children with moderate-to-severe cerebral palsy.

Kihara K, Kawasaki Y, Yagi M, Takada S. Brain Dev. 2015 Oct;37(9):853-7. doi: 10.1016/j.braindev.2015.01.007. Epub 2015 Mar 11.

OBJECTIVE: To derive the equation for estimating stature, based on tibial length, for children with moderate-tosevere cerebral palsy (CP) and lower limb joint contracture or scoliosis.

METHODS: The participants (3-12-years-old) included 50 children with moderate-to-severe CP (mean age, 8.3±2.4 years) and 38 typically developed (TD) children (mean age, 7.5±2.6 years). Thirty-four (68%) of the children with CP had a gross motor function classification system level of V. Furthermore, 40 (80%) had definite lower limb joint contracture or scoliosis. The stature and the tibial length measurements of all participants were determined. Regression equations to estimate stature, based on tibial lengths, were determined for both TD children and children with CP. Moreover, regression equations defining the relationship between tibial length and age were compared between the two groups of children, using multiple regression analysis.

RESULTS: The regression equations for estimating stature, based on tibial length, were stature=tibial length×3.25+34.45 [cm], R(2)=0.91 (TD children), and stature=tibial length×3.42+31.82 [cm], R(2)=0.81 (CP children). In children with CP, tibial lengths were significantly shorter than those in similarly aged TD children.

CONCLUSION: The stature of children with moderate-to-severe CP can be estimated from their tibial lengths, regardless of the presence of joint contracture or scoliosis. The tibial length may be a proxy for estimating stature during the growth assessment of children with moderate-to-severe CP.

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## The mechanics of activated semitendinosus are not representative of the pathological knee joint condition of children with cerebral palsy.

Ateş F, Temelli Y, Yucesoy CA. J Electromyogr Kinesiol. 2016 Apr 20;28:130-136. doi: 10.1016/j.jelekin.2016.04.002. [Epub ahead of print]

Characteristic cerebral palsy effects in the knee include a restricted joint range of motion and forcefully kept joint in a flexed position. To show whether the mechanics of activated spastic semitendinosus muscle are contributing to these effects, we tested the hypothesis that the muscle's joint range of force exertion is narrow and force production capacity in flexed positions is high. The isometric semitendinosus forces of children with cerebral palsy (n=7, mean (SD)=7years (8months), GMFCS levels III-IV, 12 limbs tested) were measured intra-operatively as a function of knee angle, from flexion (120°) to full extension (0°). Peak force measured in the most flexed position was considered as the benchmark. However, peak force (mean (SD)=112.4N (54.3N)) was measured either at intermediate or even full knee extension (three limbs) indicating no narrow joint range of force exertion. Lack of high force production capacity in flexed knee positions (e.g., at 120° negligible or below 22% of the peak force) was shown except for one limb. Therefore, our hypothesis was rejected for a vast majority of the limbs. These findings and those reported for spastic gracilis agree, indicating that the patients' pathological joint condition must rely on a more complex mechanism than the mechanics of individual spastic muscles.

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PMID: 27128957 [PubMed - as supplied by publisher]

#### The impact of walking devices on kinematics in patients with spastic bilateral cerebral palsy.

Krautwurst BK, Dreher T, Wolf SI.

Gait Posture. 2016 May;46:184-7. doi: 10.1016/j.gaitpost.2016.03.014. Epub 2016 Mar 26.

eased anterior pelvic and trunk tilt is a common finding in patients with bilateral cerebral palsy especially during walking with assistive devices. As previous studies demonstrate various gait alterations when using assistive devices, the assessment of surgical interventions may be biased when the patients become independent of (or dependent on) assistive devices after therapy. Furthermore, some of these patients in fact are able to walk without devices even though in daily life they prefer to use them. Consequently, for such patients the classification into GMFCS level II or III may be ambiguous. The specific aim of this study was therefore to assess the influence of the use of forearm crutches and posterior walker during walking and to set this influence in relation to outcome effects of surgical intervention studies. 26 ambulatory patients with spastic bilateral CP (GMFCS II-III) were included who underwent 3D gait analysis. All patients used forearm crutches or posterior walkers in everyday life even though they were able to walk without assistive devices for short distances. Independent of the type of assistive devices, the patients walk on average with more anterior trunk tilt and pelvic tilt (7°±6° and 3°±2°) and with a maximum ankle dorsiflexion decreased by 2° (±3°) when walking with assistive devices, enhancing the mal-positioning present without device. Oppositely, the knees on average are more extended by 6° (±4°) when using the assistive devices. These effects have to be taken into account when assessing gait patterns or when monitoring the outcome after intervention as assistive devices may partially hide or exaggerate therapeutic effects.

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PMID: 27131199 [PubMed - in process]

Validation of an activity monitor for children who are partly or completely wheelchair-dependent.

Nooijen CF(, de Groot JF, Stam HJ, van den Berg-Emons RJ, Bussmann HB(5); Fit for the Future Consortium. J Neuroeng Rehabil. 2015 Feb 6;12:11. doi: 10.1186/s12984-015-0004-x.

BACKGROUND: Children who are wheelchair-dependent are at risk for developing unfavorable physical behavior; therefore, assessment, monitoring and efforts to improve physical behavior should start early in life. VitaMove is an accelerometer-based activity monitor and can be used to detect and distinguish different categories of physical behavior, including activities performed in a wheelchair and activities using the legs. The purpose of this study was to assess the validity of the VitaMove activity monitor to quantify physical behavior in children who are partly or completely wheelchair-dependent.

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METHODS: Twelve children with spina bifida (SB) or cerebral palsy (CP) (mean age,  $14 \pm 4$  years) performed a series of wheelchair activities (wheelchair protocol) and, if possible, activities using their legs (n = 5, leg protocol). Activities were performed at their own home or school. In children who were completely wheelchair-dependent, VitaMove monitoring consisted of one accelerometer-based recorder attached to the sternum and one to each wrist. For children who were partly ambulatory, an additional recorder was attached to each thigh. Using video-recordings as a reference, primary the total duration of active behavior, including wheeled activity and leg activity, and secondary agreement, sensitivity and specificity scores were determined.

RESULTS: Detection of active behaviour with the VitaMove activity monitor showed absolute percentage errors of 6% for the wheelchair protocol and 10% for the leg protocol. For the wheelchair protocol, the mean agreement was 84%, sensitivity was 80% and specificity was 85%. For the leg protocol, the mean agreement was 83%, sensitivity was 78% and specificity was 90%. Validity scores were lower in severely affected children with CP.

CONCLUSIONS: The VitaMove activity monitor is a valid device to quantify physical behavior in children who are partly or completely wheelchair-dependent, except for severely affected children and for bicycling.

#### Free PMC Article

PMCID: PMC4339429 PMID: 25656614 [PubMed - indexed for MEDLINE]

### Traitement - Rééducation motrice et cognitive

#### Pharmacologie Efficacite Tolérance

#### Clinical Trial of Erythropoietin in Young Children With Cerebral Palsy.

Cho KH, Min K, Lee SH, Lee S, An SA, Kim M. J Child Neurol. 2016 May 27. pii: 0883073816650038. [Epub ahead of print]

This study was conducted to assess the safety and efficacy of recombinant human erythropoietin in young children with cerebral palsy aged between 6 months and 3 years. All participants received subcutaneous recombinant human erythropoietin and 8 weeks of rehabilitation therapy. Adverse events, changes of vital signs, and hematologic tests were monitored up to 8 weeks postinjection. Functional measures of development at 4 and 8 weeks postinjection were compared with baseline values, and improvements were compared with those of an age-matched historical control group. Nine participants completed the trial from June 2012 to February 2015. No adverse events were related to recombinant human erythropoietin. Erythropoiesis was noted, although within normal range. Functional improvements were observed in all participants (P < .05) and increases in motor function were higher in recombinant human erythropoietin group than the control group. Accordingly, recombinant human erythropoietin administration was safe without any significant adverse events and improved the functional outcomes in young children with cerebral palsy.

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PMID: 27233796 [PubMed - as supplied by publisher]

#### Management of drooling in children with cerebral palsy: A French survey.

Chaléat-Valayer E, Porte M, Buchet-Poyau K, Roumenoff-Turcant F, D'Anjou MC, Boulay C, Bernard JC, Touzet S. *Eur J Paediatr Neurol. 2016 Apr 22. pii: S1090-3798(16)30014-9. doi: 10.1016/j.ejpn.2016.04.010. [Epub ahead of print]* 

AIM: To characterise children with cerebral palsy (CP) and pathological drooling in France, and to describe care pathways, assessment and treatment.

METHOD: A transversal, observational, descriptive survey of the practices and opinions of 400 health professionals potentially involved in the care of children with CP, was carried out nationally across France in 2013.

RESULTS: The response rate was 36%. Seventy-five questionnaires were returned and analysed (52%). A small proportion of children were specifically treated for drooling (<25%). Assessments were carried out in 75% of cases and 91% of professionals prescribed treatments. Use of assessment tools varied widely. The most common treatment was oro-facial rehabilitation (95% of professionals), followed by anticholinergic drugs (Scopolamine(<sup>®</sup>)) (94%) botulinum toxin injections (BT) (66%) and surgery (34%). Scopolamine was considered to be less effective than BT and to have more side effects.

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CONCLUSION: The rate of pathological drooling in children with CP is likely underestimated and under treated in France. There is a lack of knowledge regarding assessment tools. Aside from rehabilitation, current practice is to prescribe medication as the first-line treatment, however professionals consider that BT is more effective and has less side effects.

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Intrathecal baclofen pumps do not accelerate progression of scoliosis in quadriplegic spastic cerebral palsy.

Rushton PR, Nasto LA, Aujla RK, Ammar A Grevitt MP, Vloeberghs MH. *Eur Spine J. 2016 May 6. [Epub ahead of print]* 

PURPOSE: To compare scoliosis progression in quadriplegic spastic cerebral palsy with and without intrathecal baclofen (ITB) pumps.

METHODS: A retrospective matched cohort study was conducted. Patients with quadriplegic spastic cerebral palsy, GMFCS level 5, treated with ITB pumps with follow-up >1 year were matched to comparable cases by age and baseline Cobb angle without ITB pumps. Annual and peak coronal curve progression, pelvic obliquity progression and need for spinal fusion were compared.

RESULTS: ITB group: 25 patients (9 female), mean age at pump insertion 9.4 and Risser 0.9. Initial Cobb angle 25.6° and pelvic tilt 3.2°. Follow-up 4.3 (1.0-7.8) years. Cobb angle at follow-up 76.1° and pelvic tilt 18.9°. Non-ITB group: 25 patients (14 female), mean age at baseline 9.2 and Risser 1.0. Initial Cobb angle 29.7° and pelvic tilt 7.1°. Follow-up 3.5 (1.0-7.5) years. Cobb angle at follow-up 69.1° and pelvic tilt 21.0°. The two groups were statistically similar for baseline age, Cobb angle and Risser grade. Mean curve progression was 13.6°/year for the ITB group vs 12.6°/year for the non-ITB group (p = 0.39). Peak curve progression was similar between the groups. Pelvic tilt progression was comparable; ITB group 4.5°/year vs non-ITB 4.6°/year (p = 0.97). During follow-up 5 patients in the ITB group and 9 in the non-ITB group required spinal fusion surgery for curve progression (p = 0.35).

CONCLUSIONS: Patients with quadriplegic spastic cerebral palsy with and without ITB pumps showed significant curve progression over time. ITB pumps do not appear to alter the natural history of curve progression in this population.

PMID: 27154169 [PubMed - as supplied by publisher]

#### Intrathecal Baclofen Therapy in a Child With Severe Scoliosis: Report of 2 Cases.

Sasaki N, Ogiwara H.

Neuromodulation. 2016 May 18. doi: 10.1111/ner.12449. [Epub ahead of print]

OBJECTIVE: Scoliosis is commonly found in children with cerebral palsy. Many patients with cerebral palsy and scoliosis undergo intrathecal baclofen (ITB) pump placement. The authors report 2 cases with cerebral palsy and severe scoliosis treated with intrathecal baclofen.

METHOD: The case of a 7-year-old boy with shunted hydrocephalus required surgical revision of the intrathecal catheter, while the other patient without shunt did not require revision. In the patient with shunted hydrocephalus, after the initial placement of baclofen pump and catheter at Th3 level, spasticity of lower extremities did not improve. The Indium(111) diethylenetriamine pentaacetic acid (In(111) DTPA) scintigraphy with injection of In(111) DTPA through the pump did not demonstrate distribution of the tracer to the lumbosacral area. Conversely, by direct injection of In(111) DTPA through lumbar puncture, the tracer distributed in the whole spinal canal.

RESULT: Replacement of the tip of the catheter caudal to the curve of the scoliosis improved the symptom.

CONCLUSION: The authors suggest that, in patients with severe scoliosis and shunted hydrocephalus, it may be necessary to place the tip of the catheter caudal to the curve of the scoliosis for correction of spasticity of lower extremities.

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PMID: 27191796 [PubMed - as supplied by publisher]

#### Muscle histopathology in children with spastic cerebral palsy receiving botulinum toxin type A.

Valentine J, Stannage K, Fabian V, Ellis K, Reid S, Pitcher C, Elliott C. Muscle Nerve. 2016 Mar;53(3):407-14. doi: 10.1002/mus.24763. Epub 2016 Jan 5.

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INTRODUCTION: Botulinum toxin A (BoNTA) is routine treatment for hypertonicity in children with cerebral palsy (CP).

METHODS: This single-blind, prospective, cross-sectional study of 10 participants (mean age 11 years 7 months) was done to determine the relationship between muscle histopathology and BoNTA in treated medial gastrocnemius muscle of children with CP. Open muscle biopsies were taken from medial gastrocnemius muscle and vastus lateralis (control) during orthopedic surgery.

RESULTS: Neurogenic atrophy in the medial gastrocnemius was seen in 6 participants between 4 months and 3 years post-BoNTA. Type 1 fiber loss with type 2 fiber predominance was significantly related to the number of BoNTA injections (r = 0.89, P < 0.001).

CONCLUSIONS: The impact of these changes in muscle morphology on muscle function in CP is not clear. It is important to consider rotating muscle selection or injection sites within the muscle or allowing longer time between injections.

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PMID: 26154631 [PubMed - indexed for MEDLINE]

Safety profile of incobotulinum toxin A [Xeomin(<sup>®</sup>)] in gastrocnemious muscles injections in children with cerebral palsy: Randomized double-blind clinical trial.

Carraro E, Trevisi E, Martinuzzi A.

Eur J Paediatr Neurol. 2016 Apr 22. pii: S1090-3798(16)30012-5. doi: 10.1016/j.ejpn.2016.04.008. [Epub ahead of print]

BACKGROUND: The only two preparations of botulinum toxin A for which there are published evidences of efficacy in children with cerebral palsy are onabotulinum toxin A (Botox(<sup>®</sup>)) and abobotulinum toxin A (Dyport(<sup>®</sup>)); these toxins should be considered generally safe and appropriate in the treatment for localized upper and lower limb spacticity.

and lower limb spasticity.

AIMS: To establish the safety profile of incobotulinum toxin A (Xeomin(<sup>®</sup>)) in children with cerebral palsy and muscle spasticity.

METHODS: Randomized double-blind controlled trial that involved the recruitment of children of both sexes with spastic hemiplegia or diplegia in cerebral palsy, aged between 3 and 18 years. Children were randomized to either the study group (SG, incobotulinum toxin A) or the control group (CG, onabotulinum toxin A) both to be injected with 5units/kg on gastrocnemius (medialis and lateralis) muscles. The occurrence of adverse events at baseline, after 48 h, 10 days and 3 months was recorded by the caregivers in a checklist that listed both common and uncommon side effects.

RESULTS: 35 patients were treated (CG = 18; SG = 17); the 2 groups were well balanced regarding demographics and anthropometry characteristics. At least 1 adverse event occurred in 49% of patients within first 2 days, 46% between 2 and 10 days, and 12% between 10 and 90 days. All the reported events were minor; no serious adverse event was recorded. Fatigue was the most frequent complaint. There was no significant difference in frequency and type of events between the 2 groups.

CONCLUSION: Incobotulinum toxin A and onabotulinum toxin A share similar profile of safety in the treatment of lower limb spasticity in CP children.

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#### Treatment with botulinum toxin in children with cerebral palsy: a qualitative study of parents' experiences.

Lorin K, Forsberg A.

Child Care Health Dev. 2016 May 19. doi: 10.1111/cch.12350. [Epub ahead of print]

BACKGROUND: In children with cerebral palsy everyday movements such as walking, standing and using one's hands can be difficult to perform because of spasticity. Botulinum neurotoxin type A (BoNT-A) are often used to reduce spasticity. The aim of this study was to describe how parents of children with cerebral palsy experienced the child's treatment with BoNT-A, how the child was affected by the treatment and how spasticity affected the child.

METHODS: A qualitative study in which 15 parents of children (6-13 years old) with cerebral palsy were interviewed about their experiences of the BoNT-A treatment. The children had received several BoNT-A treatments. An

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interview guide was used with topics: the child's functions before and after the treatment, the outcomes of the treatment and how they valued the BoNT-A treatment. Content analysis was used to analyse the interviews.

RESULTS: The analyses resulted in two themes: 'When softness comes and goes' and 'Both want and do not want'. The reduction of spasticity - softness – was described to promote motor functions, and facilitate the next step in motor development. The children were described as being more active out of their own initiative and having a happier mood. Spasticity, described as stiffness, was described to make walking more strenuous as well as interfering with activities. The BoNT-A injection procedure was perceived as troublesome and painful for the child, and sometimes traumatic for both children and parents.

CONCLUSIONS: Treatment with BoNT-A was described as facilitating motor development and activity. The children's and the parents' negative experiences of the injection procedure should be addressed.

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#### <u>Chirurgie</u>

Factors Contributing to Satisfaction with Changes in Physical Function after Orthopedic Surgery for Musculoskeletal Dysfunction in Patients with Cerebral Palsy.

Kusumoto Y, Nitta O, Matsuo A, Takaki K, Matsuda T.

PLoS One. 2016 May 2;11(5):e0154749. doi: 10.1371/journal.pone.0154749. eCollection 2016.

BACKGROUND: The recognition of required treatments for cerebral palsy (CP) patients, including orthopedic surgery, differs according to region. This study was performed to identify factors associated with satisfactory changes in physical function after orthopedic surgery.

METHODS: 358 patients were selected for the questionnaire survey. The following information was collected: gender, primary disease, age of initial surgery, total procedural count, operated sites, satisfaction of postoperative rehabilitation frequency, ideal amount of postoperative rehabilitation sessions per week, frequency of voluntary home training per week, satisfaction of the timing of surgery and the current satisfaction with the changes in physical function after the orthopedic surgery. We classified the patients into the satisfied and dissatisfied group according to satisfactory changes in physical function after the surgery. We performed unpaired t-tests and chi-square tests to determine the variables that differed significantly between the groups. Variables with a p value of <0.2 were included in the multivariate logistic regression analysis.

RESULTS: The logistic model was revised and summed up to two potential predictors of postsurgical satisfaction with physical function: satisfaction with the frequency of postoperative rehabilitation sessions and the orthopedic surgery of the hip (distinction hit ratio, 75.4%).

CONCLUSIONS: This study demonstrated that the frequency of postoperative rehabilitation and history of hip surgery seemed to be related to the satisfaction with the changes in physical function after orthopedic surgery.

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PMCID: PMC4852902 PMID: 27135609 [PubMed - in process]

## Long-term outcomes after multilevel surgery including rectus femoris, hamstring and gastrocnemius procedures in children with cerebral palsy.

Õunpuu S, Solomito M, Bell K, DeLuca P, Pierz K. Gait Posture. 2015 Sep;42(3):365-72. doi: 10.1016/j.gaitpost.2015.07.003. Epub 2015 Jul 23.

BACKGROUND/AIMS: Multilevel surgical intervention is a common approach for the correction of gait abnormalities in children with cerebral palsy (CP). The short-term outcomes for the combination of rectus femoris transfer, hamstring lengthening and gastrocnemius lengthening have been well documented using three-dimensional motion analysis. However, the impact of time, growth, and puberty on these short-term outcomes of this combination of procedures is not well understood. The purpose of this study was to evaluate the long-term outcomes of these procedures on gait in patients with CP.

METHODS: Twenty-two patients underwent rectus femoris transfers, medial hamstring lengthenings and gastrocnemius lengthenings in combination with a selection of other soft tissue and/or bony procedures of the lower

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limb. All patients had a pre-operative motion analysis and post-operative analysis one and 11 years following surgery.

RESULTS: Significant changes in both clinical and gait variables from pre to 1 year post surgery confirmed the shortterm gait benefits of this combination of surgical procedures. Long-term follow-up data indicated that the passive range of motion gains noted 1 year after surgery were lost at the knee and ankle. However, the improvements in ankle dorsiflexion and knee extension at initial contact were maintained over 11 years. As well, peak ankle dorsiflexion in stance was maintained and peak ankle plantar flexor moments and powers did not show declines long-term. Peak knee flexion showed a decline over the long-term, however, the timing of peak knee flexion in swing was maintained.

CONCLUSION: When compared to declines in gait kinematics in persons with CP without surgery, these results demonstrate the possible long-term benefits of surgical intervention.

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PMID: 26260009 [PubMed - indexed for MEDLINE]

#### Long-term Outcomes of Triple Arthrodesis in Cerebral Palsy Patients.

Trehan SK, Ihekweazu UN, Root L. J Pediatr Orthop. 2015 Oct-Nov;35(7):751-5. doi: 10.1097/BPO.0000000000000361.

BACKGROUND: Triple arthrodesis in the appropriately indicated cerebral palsy patient with a painful and/or rigid foot deformity can significantly alleviate pain and improve function. Limited data on long-term outcomes of triple arthrodesis in this patient population exist. In addition, there have been concerns about the long-term consequences of altered biomechanics in these patients on the tibiotalar (ankle) joint.

METHODS: We retrospectively reviewed 21 cerebral palsy patients who had undergone triple arthrodesis for a painful and/or rigid foot deformity at our institution with at least 10 years of clinical or radiographic follow-up. Preoperative, and the most recent, clinical evaluations and radiographs were reviewed. In addition, all 21 patients and/or caretakers responded to a questionnaire at the time of this study by means of telephone to assess subjective pain, analgesia use, walking aid necessity, walking distance, and satisfaction with the procedure.

RESULTS: In this series of 21 cerebral palsy patients, 5 patients had bilateral surgery, resulting in 26 operative feet. The mean age at the time of surgery was 19.4 years and most recent clinical or radiographic follow-up was 22.1 years postoperatively. Preoperative foot deformity was characterized by hindfoot valgus in 66.7% (14/21) and varus in 33.3% (7/21) of patients. Postoperatively, fusion was achieved in 96.2% (25/26) of feet. At final follow-up, 3 feet (11.5%) demonstrated tibiotalar joint arthritis, 1 (3.8%) had midfoot arthritis, and 10 (38.5%) had residual deformity. Of the total patients, 95.2% (20/21) were satisfied with the outcome and 61.9% (13/21) reported pain-free ambulation. There was no association between eventual functional outcome and preoperative diagnosis, preoperative foot deformity, postoperative tibiotalar joint arthritis, or postoperative residual deformity.

CONCLUSIONS: Triple arthrodesis is a surgical option in cerebral palsy patients with painful and/or rigid foot deformities. From this series, successful outcomes can be expected as long as bony union is achieved. The incidence of tibiotalar arthritis is relatively low and not associated with long-term functional outcome. In addition, preoperative and residual postoperative foot deformity is not

associated with long-term outcome.

PMID: 25393571 [PubMed - indexed for MEDLINE]

#### Perioperative complications of orthopedic surgery for lower extremity in patients with cerebral palsy.

Lee SY, Sohn HM, Chung CY Do SH, Lee KM, Kwon SS, Sung K, Lee SH, Park MS.

J Korean Med Sci. 2015 Apr;30(4):489-94. doi: 10.3346/jkms.2015.30.4.489. Epub 2015 Mar 19.

Because complications are more common in patients with cerebral palsy (CP), surgeons and anesthesiologists must be aware of perioperative morbidity and be prepared to recognize and treat perioperative complications. This study aimed to determine the incidence of and risk factors for perioperative complications of orthopedic surgery on the lower extremities in patients with CP. We reviewed the medical records of consecutive CP patients undergoing orthopedic surgery. Medical history, anesthesia emergence time, intraoperative body temperature, heart rate, blood pressure, immediate postoperative complications, Gross Motor Function Classification System (GMFCS) level, Cormack-Lehane classification, and American Society of Anesthesiologists physical status classification were analyzed. A total of 868 patients was included. Mean age at first surgery was 11.8 (7.6) yr. The incidences of intraoperative hypothermia, absolute hypotension, and absolute bradycardia were 26.2%, 4.4%, and 20.0%, Science Infos Paralysie Cérébrale, mai 2016, FONDATION MOTRICE 67 rue Vergniaud 75013 Paris - tel +33 1 30 45 54 03 03 contact: Christine Doumergue cdoumergue@lafondationmotrice.org respectively. Twenty (2.3%) patients had major complications, and 35 (4.0%) patients had minor complications postoperatively. The incidences of intraoperative hypothermia, absolute hypotension, and major postoperative complications were significantly higher in patients at GMFCS levels IV and V compared with patients at GMFCS levels I to III (P<0.001). History of pneumonia was associated with intraoperative absolute hypotension and major postoperative complications (P<0.001). These results revealed that GMFCS level, patient age, hip reconstructive surgery, and history of pneumonia are associated with adverse effects on intraoperative body temperature, the cardiovascular system, and immediate postoperative complications.

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PMCID: PMC4366972 PMID: 25829819 [PubMed - indexed for MEDLINE]

#### Salvage procedures for the painful chronically dislocated hip in cerebral palsy.

Hwang JH, Varte L, Kim HW, Lee DH Park H. Bone Joint J. 2016 Jan;98-B(1):137-43. doi: 10.1302/0301-620X.98B1.35202.

The aims of this study were to report functional outcomes of salvage procedures for patients with cerebral palsy (CP) who have chronic dislocation of the hip using validated scoring systems, and to compare the results of three surgical techniques. We reviewed 37 patients retrospectively. The mean age at the time of surgery was 12.2 years (8 to 22) and the mean follow-up was 56 months (24 to 114). Patients were divided into three groups: 14 who underwent proximal femoral resection arthroplasty (PFRA group 1), ten who underwent subtrochanteric valgus osteotomy (SVO group 2), and 13 who underwent subtrochanteric valgus osteotomy with resection of the femoral head (SVO with FHR group 3). All patients were evaluated using the Caregiver Priorities and Child Health Index of Life with Disabilities CPCHILD) and the Pediatric Quality of Life Inventory (PedsQL). Significant improvements occurred in most CPCHILD and PedsQL subsection scores following surgery in all patients, without significant differences between the groups. There were 12 post-operative complications. Less severe complications were seen in group 1 than in groups 2 and 3. Salvage surgery appears to provide pain relief in patients with CP who have painful chronic dislocation of the hip. The three salvage procedures produced similar results, however, we recommend the use of PFRA as the complications are less severe.TAKE HOME MESSAGE: Salvage surgery can be of benefit to patients with CP with chronic painful hip dislocation, but should be limited to selected patients considering complications.

PMID: 26733527 [PubMed - indexed for MEDLINE]

#### **Réadaptation fonctionnelle**

A Perceptual Motor Intervention Improves Play Behavior in Children with Moderate to Severe Cerebral Palsy.

Ryalls BO, Harbourne R, Kelly-Vance L, Wickstrom J, Stergiou N, Kyvelidou A. *Front Psychol. 2016 May 3;7:643. doi: 10.3389/fpsyg.2016.00643. eCollection 2016.* 

For children with moderate or severe cerebral palsy (CP), a foundational early goal is independent sitting. Sitting offers additional opportunities for object exploration, play and social engagement. The achievement of sitting coincides with important milestones in other developmental areas, such as social engagement with others, understanding of spatial relationships, and the use of both hands to explore objects. These milestones are essential skills necessary for play behavior. However, little is known about how sitting and play behavior might be affected by a physical therapy intervention in children with moderate or severe CP. Therefore, our overall purpose in this study was to determine if sitting skill could be advanced in children with moderate to severe CP using a perceptual motor intervention, and if play skills would change significantly as sitting advanced. Thirty children between the ages of 18 months and 6 years who were able to hold prop sitting for at least 10 s were recruited for this study. Outcome measures were the sitting subsection of the Gross Motor Function Measure (GMFM), and the Play Assessment of Children with Motor Impairment play assessment scale, which is a modified version of the Play in Early Childhood Evaluation System. Significant improvements in GMFM sitting scores (p < 0.001) and marginally significant improvement in play assessment scores (p = 0.067) were found from pre- to post-intervention. Sitting change explained a significant portion of the variance in play change for children over the age of 3 years, who were more severely affected by CP. The results of this study indicate that advances in sitting skill may be a factor in supporting improvements in functional play, along with age and severity of physical impairment.

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## A Prospective Case-Control Study of Radial Extracorporeal Shock Wave Therapy for Spastic Plantar Flexor Muscles in Very Young Children With Cerebral Palsy.

Wang T, Du L, Shan L, Dong H, Feng J, Kiessling MC, Angstman NB, Schmitz C, Jia F. Medicine (Baltimore). 2016 May;95(19):e3649. doi: 10.1097/MD.000000000003649.

To assess the effects of radial extracorporeal shock wave therapy (rESWT) on plantar flexor muscle spasticity and gross motor function in very young patients with cerebral palsy (CP). The design was case-control study (level of evidence 3). The setting was the Department of Pediatric Neurology and Neurorehabilitation, First Hospital of Jilin University, Changchun, China. Those with a diagnosis of CP and spastic plantar flexor muscles were recruited between April 2014 and April 2015. According to the parents' decision, patients received 1 ESWT session per week for 3 months, with 1500 radial shock waves per ESWT session and leg with positive energy flux density of 0.03 mJ/mm, combined with traditional conservative therapy (rESWT group) or traditional conservative therapy alone (control group). The Modified Ashworth Scale (MAS) (primary outcome measure) and passive range of motion (pROM) measurements were collected at baseline (BL), 1 month (M1), and 3 months (M3) after BL. The Gross Motor Function Measure (GMFM)-88 was collected at BL and M3.Sixty-six patients completed the final review at 3 months and were included in the study. Subjects ranged in age from 12 to 60 months (mean age 27.0±13.6 months; median age 22.0 months; 33.3% female). For the rESWT group (n = 34), mean MAS grades at BL, M1, and M3 were 2.6, 1.9, and 1.5 on the left side and 1.9, 1.7, and 1.2 on the right side. For the control group (n = 32), mean MAS grades at BL, M1, and M3 were 2.5, 2.4, and 2.1 on the left side and 1.8, 1.8, and 1.5 on the right side. The within-subject effects time×side and time×treatment were statistically significant (P<0.01). Similar results were found for the improvement of mean pROM. GMFM-88 improved from BL to M3, but showed no statistically significant difference between the groups. There were no significant complications. This study demonstrates that the combination of rESWT and traditional conservative therapy is more effective than traditional conservative therapy alone in the treatment of spasticity in very young patients with CP.

#### Free Article

PMID: 27175689 [PubMed - in process]

Comparison of Rehabilitation Outcomes for Long Term Neurological Conditions: A Cohort Analysis of the Australian Rehabilitation Outcomes Centre Dataset for Adults of Working Age.

Turner-Stokes L, Vanderstay R, Stevermuer T, Simmonds F, Khan F, Eagar K.

PLoS One. 2015 Jul 13;10(7):e0132275. doi: 10.1371/journal.pone.0132275. eCollection 2015.

OBJECTIVE: To describe and compare outcomes from in-patient rehabilitation (IPR) in working-aged adults across different groups of long-term neurological conditions, as defined by the UK National Service Framework.

DESIGN: Analysis of a large Australian prospectively collected dataset for completed IPR episodes (n = 28,596) from 2003-2012.

METHODS: De-identified data for adults (16-65 years) with specified neurological impairment codes were extracted, cleaned and divided into 'Sudden-onset' conditions: (Stroke (n = 12527), brain injury (n = 7565), spinal cord injury (SCI) (n = 3753), Guillain-Barré syndrome (GBS) (n = 805)) and 'Progressive/stable' conditions (Progressive (n = 3750) and Cerebral palsy (n = 196)). Key outcomes included Functional Independence Measure (FIM) scores, length of stay (LOS), and discharge destination.

RESULTS: Mean LOS ranged from 21-57 days with significant group differences in gender, source of admission and discharge destination. All six groups showed significant change (p<0.001) between admission and discharge that was likely to be clinically important across a range of items. Significant between-group differences were observed for FIM Motor and Cognitive change scores (Kruskal-Wallis p<0.001), and item-by-item analysis confirmed distinct patterns for each of the six groups. SCI and GBS patients were generally at the ceiling of the cognitive subscale. The 'Progressive/stable' conditions made smaller improvements in FIM score than the 'Sudden-onset conditions', but also had shorter LOS.

CONCLUSION: All groups made gains in independence during admission, although pattern of change varied between conditions, and ceiling effects were observed in the FIM-cognitive subscale. Relative cost-efficiency between groups can only be indirectly inferred. Limitations of the current dataset are discussed, together with opportunities for expansion and further development.

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#### Development of an EMG-ACC-Based Upper Limb Rehabilitation Training System.

Liu L, Chen X, Lu Z, Cao S, Wu, Zhang X. IEEE Trans Neural Syst Rehabil Eng. 2016 Apr 29. [Epub ahead of print]

This paper focuses on the development of an upper limb rehabilitation training system designed for use by children with cerebral palsy (CP). It attempts to meet the requirements of in-home training by taking advantage of the combination of portable accelerometers (ACC) and surface electromyography (SEMG) sensors worn on the upper limb to capture functional movements. In the proposed system, the EMG-ACC acquisition device works essentially as wireless game controller, and three rehabilitation games were designed for improving upper limb motor function under a clinician's guidance. The games were developed on the Android platform based on a physical engine called Box2D. The results of a system performance test demonstrated that the developed games can respond to the upper limb actions within 210ms. Positive questionnaire feedbacks from twenty CP subjects who participated in the game test verified both the feasibility and usability of the system. Results of a long-term game training conducted with three CP subjects demonstrated that CP patients could improve in their game performance through repetitive training, and persistent training was needed to improve and enhance the rehabilitation effect. According to our experimental results, the novel multi-feedback SEMG-ACC-based user interface improved the users' initiative and performance in rehabilitation training.

PMID: 27164594 [PubMed - as supplied by publisher]

## External Mechanical Work and Pendular Energy Transduction of Overground and Treadmill Walking in Adolescents with Unilateral Cerebral Palsy.

Zollinger M, Degache F, Currat G, Pochon L, Peyrot N, Newman CJ, Malatesta D. Front Physiol. 2016 Apr 13;7:121. doi: 10.3389/fphys.2016.00121. eCollection 2016.

PURPOSE: Motor impairments affect functional abilities and gait in children and adolescents with cerebral palsy (CP). Improving their walking is an essential objective of treatment, and the use of a treadmill for gait analysis and training could offer several advantages in adolescents with CP. However, there is a controversy regarding the similarity between treadmill and overground walking both for gait analysis and training in children and adolescents. The aim of this study was to compare the external mechanical work and pendular energy transduction of these two types of gait modalities at standard and preferred walking speeds in adolescents with unilateral cerebral palsy (UCP) and typically developing (TD) adolescents matched on age, height and body mass.

METHODS: Spatiotemporal parameters, external mechanical work and pendular energy transduction of walking were computed using two inertial sensors equipped with a triaxial accelerometer and gyroscope and compared in 10 UCP (14.2  $\pm$  1.7 year) and 10 TD (14.1  $\pm$  1.9 year) adolescents during treadmill and overground walking at standard and preferred speeds.

RESULTS: The treadmill induced almost identical mechanical changes to overground walking in TD adolescents and those with UCP, with the exception of potential and kinetic vertical and lateral mechanical works, which are both significantly increased in the overground-treadmill transition only in UCP (P < 0.05).

CONCLUSIONS: Adolescents with UCP have a reduced adaptive capacity in absorbing and decelerating the speed created by a treadmill (i.e., dynamic stability) compared to TD adolescents. This may have an important implication in rehabilitation programs that assess and train gait by using a treadmill in adolescents with UCP.

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PMCID: PMC4829600 PMID: 27148062 [PubMed]

# FAST CP: protocol of a randomised controlled trial of the efficacy of a 12-week combined Functional Anaerobic and Strength Training programme on muscle properties and mechanical gait deficiencies in adolescents and young adults with spastic-type cerebral palsy.

Gillett JG, Lichtwark GA, Boyd RN, Barber LA BMJ Open. 2015 Jun 26;5(6):e008059. doi: 10.1136/bmjopen-2015-008059.

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Erratum in BMJ Open. 2015;5(7):e008059corr1.

INTRODUCTION: Individuals with cerebral palsy (CP) have muscles that are smaller, weaker and more resistant to stretch compared to typically developing people. Progressive resistance training leads to increases in muscle size and strength. In CP, the benefits of resistance training alone may not transfer to improve other activities such as walking; however, the transfer of strength improvements to improved mobility may be enhanced by performing training that involves specific functional tasks or motor skills. This study aims to determine the efficacy of combined functional anaerobic and strength training in (1) influencing muscle strength, structure and function and (2) to determine if any changes in muscle strength and structure following training impact on walking ability and gross motor functional capacity and performance in the short (following 3 months of training) and medium terms (a further 3 months post-training).

METHODS AND ANALYSIS: 40 adolescents and young adults with CP will be recruited to undertake a 12-week training programme. The training programme will consist of  $3 \times 75$  min sessions per week, made up of 5 lower limb resistance exercises and 2-3 functional anaerobic exercises per session. The calf muscles will be specifically targeted, as they are the most commonly impacted muscles in CP and are a key muscle group involved in walking. If, as we believe, muscle properties change following combined strength and functional training, there may be long-term benefits of this type of training in slowing the deterioration of muscle function in people with spastic-type CP.

ETHICS AND DISSEMINATION: Ethical approval has been obtained from the ethics committees at The University of Queensland (2014000066) and Children's Health Queensland (HREC/15/QRCH/30). The findings will be disseminated by publications in peer-reviewed journals, conferences and local research organisations' media.

TRIAL REGISTRATION NUMBER: Australian and New Zealand Clinical Trials Registry (ACTRN12614001217695).

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NOOIJENPMCID: PMC4486965 PMID: 26116614 [PubMed - indexed for MEDLIN

## Improvements in bimanual hand function after baby-CIMT in two-year old children with unilateral cerebral palsy: A retrospective study.

Nordstrand L, Holmefur M, Kits A, Eliasson AC Res Dev Disabil. 2015 Jun-Jul;41-42:86-93. doi: 10.1016/j.ridd.2015.05.003. Epub 2015 Jun 19.

The common assumption that early-onset intensive intervention positively affects motor development has rarely been investigated for hand function in children with unilateral cerebral palsy (CP). This retrospective study explored the possible impact of baby constraint-induced movement therapy (baby-CIMT) on hand function at two years of age. We hypothesized that baby-CIMT in the first year of life would lead to better bimanual hand use at two years of age than would not receiving baby-CIMT. The Assisting Hand Assessment (AHA) was administered at age 21 months (SD 2.4 months) in 72 children with unilateral CP, 31 of who received baby-CIMT. When dividing the children into four functional levels based on AHA, the proportional distribution differed between the groups in favour of baby-CIMT. Logistic regression analysis indicated that children in the baby-CIMT group were more likely than were children in the no baby-CIMT group to have a high functional level, even when controlling for the effect of brain lesion type (OR 5.83, 95% CI 1.44-23.56, p = 0.001). However, no difference was found between groups in the odds of having a very low functional level (OR 0.31, 95% CI 0.08-1.17, p = 0.084). The result shows that baby-CIMT at early age can have a positive effect. Children who received baby-CIMT group.

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PMID: 26100242 [PubMed - indexed for MEDLINE]

## Increased Adaptation Rates and Reduction in Trial-by-Trial Variability in Subjects with Cerebral Palsy Following a Multi-session Locomotor Adaptation Training.

Mawase F, Bar-Haim S, Joubran K, Rubin L, Karniel A, Shmuelof L Front Hum Neurosci. 2016 May 4;10:203. doi: 10.3389/fnhum.2016.00203. eCollection 2016.

Cerebral Palsy (CP) results from an insult to the developing brain and is associated with deficits in locomotor and manual skills and in sensorimotor adaptation. We hypothesized that the poor sensorimotor adaptation in persons

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with CP is related to their high execution variability and does not reflect a general impairment in adaptation learning. We studied the interaction between performance variability and adaptation deficits using a multi-session locomotor adaptation design in persons with CP. Six adolescents with diplegic CP were exposed, during a period of 15 weeks, to a repeated split-belt treadmill perturbation spread over 30 sessions and were tested again 6 months after the end of training. Compared to age-matched healthy controls, subjects with CP showed poor adaptation and high execution variability in the first exposure to the perturbation. Following training they showed marked reduction in execution variability and an increase in learning rates. The reduction in variability and the improvement in adaptation were highly correlated in the CP group and were retained 6 months after training. Interestingly, despite reducing their variability in the washout phase, subjects with CP did not improve learning rates during washout phases that were introduced only four times during the experiment. Our results suggest that locomotor adaptation in subjects with CP is related to their execution variability. Nevertheless, while variability reduction is generalized to other locomotor contexts, the development of savings requires both reduction in execution variability and multiple exposures to the perturbation.

#### Free PMC Article

PMCID: PMC4854882 PMID: 27199721 [PubMed]

## Reliability and Validity of Objective Measures of Physical Activity in Youth With Cerebral Palsy Who Are Ambulatory.

O'Neil ME, Fragala-Pinkham M, Lennon N, George A, Forman J, Trost SG Phys Ther. 2016 Jan;96(1):37-45. doi: 10.2522/ptj.20140201. Epub 2015 Jun 18.

BACKGROUND: Physical therapy for youth with cerebral palsy (CP) who are ambulatory includes interventions to increase functional mobility and participation in physical activity (PA). Thus, reliable and valid measures are needed to document PA in youth with CP.

OBJECTIVE: The purpose of this study was to evaluate the inter-instrument reliability and concurrent validity of 3 accelerometer-based motion sensors with indirect calorimetry as the criterion for measuring PA intensity in youth with CP.

METHODS: Fifty-seven youth with CP (mean age=12.5 years, SD=3.3; 51% female; 49.1% with spastic hemiplegia) participated. Inclusion criteria were: aged 6 to 20 years, ambulatory, Gross Motor Function Classification System (GMFCS) levels I through III, able to follow directions, and able to complete the full PA protocol. Protocol activities included standardized activity trials with increasing PA intensity (resting, writing, household chores, active video games, and walking at 3 self-selected speeds), as measured by weight-relative oxygen uptake (in mL/kg/min). During each trial, participants wore bilateral accelerometers on the upper arms, waist/hip, and ankle and a portable indirect calorimeter. Intraclass coefficient correlations (ICCs) were calculated to evaluate inter-instrument reliability (left-to-right accelerometer placement). Spearman correlations were used to examine concurrent validity between accelerometer output (activity and step counts) and indirect calorimetry. Friedman analyses of variance with post hoc pair-wise analyses were conducted to examine the validity of accelerometers to discriminate PA intensity across activity trials.

RESULTS: All accelerometers exhibited excellent inter-instrument reliability (ICC=.94-.99) and good concurrent validity (rho=.70-.85). All accelerometers discriminated PA intensity across most activity trials.

LIMITATIONS: This PA protocol consisted of controlled activity trials.

CONCLUSIONS: Accelerometers provide valid and reliable measures of PA intensity among youth with CP.

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PMCID: PMC4706594 [Available on 2016-07-01]

PMID: 26089043 [PubMed - indexed for MEDLINE]

## Single blind randomised controlled trial of GAME (Goals - Activity – Motor Enrichment) in infants at high risk of cerebral palsy.

Morgan C, Novak I, Dale RC, Guzzetta A, Badawi N Res Dev Disabil. 2016 May 7;55:256-267. doi: 10.1016/j.ridd.2016.04.005. [Epub ahead of print]

BACKGROUND: Cerebral palsy (CP) is caused by a lesion in the developing infant brain. Recent neuroplasticity literature suggests that intensive, task-specific intervention ought to commence early, during the critical period of neural development.

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AIMS: To determine whether "GAME" (Goals - Activity - Motor Enrichment), a motor learning, environmental enrichment intervention, is effective for improving motor skills in infants at high risk of CP.

METHODS AND PROCEDURES: Single blind randomised controlled trial of GAME versus standard care. Primary outcome was motor skills on the Peabody Developmental Motor Scales-2 (PDMS-2). Secondary outcomes included Canadian Occupational Performance Measure (COPM), Bayley Scales of Infant and Toddler Development (BSID-III) and Gross Motor Function Measure-66 (GMFM-66). Outcome assessors were masked to group allocation and data analyzed with multiple regression.

OUTCOMES AND RESULTS: All n=30 infants enrolled received the assigned intervention until 16 weeks post enrolment. At 12 months of age, n=26 completed assessments. Significant between group differences were found in raw scores on the PDMS-2 in favour of GAME (B=20.71, 95%CI 1.66-39.76, p=0. 03) and at 12 months on the total motor quotient (B=8.29, 95%CI 0.13-16.45,p =0.05). Significant between group differences favored GAME participants at 12 months on the cognitive scale of the BSID-III and satisfaction scores on the COPM.

CONCLUSION: GAME intervention resulted in advanced motor and cognitive outcomes when compared with standard care.

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PMID: 27164480 [PubMed - as supplied by publisher]

## The Effects of a Home-Based Connective Tissue Targeting Therapy on Hip Development in Children With Cerebral Palsy: Six Case Reports.

Drewes E, Driscoll M, Blyum L, Vincentz D Explore (NY). 2016 Apr 19. pii: S1550-8307(16)30025-8. doi: 10.1016/j.explore.2016.04.004. [Epub ahead of print]

Hip subluxation in children with Cerebral Palsy (CP) has an incidence of 10-30 %, and children with severe CP having the highest incidence. The condition deteriorates if left untreated. Surgery is the most common method used in managing hip subluxation because standard conservative therapies do not improve it. Surgery may have to be repeated and comes at a biological cost to the child. A new home-based CAM, Advanced Biomechanical Rehabilitation (ABR), has shown encouraging results leading to improved spinal stability and stability in sitting in children with severe CP. This case report examines hip development over time in six children with severe CP in the ABR Program. Changes in their clinical picture and pelvic X-Rays are reported. ABR appeared to help stabilize and improve hip subluxation, resulting in these children not requiring further surgical intervention. These findings warrant further investigation of ABR as a noninvasive therapy for hip subluxation.

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PMID: 27198038 [PubMed - as supplied by publisher]

## WalkAide Efficacy on Gait and Energy Expenditure in Children with Hemiplegic Cerebral Palsy: A Randomized Controlled Trial.

El-Shamy SM, Abdelaal AA. Am J Phys Med Rehabil. 2016 May 4. [Epub ahead of print]

oBJECTIVE: The aim of this study was to investigate the effects of WalkAide functional electrical stimulation on gait pattern and energy expenditure in children with hemiplegic cerebral palsy.

DESIGN: Seventeen children were assigned to the study group, whose members received functional electrical stimulation (pulse width, 300 µs; frequency, 33 Hz, 2 hours/d, 3 days/week for 3 consecutive months). Seventeen other children were assigned to the control group, whose members participated in a conventional physical therapy exercise program for 3 successive months. Baseline and posttreatment assessments were performed using the GAITRite system to evaluate gait parameters and using an open-circuit indirect calorimeter to evaluate energy expenditure.

RESULTS: Children in the study group showed a significant improvement when compared with those in the control group (P < 0.005). The gait parameters (stride length, cadence, speed, cycle time, and stance phase percentage) after treatment were (0.74 m,119 steps/min, 0.75 m/s, 0.65 s, 55.9%) and (0.5 m,125 steps/min, 0.6 m/s, 0.49 s, 50.4%) for the study group and control group, respectively. The mean energy expenditures after treatment were 8.18  $\pm$  0.88 and 9.16  $\pm$  0.65 mL/kg per minute for the study and control groups, respectively.

CONCLUSIONS: WalkAide functional electrical stimulation may be a useful tool for improving gait pattern and energy expenditure in children with hemiplegic cerebral palsy.

PMID: 27149586 [PubMed - as supplied by publisher]

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#### **Orthèse**

## An individual approach for optimizing ankle-foot orthoses to improve mobility in children with spastic cerebral palsy walking with excessive knee flexion.

*Gait Posture. 2016 May;46:104-11. doi: 10.1016/j.gaitpost.2016.03.001. Epub 2016 Mar 9.* Kerkum YL, Harlaar J, Buizer AI, van den Noort JC, Becher JG, Brehm MA

Ankle-Foot Orthoses (AFOs) are commonly prescribed to promote gait in children with cerebral palsy (CP). The AFO prescription process is however largely dependent on clinical experience, resulting in confusing results regarding treatment efficacy. To maximize efficacy, the AFO's mechanical properties should be tuned to the patient's underlying impairments. This study aimed to investigate whether the efficacy of a ventral shell AFO (vAFO) to reduce knee flexion and walking energy cost could be improved by individually optimizing AFO stiffness in children with CP walking with excessive knee flexion. Secondarily, the effect of the optimized vAFO on daily walking activity was investigated. Fifteen children with spastic CP were prescribed with a hinged vAFO with adjustable stiffness. Effects of a rigid, stiff, and flexible setting on knee angle and the net energy cost (EC) [Jkg(-1)m(-1)] were assessed to individually select the optimal stiffness. After three months, net EC, daily walking with shoes-only. A near significant 9% (p=0.077) decrease in net EC (-0.5Jkg(-1)m(-1)) was found for walking with the optimized vAFO compared to shoes-only. Daily activity remained unchanged. Knee flexion in stance was reduced by 2.4° (p=0.006). These results show that children with CP who walk with excessive knee flexion show a small, but significant reduction of knee flexion in stance as a result of wearing individually optimized vAFOs. Data suggest that this also improves gait efficiency for which an individual approach to AFO prescription is emphasized.

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PMID: 27131186 [PubMed - in process]

## Minimising impairment: Protocol for a multicentre randomised controlled trial of upper limb orthoses for children with cerebral palsy.

Imms C, Wallen M, Elliott C, Hoare B, Randall M, Greaves S, Adair B, Bradshaw E, Carter R, Orsini F, Shih ST, Reddihough D

BMC Pediatr. 2016 May 27;16(1):70. doi: 10.1186/s12887-016-0608-8.

BACKGROUND: Upper limb orthoses are frequently prescribed for children with cerebral palsy (CP) who have muscle overactivity predominantly due to spasticity, with little evidence of long-term effectiveness. Clinical consensus is that orthoses help to preserve range of movement: nevertheless, they can be complex to construct, expensive, uncomfortable and require commitment from parents and children to wear. This protocol paper describes a randomised controlled trial to evaluate whether long-term use of rigid wrist/hand orthoses (WHO) in children with CP, combined with usual multidisciplinary care, can prevent or reduce musculoskeletal impairments, including muscle stiffness/tone and loss of movement range, compared to usual multidisciplinary care alone. METHODS/DESIGN: This pragmatic, multicentre, assessor-blinded randomised controlled trial with economic analysis will recruit 194 children with CP, aged 5-15 years, who present with flexor muscle stiffness of the wrist and/or fingers/thumb (Modified Ashworth Scale score  $\geq$ 1). Children, recruited from treatment centres in Victoria, New South Wales and Western Australia, will be randomised to groups (1:1 allocation) using concealed procedures. All children will receive care typically provided by their treating organisation. The treatment group will receive a custom-made serially adjustable rigid WHO, prescribed for 6 h nightly (or daily) to wear for 3 years. An application

developed for mobile devices will monitor WHO wearing time and adverse events. The control group will not receive a WHO, and will cease wearing one if previously prescribed. Outcomes will be measured 6 monthly over a period of 3 years. The primary outcome is passive range of wrist extension, measured with fingers extended using a goniometer at 3 years. Secondary outcomes include muscle stiffness, spasticity, pain, grip strength and hand deformity. Activity, participation, quality of life, cost and cost-effectiveness will also be assessed.

DISCUSSION: This study will provide evidence to inform clinicians, services, funding agencies and parents/carers of children with CP whether the provision of a rigid WHO to reduce upper limb impairment, in combination with usual multidisciplinary care, is worth the effort and costs.

TRIAL REGISTRATION: ANZ Clinical Trials Registry: U1111-1164-0572.

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#### Stimulation cérébrale - Stimulation neurosensorielle

#### Transcranial Direct-Current Stimulation Can Enhance Motor Learning in Children.

Ciechanski P, Kirton A Cortex. 2016 May 10. pii: bhw114. [Epub ahead of print]

This study aims to determine the effects of transcranial direct-current stimulation (tDCS) on motor learning in healthy school-aged children. Safety, tolerability, and translation of effects to untrained tasks were also explored. We recruited 24 right-handed children for a randomized, sham-controlled, double-blinded trial to receive: right primary motor cortex (M1) 1 mA anodal (1A-tDCS), left M1 1 mA cathodal (1C-tDCS), left M1 2 mA cathodal tDCS (2C-tDCS), or sham tDCS over 3 consecutive days of motor task practice. Participants trained their left hand to perform the Purdue Pegboard Test (PPT) during tDCS application. Right hand and bimanual PPT, the Jebsen-Taylor Test (JTT), and the Serial Reaction Time Task (SRTT) were tested at baseline and post-training. All measures were retested 6 weeks later. Active tDCS montages enhanced motor learning compared with sham (all P < 0.002). Effects were sustained at 6 weeks. Effect sizes were large and comparable across montages: contralateral 1A-tDCS (Cohen's d = 2.58) and ipsilateral 1C-tDCS (3.44) and 2C-tDCS (2.76). Performance in the untrained hand PPT, bilateral JTT, and SRTT often improved with tDCS. tDCS was well-tolerated and safe with no adverse events. These first principles will advance the pairing of tDCS with therapy to enhance rehabilitation for disabled children such as those with cerebral palsy.

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## Transcranial direct current stimulation combined with integrative speech therapy in a child with cerebral palsy: A case report.

Carvalho Lima VL, Collange Grecco LA, Marques VC, Fregni F, Brandão de Ávila CR J Body Mov Ther. 2016 Apr;20(2):252-7. doi: 10.1016/j.jbmt.2015.03.007. Epub 2015 Mar 25.

The aim of this study was to describe the results of the first case combining integrative speech therapy with anodal transcranial direct current stimulation (tDCS) over Broca's area in a child with cerebral palsy. The ABFW phonology test was used to analyze speech based on the Percentage of Correct Consonants (PCC) and Percentage of Correct Consonants - Revised (PCC-R). After treatment, increases were found in both PCC (Imitation: 53.63%-78.10%; Nomination: 53.19%-70.21%) and PPC-R (Imitation: 64.54%-83.63%; Nomination: 61.70%-77.65%). Moreover, reductions occurred in distortions, substitutions and improvement was found in oral performance, especially tongue mobility (AMIOFE-mobility before = 4 after = 7). The child demonstrated a clinically important improvement in speech fluency as shown in results of imitation number of correct consonants and phonemes acquire. Based on these promising findings, continuing research in this field should be conducted with controlled clinical trials. Copyright © 2015 Elsevier Ltd. All rights reserved.

PMID: 27210840 [PubMed - in process]

#### Transcranial direct current stimulation in children and adolescents: a comprehensive review.

Palm U, Segmiller FM, Epple AN), Freisleder FJ, Koutsouleris N, Schulte-Körne G, Padberg F J Neural Transm (Vienna). 2016 May 12. [Epub ahead of print]

Transcranial direct current stimulation (tDCS) is a non-invasive brain stimulation method that has shown promising results in various neuropsychiatric disorders in adults. This review addresses the therapeutic use of tDCS in children and adolescents including safety, ethical, and legal considerations. There are several studies addressing the dosage of tDCS in children and adolescents by computational modeling of electric fields in the pediatric brain. Results suggest halving the amperage used in adults to obtain the same peak electric fields, however, there are some studies reporting on the safe application of tDCS with standard adult parameters in children (2 mA; 20-30 min). There are several randomized placebo controlled trials suggesting beneficial effects of tDCS for the treatment of cerebral palsy.

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For dystonia there are mixed data. Some studies suggest efficacy of tDCS for the treatment of refractory epilepsy, and for the improvement of attention deficit/hyperactivity disorder and autism. Interestingly, there is a lack of data for the treatment of childhood and adolescent psychiatric disorders, i.e., childhood onset schizophrenia and affective disorders. Overall, tDCS seems to be safe in pediatric population. More studies are needed to confirm the preliminary encouraging results; however, ethical deliberation has to be weighed carefully for every single case. PMID: 27173384 [PubMed - as supplied by publisher]

#### Réalité virtuelle - Jeux video

#### Efficacy of neurodevelopmental treatment combined with the Nintendo(<sup>®</sup>) Wii in patients with cerebral palsy.

Acar G, Altun GP, Yurdalan S, Polat MG

J Phys Ther Sci. 2016 Mar;28(3):774-80. doi: 10.1589/jpts.28.774. Epub 2016 Mar 31.

[Purpose] The aim of this study was to investigate the efficiency of Nintendo(<sup>®</sup>) Wii games in addition to neurodevelopmental treatment in patients with cerebral palsy. [Subjects and Methods] Thirty hemiparetic cerebral palsy patients (16 females, 14 males; mean age, 6-15 years) were included in the study and divided into two groups: a neurodevelopmental treatment+Nintendo Wii group (group 1, n=15) and a neurodevelopmental treatment group (group 2, n=15). Both groups received treatment in 45-minute sessions 2 days/week for six weeks. Use of the upper extremities, speed, disability and functional independence were evaluated using the Quality of Upper Extremity Skills Test, Jebsen Taylor Hand Function Test, ABILHAND-Kids test, and Pediatric Functional Independence Measure (self-care) before and after treatment. [Results] There were statistically significant improvements in all parameters for group 1 and group 2 (except quality of function) after six weeks of treatment. Intergroup analysis showed that group 1 was superior to group 2 in mean change differences in the Jebsen Taylor Hand Function Test. [Conclusion] Our results showed that neurodevelopmental treatment is effective for improving hand functions in hemiplegic cerebral palsy. To provide a enjoyable, motivational, safe, and effective rehabilitation program, the Nintendo(<sup>®</sup>) Wii may be used in addition to neurodevelopmental treatment.

Free PMC Article PMCID: PMC4842438 PMID: 27134357 [PubMed]

#### Thérapie cellulaire Médecine regénérative

#### Human Umbilical Cord Blood Cells Ameliorate Motor Deficits in Rabbits in a Cerebral Palsy Model.

Drobyshevsky A, Cotten CM, Shi Z, Luo K, Jiang R, Derrick M, Tracy ET, Gentry T, Goldberg RN, Kurtzberg J, Tan S. *Dev Neurosci.* 2015;37(4-5):349-62. doi: 10.1159/000374107. Epub 2015 Mar 17.

Cerebral palsy (CP) has a significant impact on both patients and society, but therapy is limited. Human umbilical cord blood cells (HUCBC), containing various stem and progenitor cells, have been used to treat various brain genetic conditions. In small animal experiments, HUCBC have improved outcomes after hypoxic-ischemic (HI) injury. Clinical trials using HUCBC are underway, testing feasibility, safety and efficacy for neonatal injury as well as CP. We tested HUCBC therapy in a validated rabbit model of CP after acute changes secondary to HI injury had subsided. Following uterine ischemia at 70% gestation, we infused HUCBC into newborn rabbit kits with either mild or severe neurobehavioral changes. Infusion of high-dose HUCBC (5 × 10(6) cells) dramatically altered the natural history of the injury, alleviating the abnormal phenotype including posture, righting reflex, locomotion, tone, and dystonia. Half the high dose showed lesser but still significant improvement. The swimming test, however, showed that joint function did not restore to naïve control function in either group. Tracing HUCBC with either MRI biomarkers or PCR for human DNA found little penetration of HUCBC in the newborn brain in the immediate newborn period, suggesting that the beneficial effects were not due to cellular integration or direct proliferative effects but rather to paracrine signaling. This is the first study to show that HUCBC improve motor performance in a dose-dependent manner, perhaps by improving compensatory repair processes.

#### Free Article

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#### How to make an oligodendrocyte.

Goldman SA, Kuypers NJ Development. 2015 Dec 1;142(23):3983-95. doi: 10.1242/dev.126409.

Oligodendrocytes produce myelin, an insulating sheath required for the saltatory conduction of electrical impulses along axons. Oligodendrocyte loss results in demyelination, which leads to impaired neurological function in a broad array of diseases ranging from pediatric leukodystrophies and cerebral palsy, to multiple sclerosis and white matter stroke. Accordingly, replacing lost oligodendrocytes, whether by transplanting oligodendrocyte progenitor cells OPCs) or by mobilizing endogenous progenitors, holds great promise as a therapeutic strategy for the diseases of central white matter. In this Primer, we describe the molecular events regulating oligodendrocyte development and how our understanding of this process has led to the establishment of methods for producing OPCs and oligodendrocytes from embryonic stem cells and induced pluripotent stem cells, as well as directly from somatic cells. In addition, we will discuss the safety of engrafted stem cell-derived OPCs, as well as approaches by which to modulate their differentiation and myelinogenesis in vivo following transplantation.

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#### Free Article

PMCID: PMC4712837 [Available on 2016-12-01] PMID: 26628089 [PubMed - indexed for MEDLINE]

#### Platelet-Rich Plasma in a Patient with Cerebral Palsy.

Alcaraz J, Oliver A, Sánchez JM. Am J Case Rep. 2015 Jul 17;16:469-72. doi: 10.12659/AJCR.893805.

BACKGROUND: The use of platelet-rich plasma is a now a common medical technique known as regenerative medicine, through power cell activation and differentiation, which produces growth factors called platelets derived both locally and systematically. Here, we report the case of a cerebral palsy patient who received intravenous platelet-rich plasma.

CASE REPORT: We administered an intravenous injection of concentrated platelet-rich plasma (25 cc) in a 6-year-old boy with perinatal cerebral palsy, cognitive impairment, and marked and severe generalized spasticity. We performed follow-up at 3 and 6 months after the injection. All serum samples for determination were obtained by ELISA technique. Cognitive scales (Bayley, Battelle, M.S.C.A, Kaufman ABC, and Stanford-Binet Intelligence scale) were used before and after treatment. The determination protocol that was applied before the analysis was performed manually and the autotransfusion was considered suitable for treatment. We determined the plasma levels of factor similar to insulin-1 (IGF-1), platelet-derived growth factor (PDGF), vasculo-endothelial growth factor (VEGF), and transforming growth factor B (TGF-B) before and during treatment monitoring.

CONCLUSIONS: No adverse effects were observed in the patient except for a small hematoma in the area channeling venous access. We observed a clear improvement in the cognitive sphere (memory, ability to perform more complex tasks, and acquisition of new skills) and in language, maintaining stable levels of growth factor in plasma 3-5 times higher than average for his age group at both 3- and 6-month follow-up. Positron emission tomography (PET) images showed an evident increased demarcation in the cerebral cortex. We propose that this therapy is useful in these patients to harness the neurostimulative and neuroregenerative power of endogenous growth factors derived from platelets.

#### Free PMC Article

PMCID: PMC4509427 PMID: 26185982 [PubMed - indexed for MEDLINE]

#### **Autres methodes**

Dog-assisted therapies and activities in rehabilitation of children with cerebral palsy and physical and mental disabilities.

Elmacı DT, Cevizci S Int J Environ Res Public Health. 2015 May 12;12(5):5046-60. doi:10.3390/ijerph120505046.

The aim of the present study was to evaluate dog-assisted therapies and activities in the rehabilitation of children with cerebral palsy and physical and mental disabilities who have difficulties in benefiting from well-being and Science Infos Paralysie Cérébrale, mai 2016, FONDATION MOTRICE 67 rue Vergniaud 75013 Paris - tel +33 1 40 45 54 03 03 contact: Christine Doumergue cdoumergue@lafondationmotrice.org

health-improving services. This descriptive-explanatory study was conducted in disabled children of various ages between 2008 and 2011 by an experienced team in a private training and rehabilitation center in Antalya (Turkey). In this study, five study groups were formed among the children with physical and mental disabilities. During the therapy studies, three dogs were used. For each therapy group, the goals for the children and therapist were defined, and the activities were determined according to these goals. The entire study process was followed using audio-records and photographs of patients. The expected targets were reached in all study groups. The children who experienced fear, anxiety and difficulties due to their disabilities in daily life learned to cope with their anxieties and fears, set goals and make plans to achieve their aims. During this study, the children improved their abilities to use their bodies according to their capabilities. Accordingly, they improved their ability to develop empathy between themselves and a therapy dog, to receive and present help, and to communicate. The results of the present study revealed that dog-assisted therapies and activities can be a supportive method for routine treatment procedures in the rehabilitation of children with cerebral palsy and physical and mental disabilities.

#### Free PMC Article

PMCID: PMC4454953 PMID: 25985307 [PubMed - indexed for MEDLINE]

## Effects of Hippotherapy on Psychosocial Aspects in Children With Cerebral Palsy and Their Caregivers: A Pilot Study.

Ann Rehabil Med. 2016 Apr;40(2):230-6. doi: 10.5535/arm.2016.40.2.230. Epub 2016 Apr 25. Jang CH, Joo MC, Noh SE, Lee SY, Lee DB, Lee SH, Kim HK, Park HI

OBJECTIVE: To investigate the effects of hippotherapy on psychosocial and emotional parameters in children with cerebral palsy (CP) and their caregivers.

METHODS: Eight children with CP were recruited (three males and five females; mean age, 7.3 years; Gross Motor Function Classification System levels 1-3). Hippotherapy sessions were conducted for 30 minutes once weekly for 10 consecutive weeks in an indoor riding arena. The Gross Motor Function Measure (GMFM), Pediatric Balance Scale (PBS), and the Korean version of the Modified Barthel Index were evaluated. All children were evaluated by the Children's Depression Inventory, Trait Anxiety Inventory for Children, State Anxiety Inventory for Children, Rosenberg Self Esteem Scale, and the Korean-Satisfaction with Life Scale (K-SWLS). Their caregivers were evaluated with the Beck Depression Inventory, the Beck Anxiety Inventory, and the K-SWLS. We assessed children and their caregivers with the same parameters immediately after hippotherapy.

RESULTS: Significant improvements on the GMFM, dimension E in the GMFM, and the PBS were observed after hippotherapy compared with the baseline assessment (p<0.05). However, no improvements were detected in the psychosocial or emotional parameters in children with CP or their caregivers. None of the participants showed any adverse effects or accidents during the 10 weeks hippotherapy program.

CONCLUSIONS: Hippotherapy was safe and effectively improved gross motor and balance domains in children with CP. However, no improvements were observed in psychosocial or emotional parameters.

Free PMC Article

PMCID: PMC4855116 PMID: 27152272 [PubMed]

## Horseback riding therapy in addition to conventional rehabilitation program decreases spasticity in children with cerebral palsy: A small sample study.

Alemdaroğlu E, Yanıkoğlu İ, Öken Ö, Uçan H, Ersöz M, Köseoğlu BF, Kapıcıoğlu Mİ Complement Ther Clin Pract. 2016 May;23:26-9. doi: 10.1016/j.ctcp.2016.02.002. Epub 2016 Feb 13.

OBJECTIVE: To evaluate the short-term effects of horseback riding therapy in addition to a conventional rehabilitation program in children with cerebral palsy.

METHODS: Nine children receiving horseback riding therapy in addition to conventional rehabilitation (Group 1) and seven children receiving conventional rehabilitation alone (Group 2) were assessed at baseline and 5 weeks later.

Assessed were: modified functional reach test (MFRT), hip abduction angle, the Ashworth Scale for hip adductor muscle spasticity, knee distance test, and the Gross Motor Function Classification System (GMFCS).

RESULTS: The percentage change in hip adductor spasticity on the Ashworth Scale was 22% in Group 1 and 0% in Group 2 (significant difference; p = 0.016). Comparison of changes on the MFRT, GMFCS, knee distance test and hip abduction angle showed that the differences between Groups 1 and 2 were not significant.

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CONCLUSIONS: In these children, horseback riding therapy in addition to conventional rehabilitation resulted in significant improvement in adductor spasticity on short-term follow-up. Copyright © 2016 Elsevier Ltd. All rights reserved. PMID: 27157954 [PubMed - in process]

#### Laser acupuncture as an adjunctive therapy for spastic cerebral palsy in children.

Dabbous OA, Mostafa YM, El Noamany HA, El Shennawy SA, El Bagoury MA Lasers Med Sci. 2016 May 4. [Epub ahead of print]

Laser acupuncture is widely used as an alternative line of treatment in several chronic pediatric diseases. To investigate whether biostimulation by low-level laser on acupuncture points adds a clinical benefit to conventional physiotherapy in hemiplegic spastic cerebral palsy (CP) children. Forty spastic hemiplegic cerebral palsy children by age 1-4 years were chosen from the pediatric outpatient clinic of the National Institute of Laser Enhanced Sciences (NILES), Cairo University, and Menofyia University hospitals. They were randomly divided into control and study groups; 20 children each. Both groups received physiotherapy for 3 months, while only the study group also received laser acupuncture (low-level laser 650 nm with 50 mW power was applied at each acupoint for 30 s giving an energy density of 1.8 J/cm(2)). Preassessment and postassessment of muscle tone, the range of motion (ROM), and gross motor function measurements (GMFMs) were obtained, and the results were statistically analyzed. Comparison between posttreatment measures for the control vs. Study group, showed significant difference in muscle tone (wrist flexors and plantar flexors) in favor of the study group, while range of motion showed no significant difference in total score while there was a significant difference in goal total score (sum of % scores for each dimension identified as goal area divided by number of goal areas) in favor of the study group. Laser acupuncture has a beneficial effect on reducing spasticity in spastic cerebral palsy and may be helpful in improving their movement.

PMID: 27147077 [PubMed - as supplied by publisher]

## Langage -parole-communication

#### Mastery motivation in children with complex communication needs: longitudinal data analysis.

Medeiros KF, Cress CJ, Lambert MC

Augment Altern Commun. 2016 May 17:1-11. [Epub ahead of print]

This study compared longitudinal changes in mastery motivation during parent-child free play for 37 children with complex communication needs. Mastery motivation manifests as a willingness to work hard at tasks that are challenging, which is an important quality to overcoming the challenges involved in successful expressive communication using AAC. Unprompted parent-child play episodes were identified in three assessment sessions over an 18-month period and coded for nine categories of mastery motivation in social and object play. All of the object-oriented mastery motivation categories and one social mastery motivation category showed an influence of motor skills after controlling for receptive language. Object play elicited significantly more of all of the object-focused mastery motivation categories than social play, and social play elicited more of one type of social-focused mastery motivation behavior than object play. Mastery motivation variables did not differ significantly over time for children. Potential physical and interpersonal influences on mastery motivation for parents and children with complex communication needs are discussed, including broadening the procedures and definitions of mastery motivation beyond object-oriented measurements for children with complex communication needs. PMID: 27184193 [PubMed - as supplied by publisher]

## Sommeil

#### Children with cerebral palsy: why are they awake at night? A pilot study.

J Spec Pediatr Nurs. 2015 Apr;20(2):98-104. doi: 10.1111/jspn.12106. Epub 2015 Jan 29. Petersen S, Harvey A, Reddihough D, Newall F.

PURPOSE: The purpose of this study was to assess acceptability of study design and utility of assessment tools for determining causes of sleep disturbance in children with cerebral palsy (CP) and their caregivers.

Science Infos Paralysie Cérébrale, mai 2016, FONDATION MOTRICE 67 rue Vergniaud 75013 Paris - tel +33 1 42 45 54 03 03 contact: Christine Doumergue cdoumergue@lafondationmotrice.org

DESIGN AND METHODS: This pilot study explored parental report (n = 8) of the child's sleep using a validated sleep questionnaire, a survey, and a time use diary.

RESULTS: The selected tools were appropriate. Study design provided challenges for recruitment. Future research should include qualitative data.

PRACTICE IMPLICATIONS: Although further research is required to confirm results, sleep problems for children with CP and resulting sleep disturbances for caregivers may be an important part of clinical assessment.

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PMID: 25630812 [PubMed - indexed for MEDLINE]

## Autres Troubles /troubles concomitants

## **4**Troubles musculosquelettiques, des tissue conjonctifs et osseux

#### Osteopenic features of the hip joint in patients with cerebral palsy: a hospital-based study.

Moon SY, Kwon SS, Cho BC, Chung CY, Lee KM, Sung KH, Chung MK, Zulkarnain A, Kim YS, Park MS Dev Med Child Neurol. 2016 May 4. doi: 10.1111/dmcn.13146. [Epub ahead of print]

AIM: We aimed to evaluate the bone mineral density of the hip joint in patients with cerebral palsy (CP).

METHOD: Patients with CP younger than 18 years who underwent three-dimensional hip examination by computed tomography were analysed. Bone attenuation of the acetabulum and femur was measured as Hounsfield units (HU), and was adjusted for affecting factors such as hip instability and Gross Motor Function Classification System (GMFCS).

RESULTS: One hundred and twenty-six patients with CP and 86 typically developing participants were included. The average bone attenuation was significantly lower in those with CP than in the comparison group (acetabulum: 70.8HU, 95% confidence interval [95% CI] 59.9-81.8; femur: 82.2HU, 95% CI 70.4-95.8). Compared with GMFCS levels I to III, bone attenuation was significantly lower for GMFCS levels IV (acetabulum: 30.9HU, 95% CI 15.7-46.2; femur: 39.7HU, 95% CI 19.9-59.5) and V (acetabulum: 51.7HU, 95% CI 35.9-67.5; femur: 72.5HU, 95% CI 51.9-93.0). The average bone attenuation decreased when the migration percentage was over 37% (acetabulum: 11.6HU, 95% CI 1.4-24.6; femur: 26.8HU, 95% CI 9.9-43.6).

INTERPRETATION: Bone attenuation of the acetabulum and femur was significantly affected both by GMFCS level and by severity of hip instability.

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PMID: 27145375 [PubMed - as supplied by publisher]

# Site-Specific Bone Mineral Density Is Unaltered Despite Differences in Fat-Free Soft Tissue Mass Between Affected and Nonaffected Sides in Hemiplegic Paralympic Athletes with Cerebral Palsy: Preliminary Findings.

Runciman P, Tucker R, Ferreira S, Albertus-Kajee Y, Micklesfield L, Derman W. Am J Phys Med Rehabil. 2016 May 4. [Epub ahead of print]

OBJECTIVE: This study investigated bone mineral density (BMD, g/cm), fat mass (FM, kg), and fat-free soft tissue mass (FFSTM, kg) in Paralympic athletes with cerebral palsy (CP) using dual-energy x-ray absorptiometry.

METHODS: Bone mineral density, BMD Z scores (standard deviations), FM, and FFSTM were measured for the whole body and at the lumbar spine, femoral neck, and total hip sites on both nonaffected and affected sides of 6 athletes with hemiplegic CP.

RESULTS: There were no differences between nonaffected and affected sides with respect to site-specific BMD and BMD Z scores and FM. Fat-free soft tissue mass was significantly lower on the affected side in both upper and lower limbs (15% lower; P < 0.05).

CONCLUSION: The present study is the first to describe similar BMD between sides, symmetry in FM, and asymmetry in FFSTM in Paralympic athletes with CP. These findings have important consequences for rehabilitation, as they indicate the potential for positive physiological adaptation as a result of exercise training over long periods of time. PMID: 27149600 [PubMed - as supplied by publisher]

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#### The effect of anticonvulsant use on bone mineral density in non-ambulatory children with cerebral palsy.

Cheng SW, Ko CH, Lee CY

Hong Kong Med J. 2016 May 6. doi: 10.12809/hkmj154588. [Epub ahead of print]

INTRODUCTION: Studies showed that use of anticonvulsants (antiepileptic drugs) might be associated with reduced bone mineral density. The primary objective of this study was to evaluate the effect of anticonvulsants on bone mineral density in non-ambulatory children with cerebral palsy. The secondary objective was to identify their risk factors for low bone mineral density.

METHODS: This case series with internal comparisons was conducted in a paediatric residential rehabilitation centre in Hong Kong. Overall, 32 patients were enrolled. The study group comprised 18 patients (6 males, 12 females) aged 5.0 to 19.5 years (mean  $\pm$  standard deviation, 13.8  $\pm$  4.7 years); all were prescribed anticonvulsant therapy for more than 2 years. The comparison group comprised 14 patients (6 males, 8 females) aged 7.0 to 19.1 years (mean, 16.4  $\pm$ 3.0 years) who were concomitant non-ambulatory residents with cerebral palsy and were not prescribed any anticonvulsant therapy prior to study recruitment. Patients underwent a physical examination, blood tests, nutritional assessment, and dual-energy X-ray absorptiometry scan of the total body less head. Z-scores were calculated.

RESULTS: There was no significant difference in Z-scores of total body less head between groups. Among children with low bone mineral density (Z-scores  $\leq$ -2.0) and normal bone mineral density, multivariate analysis revealed that higher weight-for-age Z-score (adjusted odds ratio=0.015) and presence of puberty (adjusted odds ratio=0.027) were independent factors for bone mineral density improvement. Hosmer-Lemeshow goodness of fit test (P=0.315) was not significant. Nagelkerke R<sup>2</sup> was 0.677, signifying a relatively well-fitting model.

CONCLUSION: There was no evidence that anticonvulsant therapy has any detrimental effect on bone mineral density in non-ambulatory children with cerebral palsy. A low weight-for-age Z-score was associated with low bone mineral density. Early nutritional intervention to optimise body weight may help to increase bone mineral density. **Free Article** 

PMID: 27149974 [PubMed - as supplied by publisher]

## **4**Métabolisme

# The Effect of Handcycle Ergometer Exercise on Glucose Tolerance in Ambulatory and Non-Ambulatory Adolescents.

Short KR, Teague AM, Klein JC, Malm-Buatsi E, Frimberger D. *Pediatr Exerc Sci. 2016 May 13. [Epub ahead of print]* 

PURPOSE: Whole body or leg exercise before a meal can increase insulin sensitivity, but it is unclear whether the same can occur with upper body exercise since a smaller muscle mass is activated. We measured the impact of a single session of handcycle exercise on glucose tolerance and insulin sensitivity.

METHODS: Non-ambulatory (Non-Amb) adolescents with spina bifida or cerebral palsy (4F/3M), or ambulatory peers (Control, 4F/7M) completed two glucose tolerance tests on separate days, preceded by either rest or a 35-minute bout of moderate-to-vigorous intermittent handcycle exercise.

RESULTS: The Non-Amb group had higher body fat (mean  $\pm$  SD: 38 $\pm$ 12%, Control: 24 $\pm$ 9, p=0.041) but similar VO2peak (17.7 $\pm$ 6.1 ml/kg/min, Control: 21.1 $\pm$ 7.9). Fasting glucose and insulin were normal for all participants. Compared to the rest trial, exercise resulted in a reduction in glucose area under the curve (11%, p = 0.008) without a significant group x trial interaction and no difference in the magnitude of change between groups. Insulin sensitivity was increased 16% (p=0.028) by exercise in the Control group but was not significantly changed in the Non-Amb group.

CONCLUSION: A single bout of handcycle exercise improves glucose tolerance in adolescents with and without mobility limitations and could therefore help maintain or improve metabolic health.

PMID: 27176627 [PubMed - as supplied by publisher]

## **H**Troubles respiratoires

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#### Factors Associated with Respiratory Illness in Children and Young Adults with Cerebral Palsy.

Blackmore AM, Bear N, Blair E, Gibson N, Jalla C, Langdon K, Moshovis L, Steer K, Wilson AC J Pediatr. 2016 Jan;168:151-7.e1. doi: 10.1016/j.jpeds.2015.09.064. Epub 2015 Oct 29.

OBJECTIVE: To describe associations between respiratory illness and its potential predictors in children and young adults with cerebral palsy (CP).

STUDY DESIGN: Cross-sectional survey of self- and caregiver-reported respiratory symptoms for individuals aged up to 26 years with CP. Respiratory illness was indicated by 2 outcomes: (1)  $\geq$ 1 respiratory hospitalizations in the past year; and (2)  $\geq$ 2 courses of antibiotics for respiratory symptoms in the past year. ORs were calculated using univariate and multivariate logistic regression.

RESULTS: There were 551 participants, aged 1-26 years, distributed across all gross motor function classification scale (GMFCS) levels. In univariate analyses, factors significantly associated with respiratory hospitalizations were weekly respiratory symptoms (OR 2.31, 95% CI 1.78-3.00), respiratory symptoms during meals (OR 3.23, 95% CI 1.50-5.80), gastroesophageal reflux (OR 3.01, 95% CI 1.71-5.31), coughing or choking on saliva (OR 4.36, 95% CI 2.38-8.01), current asthma (OR 3.56, 95% CI 1.97-6.42), age (0-3 years) (OR 3.24, 95% CI 1.19-8.80, compared with 13-17 years), seizures (OR 3.45, 95% CI 1.96-6.08), and scoliosis (OR 2.14, 95% CI 1.16-3.97). Nonambulatory individuals (GMFCS IV-V) were at significantly increased risk of hospitalizations only if they had food modifications and/or nasogastric or gastrostomy tube feeds (OR 5.36, 95% CI 2.89-9.96, compared with GMFCS I-III with no food modifications and no tube). All factors, except seizures and scoliosis, were significantly associated with multiple courses of antibiotics in univariate analyses.

CONCLUSIONS: Oromotor dysfunction is strongly associated with respiratory illness in patients with CP.

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PMID: 26520916 [PubMed - indexed for MEDLINE]

#### Tracheostomy in neurologically compromised paediatric patients: role of starplasty.

Gupta A, Stokken J, Krakovitz P, Malhotra P, Anne S J Laryngol Otol. 2015 Oct;129(10):1009-12. doi: 10.1017/S002221511500208X. Epub 2015 Aug 17.

OBJECTIVES: Starplasty tracheostomy is an alternative to traditional tracheostomy. This paper reviews neurologically compromised paediatric patients with tracheostomies and discusses the role of starplasty tracheostomy.

METHOD: A retrospective review was conducted of paediatric patients with a neurological disorder who underwent tracheostomy between 1997 and 2011.

RESULTS: Forty-eight patients, with an average age of 7.3 years, were identified. The most common indications for tracheostomy were: ventilator dependence (39.6 per cent), an inability to tolerate secretions or recurrent aspiration pneumonia (33.3 per cent), and upper respiratory obstruction or hypotonia (12.5 per cent). The most common underlying neurological diagnosis was cerebral palsy. There were no early complications. Eighteen (43 per cent) of 42 patients with follow up experienced at least 1 delayed complication. Only 12 patients (28.6 per cent) were decannulated.

CONCLUSION: Patients with primary neurological diagnoses have low rates of decannulation; starplasty tracheostomy should be considered for these patients. Patients with seizure disorder or acute neurological injury tended to have a higher short-term decannulation rate; traditional tracheostomy is recommended in these patients. PMID: 26279145 [PubMed - indexed for MEDLINE]

### **Wutrition – Troubles nutritionnels**

#### Body mass index in ambulatory children with cerebral palsy: A cohort study.

Pascoe J, Thomason P, Graham HK, Reddihough D,Sabin MA J Paediatr Child Health. 2016 Apr;52(4):417-21. doi: 10.1111/jpc.13097.

AIM: Children with cerebral palsy (CP) have reduced levels of physical activity compared with children without physical disability and experience risk factors for becoming overweight or obese. In the Australian CP population, there is little information available about the weight status of children with CP. The aims of this study were to

compare the distribution of body mass index (BMI) in a cohort of ambulant children with CP with the BMI distribution of Australian children and explore the relationship between BMI and gross motor function.

METHODS: A retrospective cohort study of 587 children with CP Gross Motor Function Classification System (GMFCS) levels I-III who attended a Gait Laboratory between July 1995 and January 2012 was carried out. The BMI and Z-score were calculated at each assessment. Data were grouped into the categories of underweight, healthy, overweight and obese according to age-specific and sex-specific percentiles.

RESULTS: There were 348 boys and 240 girls with a mean age 11.2 (standard deviation 3.2) years. Mean BMI Z-score was 0.11 (standard deviation 1.33). Seven percent of children were underweight, 73.6% healthy, 7.3% overweight and 12.1% obese. This was similar to the distribution of children without disability. The largest percentage of children in the healthy group were classified GMFCS I. The largest percentage of children in the obese group were classified GMFCS III.

CONCLUSIONS: In this cohort, 19.4% of ambulant children with CP were overweight or obese. This is of concern as BMI may impact on the outcomes of surgical intervention and rehabilitation. Further research is needed to determine the consequences of obesity for children with CP.

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and Child Health Division (Royal Australasian College of Physicians).

PMID: 27145505 [PubMed - in process]

#### Effects of maternal low-protein diet on parameters of locomotor activity in a rat model of cerebral palsy.

Silva KO, Pereira SD, Portovedo M, Milanski M, Galindo LC, Guzmán-Quevedo O, Manhães-De-Castro R, Toscano AE Int J Dev Neurosci. 2016 May 19. pii: S0736-5748(16)30079-X. doi: 10.1016/j.ijdevneu.2016.05.002. [Epub ahead of print]

Children with cerebral palsy have feeding difficulties that can contribute to undernutrition. The aim of this study was to investigate the effect of early undernutrition on locomotor activity and the expression of the myofibrillar protein MuRF-1 in an experimental model of cerebral palsy (CP). In order to achieve this aim, pregnant rats were divided into two groups according to the diet provided: Normal Protein (NP, n=9) and Low Protein (LP, n=12) groups. After birth, the pups were divided into four groups: Normal Protein Sham (NPS, n=16), Normal Protein Cerebral Palsy (NPCP, n=21), Low Protein Sham (LPS, n=20) and Low Protein Cerebral Palsy (LPCP, n=18) groups. The experimental cerebral palsy protocol consisted of two episodes of anoxia at birth and during the first days of life. Each day, nitrogen flow was used (9I/min during 12min). After nitrogen exposure, sensorimotor restriction was performed 16h per day, from the 2nd to the 28th postnatal day (PND). Locomotor activity was evaluated at 8th, 14th, 17th, 21th and 28th PND. At PND 29, soleus muscles were collected to analyse myofibrillar protein MuRF-1. Our results show that CP animals decreased body weight (p<0.001), which were associated with alterations of various parameters of locomotor activity (p<0.05), compared to their control. Undernourished animals also showed a decrease (p<0.05) in body weight and locomotor activity parameters. Moreover, CP decreased MuRF-1 levels in nourished rats (p=0.015) but not in undernourished rats. In summary, perinatal undernutrition exacerbated the negative effects of cerebral palsy on locomotor activity and muscle atrophy, but it appears not be mediated by changes in MuRF-1 levels. Copyright © 2016 ISDN. Published by Elsevier Ltd. All rights reserved.

PMID: 27211347 [PubMed - as supplied by publisher]

# Effects of Neuromuscular Electrical Stimulation on the Masticatory Muscles and Physiologic Sleep Variables in Adults with Cerebral Palsy: A Novel Therapeutic Approach.

Giannasi LC, Matsui MY, Freitas SR, Caldas BF, Grossmann E, Amorim JB, dos Santos Idos R, Oliveira LV, Oliveira CS, Gomes MF

PLoS One. 2015 Aug 6;10(8):e0128959. doi: 10.1371/journal.pone.0128959. eCollection 2015.

Cerebral palsy (CP) is a term employed to define a group of non-progressive neuromotor disorders caused by damage to the immature or developing brain, with consequent limitations regarding movement and posture. CP may impair orapharygeal muscle tone, leading to a compromised chewing function and to sleep disorders (such as obstructive sleep apnea). Thirteen adults with CP underwent bilateral masseter and temporalis neuromuscular electrical stimulation (NMES) therapy. The effects on the masticatory muscles and sleep variables were evaluated using electromyography (EMG) and polysomnography (PSG), respectively, prior and after 2 months of NMES. EMG consisted of 3 tests in different positions: rest, mouth opening and maximum clenching effort (MCE). EMG values in the rest position were 100% higher than values recorded prior to therapy for all muscles analyzed (p < 0.05); mean Science Infos Paralysie Cérébrale, mai 2016, FONDATION MOTRICE 67 rue Vergniaud 75013 Paris - tel +33 1 46 45 54 03 03 contact: Christine Doumergue cdoumergue@lafondationmotrice.org

mouth opening increased from  $38.0 \pm 8.0$  to  $44.0 \pm 10.0$  cm (p = 0.03). A significant difference in MCE was found only for the right masseter. PSG revealed an improved in the AHI from  $7.2\pm7.0/h$  to  $2.3\pm1.5/h$  (p < 0.05); total sleep time improved from 185 min to 250 min (p = 0.04) and minimun SaO2 improved from  $83.6 \pm 3.0$  to  $86.4 \pm 4.0$  (p = 0.04). NMES performed over a two-month period led to improvements in the electrical activity of the masticatory muscles at rest, mouth opening, isometric contraction and sleep variables, including the elimination of obstructive sleep apnea events in patients with CP. Trial registration: ReBEC RBR994XFS http://www.ensaiosclinicos.gov.br. **Free PMC Article** 

PMCID: PMC4527754 PMID: 26247208 [PubMed - indexed for MEDLINE]

#### Nutritional and health benefits of semi-elemental diets: A comprehensive summary of the literature.

Alexander DD, Bylsma LC, Elkayam L, Nguyen DL World J Gastrointest Pharmacol Ther. 2016 May 6;7(2):306-19. doi: 10.4292/wjgpt.v7.i2.306.

AIM: To critically review and summarize the literature on nutritional and health outcomes of semi-elemental formulations on various nutritionally vulnerable patient populations who are unable to achieve adequate nutrition from standard oral diets.

METHODS: We conducted a comprehensive literature search of Pubmed and Embase databases. We manually screened articles that examined nutritional and health outcomes (e.g., growth, disease activity, gastrointestinal impairment, mortality, and economic impact) among various patient groups receiving semi-elemental diets. This review focused on full-text articles of randomized controlled clinical trials and other intervention studies, but pertinent abstracts and case studies were also included. Results pertaining primarily to tolerance, digestion, and absorption were summarized for each patient population in this systematic review.

RESULTS: Results pertaining primarily to tolerance, digestion, and absorption were summarized for each patient population. The efficacy of semi-elemental whey hydrolyzed protein (WHP) diet have been reported in various nutritionally high risk patient populations including - Crohn's disease, short bowel syndrome, acute and chronic pancreatitis, cerebral palsy, cystic fibrosis, cerebrovascular accidents, human immunodeficiency virus, critically ill, and geriatrics. Collectively, the evidence from the medical literature indicates that feeding with a semi-elemental diet performs as well or better than parenteral or amino acid based diets in terms of tolerance, digestion, and nutrient assimilation measures across various disease conditions.

CONCLUSION: Based on this comprehensive review of the literature, patient populations who have difficulty digesting or absorbing standard diets may be able to achieve improved health and nutritional outcomes through the use of semi-elemental WHP diets.

#### Free PMC Article

PMCID: PMC4848254 PMID: 27158547 [PubMed]

## **4**Troubles digestifs

## Medical and surgical management of neurogenic bowel.

Gor RA, Katorski JR, Elliott SP. *Curr Opin Urol. 2016 May 5. [Epub ahead of print]* 

PURPOSE OF REVIEW: Neurogenic bowel dysfunction (NBoD) commonly affects patients with spina bifida, cerebral palsy, and spinal cord injury among other neurologic insults. NBoD is a significant source of physical and psychosocial morbidity. Treating NBoD requires a diligent relationship between patient, caretaker, and provider in establishing and maintaining a successful bowel program. A well designed bowel program allows for regular, predictable bowel movements and prevents episodes of fecal incontinence.

RECENT FINDINGS: Treatment options for NBoD span conservative lifestyle changes to fecal diversion depending on the nature of the dysfunction. Lifestyle changes and oral laxatives are effective for many patients. Patients requiring more advanced therapy progress to transanal irrigation devices and retrograde enemas. Those receiving enemas may opt for antegrade enema administration via a Malone antegrade continence enema or Chait cecostomy button,

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which are increasingly performed in a minimally invasive fashion. Select patients benefit from fecal diversion, which simplifies care in more severe cases.

SUMMARY: Many medical and surgical options are available for patients with NBoD. Selecting the appropriate medical or surgical treatment involves a careful evaluation of each patient's physical, psychosocial, financial, and geographic variables in an effort to optimize bowel function.

PMID: 27152922 [PubMed - as supplied by publisher]

## **4**Sphère bucco-dentaire – Troubles de la déglutition

#### A telehealth approach to conducting clinical swallowing evaluations in children with cerebral palsy.

Kantarcigil C, Sheppard JJ, Gordon AM, Friel KM, Malandraki GA. Res Dev Disabil. 2016 Apr 28;55:207-217. doi: 10.1016/j.ridd.2016.04.008. [Epub ahead of print]

BACKGROUND: Accurate and timely evaluation of dysphagia in children with cerebral palsy (CP) is critical. For children with limited access to quality healthcare, telehealth is an option; however, its reliability needs to be investigated.

AIM: To test the reliability of an asynchronous telehealth model for evaluating dysphagia in children with CP using a standardized clinical assessment.

METHODS AND PROCEDURES: Nineteen children (age range 6.9-17.5) were assessed at three mealtimes via the Dysphagia Disorder Survey (DDS) by three clinicians (face-to-face evaluations). Mealtimes were video-recorded to allow asynchronous evaluations by a remote clinician who also completed approximately 1/3 of face-to-face evaluations. Agreement was tested on DDS variables and dysphagia severity.

OUTCOMES AND RESULTS: Results revealed substantial to excellent agreement between face-to-face and remote assessments by the same rater (78-100%, KW=0.64-1) on all, but two variables (oral transport and oral pharyngeal swallow) and by different raters (69-89%, KW=0.6-0.86) on all but one variable (orienting). For dysphagia severity, intrarater agreement was excellent (100%, KW=1); interrater agreement was substantial (85%; KW=0.76).

CONCLUSIONS AND IMPLICATIONS: Asynchronous clinical swallowing evaluations using standardized tools have acceptable levels of agreement with face-to-face evaluations, and can be an alternative for children with limited access to expert swallowing care.

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PMID: 27132060 [PubMed - as supplied by publisher]

# [Submandibular gland resection for the management of sialorrhea in paediatric patients with cerebral palsy and unresponsive to type A botullinum toxin. Pilot study].[Article in Spanish]

Hernández-Palestina MS, Cisneros-Lesser JC, Arellano-Saldaña ME, Plascencia-Nieto SE.

*Cir Cir.* 2016 May 21. *pii*: S0009-7411(16)30019-6. *doi*: 10.1016/j.circir.2016.03.007. [Epub ahead of print

BACKGROUND: Sialorrhoea has a prevalence of between 10% and 58% in patients with cerebral palsy. Amongst the invasive treatments, botulinum toxin-A injections in submandibular and parotid glands and various surgical techniques are worth mentioning. There are no studies in Mexico on the usefulness of surgery to manage sialorrhoea.

OBJECTIVE: To evaluate the usefulness of submandibular gland resection in improving sialorrhoea in patients with cerebral palsy and with a poor response to botulinum toxin.

MATERIAL AND METHODS: Experimental, clinical, self-controlled, prospective trial was conducted to evaluate the grade of sialorrhoea before surgery, and 8, 16 and 24 weeks after. Statistical analysis was performed using a non-parametric repetitive measure assessment, considering a p < 0.05 as significant. Complications and changes in salivary composition were evaluated.

RESULTS: Surgery was performed on 3 patients with severe sialorrhoea, and 2 with profuse sialorrhoea, with mean age of 10.8 years. The frequency and severity of sialorrhoea improved in the 5 patients, with mean of 76.7 and 87.5% improvement, respectively. The best results were seen after 6 months of surgery, with a statistically significant difference between the preoperative stage and 6 months after the procedure (p = 0.0039, 95% CI). No significant differences were observed in complications, increase in periodontal disease or cavities, or salivary composition.

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CONCLUSIONS: Submandibular gland resection is an effective technique for sialorrhoea control in paediatric patients with cerebral palsy, with a reduction in salivary flow greater than 80%. It has a low chance of producing complications compared to other techniques. It led to an obvious decrease in sialorrhoea without the need to involve other salivary glands in the procedure.

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PMID: 27221328 [PubMed - as supplied by publisher]

## **4**Troubles urinaires

#### Urodynamic Findings in Adults with Moderate to Severe Cerebral Palsy.

Cotter KJ, Levy ME, Goldfarb RA, Liberman D, Katorski J, Myers JB, Elliott SP. Urology. 2016 May 19. pii: S0090-4295(16)30220-5. doi:10.1016/j.urology.2016.05.024. [Epub ahead of print]

OBJECTIVES: To determine UDS findings in adult CP patients. Cerebral palsy (CP) patients may suffer from voiding dysfunction. Urodynamics (UDS) in children with CP has consistently shown an upper motor neuron bladder with detrusor-sphincter dyssynergia (DSD).

METHODS: We included adult CP patients seen at Gillette Transitional Urology Clinic who underwent UDS for voiding dysfunction between 2011-2014.Descriptive statistics were used to characterize findings.

RESULTS: 49/211 CP patients underwent UDS. Average age was 30 years; 55% were men. 98% had moderate to severe CP. UDS was initiated for irritative symptoms in 55%, obstructive voiding symptoms in 25%, hydronephrosis in 18%, and other reasons in 2%. Incontinence was reported in 57%. DSD was seen in 12%, detrusor overactivity (DO) in 30% and detrusor leak point pressure (DLPP) >40 cm H2O in 51%. Median compliance was 18 mL/cm H2O (0.78 - 365). Maximum cystometric capacity (MCC) was 80-1400 mL and was < 300mL in 27%. 16% had a MCC < 300mL and compliance < 20. 12% had a MCC <300 and DLPP >40.

CONCLUSIONS: UDS findings in symptomatic adult CP patients are varied. 51% had upper motor neuron bladder findings, similar to that seen in the pediatric literature, but 6% had large flaccid bladders. Half of the patients had concerning findings, such as compliance <20 or DLPP >40 cm H2O. Our results emphasize the needs to thoroughly investigate voiding dysfunction in those with CP. Further characterization of this population is needed in order to correlate these UDS findings with clinical outcomes.

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PMID: 27210572 [PubMed - as supplied by publisher]

## **4**Troubles de la vision

#### Ophthalmic profile and systemic features of pediatric facial nerve palsy.

Patil-Chhablani P, Murthy S, Swaminathan M. *Eye Sci. 2015 Dec;30(4):147-50.* 

BACKGROUND: Facial nerve palsy (FNP) occurs less frequently in children as compared to adults but most cases are secondary to an identifiable cause. These children may have a variety of ocular and systemic features associated with the palsy and need detailed ophthalmic and systemic evaluation.

METHODS: This was a retrospective chart review of all the cases of FNP below the age of 16 years, presenting to a tertiary ophthalmic hospital over the period of 9 years, from January 2000 to December 2008.

RESULTS: A total of 22 patients were included in the study. The average age at presentation was 6.08 years (range, 4 months to 16 years). Only one patient (4.54%) had bilateral FNP and 21 cases (95.45%) had unilateral FNP. Seventeen patients (77.27%) had congenital palsy and of these, five patients had a syndromic association, three had birth trauma and nine patients had idiopathic palsy. Five patients (22.72%) had an acquired palsy, of these, two had a traumatic cause and one patient each had neoplastic origin of the palsy, iatrogenic palsy after surgery for hemangioma and idiopathic palsy. Three patients had ipsilateral sixth nerve palsy, two children were diagnosed to have Moebius syndrome, one child had an ipsilateral Duane's syndrome with ipsilateral hearing loss. Corneal involvement was seen in eight patients (36.36%). Amblyopia was seen in ten patients (45.45%). Neuroimaging

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studies showed evidence of trauma, posterior fossa cysts, pontine gliosis and neoplasms such as a chloroma. Systemic associations included hemifacial macrosomia, oculovertebral malformations, Dandy Walker syndrome, Moebius syndrome and cerebral palsy

CONCLUSIONS: FNP in children can have a number of underlying causes, some of which may be life threatening. It can also result in serious ocular complications including corneal perforation and severe amblyopia. These children require a multifaceted approach to their care.

PMID: 27215002 [PubMed - in process]

## **H**Troubles de l'audition

## Comparison of Auditory Perception in Cochlear Implanted Children with and without Additional Disabilities.

Hashemi SB Md, Monshizadeh L PhD. Iran J Med Sci. 2016 May;41(3):186-190.

BACKGROUND: The number of children with cochlear implants who have other difficulties such as attention deficiency and cerebral palsy has increased dramatically. Despite the need for information on the results of cochlear implantation in this group, the available literature is extremely limited. We, therefore, sought to compare the levels of auditory perception in children with cochlear implants with and without additional disabilities.

METHODS: A spondee test comprising 20 two-syllable words was performed. The data analysis was done using SPSS, version 19.

RESULTS: Thirty-one children who had received cochlear implants 2 years previously and were at an average age of 7.5 years were compared via the spondee test. From the 31 children, 15 had one or more additional disabilities. The data analysis indicated that the mean score of auditory perception in this group was approximately 30 scores below that of the children with cochlear implants who had no additional disabilities.

CONCLUSION: Although there was an improvement in the auditory perception of all the children with cochlear implants, there was a noticeable difference in the level of auditory perception between those with and without additional disabilities. Deafness and additional disabilities depended the children on lip reading alongside the auditory ways of communication. In addition, the level of auditory perception in the children with cochlear implants who had more than one additional disability was significantly less than that of the other children with cochlear implants who had one additional disability.

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PMCID: PMC4876296 PMID: 27217602 [PubMed - as supplied by publisher]

## Qualité de vie et rapport au monde

#### Burden of Neurological Conditions in Canada.

Gaskin J, Gomes J, Darshan S, Krewski D Neurotoxicology. 2016 May 3. pii: S0161-813X(16)30066-3. doi: 10.1016/j.neuro.2016.05.001. [Epub ahead of print]

Neurological conditions are among the leading causes of disability in the Canadian population and are associated with a large public health burden. An increase in life expectancy and a declining birth rate has resulted in an aging Canadian population, and the proportion of age-adjusted mortality due to non-communicable diseases has been steadily increasing. These conditions are frequently associated with chronic disability and an increasing burden of care for patients, their families and caregivers. The National Population Health Study of Neurological Conditions (NPHSNC) aims to improve knowledge about neurological conditions and their impacts on individuals, their families, caregivers and health care system. The Systematic Review of Determinants of Neurological Conditions, a specific objective within the NPHSNC, is a compendium of systematic reviews on risk factors affecting onset and progression of the following 14 priority neurological conditions: Alzheimer's disease (AD), amyotrophic lateral sclerosis (ALS), brain tumours (BT), cerebral palsy (CP), dystonia, epilepsy, Huntington's disease (HD), hydrocephalus, multiple sclerosis (MS), muscular dystrophies (MD), neurotrauma, Parkinson's disease (PD), spina bifida (SB), and Tourette's

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syndrome (TS). The burden of neurological disease is expected to increase as the population ages, and this trend is presented in greater detail for Alzheimer's and Parkinson's disease because the incidence of these two common neurological diseases increases significantly with age over 65 years. This article provides an overview of burden of neurological diseases in Canada to set the stage for the in-depth systematic reviews of the 14 priority neurological conditions presented in subsequent articles in this issue.

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PMID: 27153747 [PubMed - as supplied by publisher]

## **Vie quotidienne**

#### Activities and participation of children with cerebral palsy: parent perspectives.

Mei C, Reilly S, Reddihough D, Mensah F, Green J, Pennington L, Morgan AT. Disabil Rehabil. 2015;37(23):2164-73. doi: 10.3109/09638288.2014.999164. Epub 2015 Jan 14.

PURPOSE: To explore parents' views of the activities and participation of children with cerebral palsy (CP) with a range of communicative abilities and the factors (personal and environmental) that influenced these.

METHODS: Thirteen parents of children with CP aged 4-9 years participated in semi-structured individual interviews. Interviews were recorded, transcribed and analysed thematically. Identified codes and themes were mapped to the domains of the International Classification of Functioning, Disability and Health – Children and Youth Version (ICF-CY).

RESULTS: Parents' responses reflected all ICF-CY domains comprising activity, participation and environmental factors. Codes were primarily mapped to the domains learning and applying knowledge, communication, mobility and interpersonal interactions and relationships. Key barriers identified included aspects of parents' own interactions with their child (e.g. not offering choices), unfamiliar people and settings, negative attitudes of others and children's frustration. Facilitators included support received from the child's family and school, being amongst children, having a familiar routine and the child's positive disposition.

CONCLUSIONS: Despite the barriers experienced, children participated in a range of activities. Parents placed importance on communication and its influence on children's independence, behaviour and relationships. Barriers and facilitators identified highlight aspects of the environment that could be modified through intervention to enhance communication and participation.

IMPLICATIONS FOR REHABILITATION: Children's activities and participation were largely related to early learning tasks (e.g. literacy), communication, mobility and interactions. Intervention aimed at improving activities and participation may address the various child, impairment, social and environment factors identified here as impacting on activities and participation (e.g. the child's personal characteristics, communication and physical impairments, the support and attitudes of others and the familiarity of the environment). Therapists will need to consider (and manage) the potential negative impact communication deficits may have on children's behaviour, independence and social skills which may in turn detrimentally impact on activity and participation.

PMID: 25586796 [PubMed - indexed for MEDLINE]

#### Development of work participation in young adults with cerebral palsy: a longitudinal study.

Verhoef JA, Bramsen I, Miedema HS, Stam HJ, Roebroeck ME; Transition and Lifespan Research Group South West Netherlands.

Collaborators: van Meeteren J, van der Slot WM, Wiegerink DJ. J Rehabil Med. 2014 Jul;46(7):648-55. doi: 10.2340/16501977-1832.

OBJECTIVE: To document the development of work participation in young adults with cerebral palsy who are transitioning into adulthood, examine associated characteristics, and investigate work limitations and barriers among employed persons.

DESIGN: Observational longitudinal cohort study.

SUBJECTS: Seventy-four young adults with cerebral palsy of average intelligence, aged 16-20 years at baseline. METHODS: Work participation in 3 categories (employed, unemployed, studying) was assessed at baseline, 2-year and 4-year follow-ups using structured interviews. At 4-year follow-up, associations of work participation with

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demographic and clinical characteristics were examined using multinomial logistic regression. Work limitations and barriers among employed persons were evaluated using questionnaires.

RESULTS: From age range 16-20 years to age range 20-24 years the proportions of subjects who were employed and unemployed increased from 12% to 49% and 3% to 17%, respectively; the proportion who were students decreased from 85% to 34%. In the age range 20-24 years, the employment rate of young adults with cerebral palsy was lower and the unemployment rate higher, than that of the general population. A lower level of gross motor function and younger age were associated with unemployment. Employed persons experienced few work limitations; 28% experienced situational or health barriers.

CONCLUSION: Young adults with cerebral palsy and average intelligence are at risk of experiencing unfavourable developments in work participation. Rehabilitation services should offer support to prevent unemployment and occupational disability.

#### Free Article

PMID: 24858956 [PubMed - indexed for MEDLINE]

#### Physical Accessibility of Routine Prenatal Care for Women with Mobility Disability.

lezzoni LI, Wint AJ Smeltzer SC, Ecker JL

J Womens Health (Larchmt). 2015 Dec;24(12):1006-12. doi: 10.1089/jwh.2015.5385. Epub 2015 Oct 20. Comment in J Womens Health (Larchmt). 2015 Dec;24(12):963-5.

BACKGROUND: Routine prenatal care includes physical examinations and weight measurement. Little is known about whether access barriers to medical diagnostic equipment, such as examination tables and weight scales, affect prenatal care among pregnant women with physical disabilities.

METHODS: We conducted 2-hour, in-depth telephone interviews with 22 women using a semistructured, openended interview protocol. All women had significant mobility difficulties before pregnancy and had delivered babies within the prior 10 years. We recruited most participants through social networks. We sorted interview transcript texts using used NVivo software and conducted conventional content analyses to identify major themes.

RESULTS: Interviewee's mean (standard deviation) age was 34.8 (5.3) years. Most were white, well-educated, and higher income; 8 women had spinal cord injuries, 4 cerebral palsy, and 10 had other conditions; 18 used wheeled mobility aids. Some women's obstetricians had height adjustable examination tables, which facilitated transfers for physical examinations. Other women had difficulty transferring onto fixed height examination tables and were examined while sitting in their wheelchairs. Family members and/or clinical staff sometimes assisted with transfers; some women reported concerns about transfer safety. No women reported being routinely weighed on an accessible weight scale by their prenatal care clinicians. A few were never weighed during their pregnancies.

CONCLUSIONS: Inaccessible examination tables and weight scales impede some pregnant women with physical disabilities from getting routine prenatal physical examinations and weight measurement. This represents substandard care. Adjustable height examination tables and wheelchair accessible weight scales could significantly improve care and comfort for pregnant women with physical disabilities.

PMCID: PMC4683562 [Available on 2016-12-01]

PMID: 26484689 [PubMed - indexed for MEDLINE]

## Upper limb function in everyday life of children with cerebral palsy: description and review of parent report measures.

Wallen M, Stewart K. Disabil Rehabil. 2015;37(15):1353-61. doi: 10.3109/09638288.2014.963704. Epub 2014 Sep 29.

PURPOSE: To determine the role, in clinical practice and measurement of outcomes of upper limb interventions, of cerebral palsy-specific self- or parent-report measures of upper limb performance in everyday activities.

METHOD: Search of databases and handsearching for information on test development procedures, psychometric properties or relevant studies to inform study objectives.

RESULTS: Children's Hand-use Experience Questionnaire holds most promise for guiding treatment planning but requires more psychometric evidence. ABILHAND-Kids has the strongest evidence for reliability, validity and sensitivity to change; evaluates impact of intervention on bimanual performance and can be used for children with unilateral or bilateral cerebral palsy. The original and revised versions of the Pediatric Motor Activity Log (PMAL) evaluate unilateral rather than bimanual upper limb performance. Neither ABILHAND-Kids nor PMAL offer

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information to assist treatment planning. PMAL-R is the only measure for the 2-5-year age group. No measure was adequate for children younger than 2 years to ascertain parents' perception of upper limb function in everyday activities.

CONCLUSIONS: Understanding upper limb performance in everyday life, as perceived by children with cerebral palsy and their families, informs a comprehensive assessment and acknowledges the importance of the perspectives of child and family. Implications for Rehabilitation Cerebral palsy-specific self- or parent-report measures of upper limb performance in everyday life complement observational assessments in understanding upper limb performance CHEQ provides clinical information, ABILHAND-Kids is validated for children with unilateral and bilateral cerebral palsy and possesses the most robust psychometric properties, Revised PMAL measures unilateral upper limb use. No adequate measure for children under 2 years exists.

PMID: 25264734 [PubMed - indexed for MEDLINE]

## **4**Activité physique - Sport

#### Cardiovascular diseases in Paralympic athletes.

Pelliccia A, Quattrini FM, Squeo MR, Caselli S, Culasso F, Link MS, Spataro A, Bernardi M Br J Sports Med. 2016 May 26. pii: bjsports-2015-095867. doi: 10.1136/bjsports-2015-095867. [Epub ahead of print]

BACKGROUND: Sport participation (SP) of individuals with impairments has recently grown exponentially. Scarce scientific data, however, exist regarding cardiovascular (CV) risk associated with competitive SP.

OBJECTIVE: Assessing the prevalence of CV abnormalities and the risk for SP in Paralympic athletes (PA).

METHODS: PA (n=267; 76% men), aged 35±9 years, engaged in 18 sport disciplines, with a spectrum of lesions including: spinal cord injury (paraplegia and spina bifida) (n=116); amputation, poliomyelitis, cerebral palsy and other neuromuscular and/or skeletal disorders (Les autres) or visual impairment (n=151) entered the study. CV evaluation included history, PE, 12-lead and exercise ECG, echocardiography. Of these, 105 participated in  $\geq$ 2 consecutive games, and had evaluations available over a 6±4 year follow-up.

RESULTS: Structural CV abnormalities were identified in 33 athletes (12%), including arrhythmogenic cardiomyopathies in 3, aortic root dilation in 3, valvular diseases in 7 (mitral valve prolapse in 4, bicuspid aortic valve in 3) and systemic hypertension in 11 (4%). In addition, ventricular (polymorphic, couplets or non-sustained ventricular tachycardia) or supraventricular tachyarrhythmias (atrial flutter, paroxysmal atrial fibrillation or SVT) were identified in 9 others. Over a 6-year follow-up, 6 of the 105 athletes (6%) developed CV diseases, including dilated cardiomyopathy in 1 and systemic hypertension in 5.

CONCLUSIONS: PA present an unexpected high prevalence of CV abnormalities (12%), including a non-trivial proportion of diseases at risk for sudden death (2%), such as arrhythmogenic cardiomyopathies and dilated aortic root. This observation suggests that tailored recommendations for preparticipation screening and safe SP in this special athletic population are timely and appropriate.

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PMID: 27231335 [PubMed - as supplied by publisher]

# Comparison of Patterns of Physical Activity and Sedentary Behavior Between Children With Cerebral Palsy and Children With Typical Development.

Ryan JM, Forde C, Hussey JM, Gormley J.

Phys Ther. 2015 Dec;95(12):1609-16. doi: 10.2522/ptj.20140337. Epub 2015 May 28.

BACKGROUND: Reduced participation in physical activity and increased time spent in sedentary behavior are associated with overweight, chronic disease, and disability. In order to optimize recommendations and interventions to increase physical activity and reduce sedentary behavior in children with cerebral palsy (CP), knowledge of their physical activity and sedentary behavior is needed.

OBJECTIVES: The aim of this study was to describe light, moderate, and vigorous physical activity and sedentary behavior in preadolescent children with and without CP and compare physical activity and sedentary behavior between the 2 groups.

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DESIGN: This was a cross-sectional study of 33 children, aged 6 to 10 years, with CP (Gross Motor Function Classification System [GMFCS] levels I-III) and 33 age- and sex-matched children with typical development.

METHODS: Physical activity was measured using the RT3 accelerometer over 7 days.

RESULTS: Children with CP spent more time in sedentary behavior and accumulated less total activity, moderate activity, vigorous activity, and sustained bouts of moderate-to-vigorous activity (MVPA). They also accumulated a fewer number of bouts of MVPA and vigorous activity, despite spending a similar amount of time in each bout.

LIMITATIONS: The small number of children in GMFCS levels II and III did not allow for adjustment for GMFCS level when comparing physical activity between children with and without CP.

CONCLUSIONS: Preadolescent children with CP spent less time in moderate and vigorous activity and more time in sedentary behavior than children with typical development. Children with CP also accumulated less continuous MVPA and vigorous activity as a result of achieving fewer sustained bouts of MVPA and vigorous activity throughout the day.

© 2015 American Physical Therapy Association. PMID: 26023216 [PubMed - indexed for MEDLINE]

#### Health-related physical fitness of ambulatory adolescents and young adults with spastic cerebral palsy.

Nooijen C, Slaman J, van der Slot W, Stam H, Roebroeck M, van den Berg-Emons R; Learn2Move Research Group. J Rehabil Med. 2014 Jul;46(7):642-7. doi: 10.2340/16501977-1821.

OBJECTIVE: To describe in detail the health-related physical fitness of adolescents and young adults with cerebral palsy, compared with able-bodied references, and to assess differences related to Gross Motor Functioning Classification System (GMFCS) level and distribution of cerebral palsy.

DESIGN: Cross-sectional.

SUBJECTS: Fifty ambulatory persons with spastic cerebral palsy, GMFCS level I or II, aged 16-24 years.

METHODS: Physical fitness measures were: (i) cardiopulmonary fitness by maximal cycle ergometry, (ii) muscle strength, (iii) body mass index and waist circumference, (iv) skin-folds, and (v) lipid profile.

RESULTS: Regression analyses, corrected for age and gender, showed that persons with bilateral cerebral palsy had lower cardiopulmonary fitness and lower hip abduction muscle strength than those with unilateral cerebral palsy. Comparisons between persons with GMFCS levels I and II showed a difference only in peak power during cycle ergometry. Cardiopulmonary fitness, hip flexion and knee extension strength were considerably lower (< 75%) in persons with cerebral palsy than reference values.

CONCLUSION: The distribution of cerebral palsy affects fitness more than GMFCS level does. Furthermore, adolescents and young adults with cerebral palsy have reduced health-related physical fitness compared with ablebodied persons. This stage of life has a strong influence on adult lifestyle, thus it is an important period for intervention.

#### Free Article

PMID: 24714702 [PubMed - indexed for MEDLINE]

# Metabolic, cardiorespiratory, and neuromuscular fitness performance in children with cerebral palsy: A comparison with healthy youth.

García CC, Alcocer-Gamboa A, Ruiz MP, Caballero IM, Faigenbaum AD, Esteve-Lanao J, Saiz BM, Lorenzo TM, Lara SL J Exerc Rehabil. 2016 Apr 26;12(2):124-31. doi: 10.12965/jer.1632552.276. eCollection 2016.

The aim of this study was to assess metabolic, cardiorespiratory, and neuromuscular fitness parameters in children with spastic cerebral palsy (CP) and to compare these findings with typically developing children. 40 children with CP (21 males, 19 females; mean age, 11.0±3.3 yr; range, 6.5-17.1 yr; Gross Motor Function Classification System levels 1 or 2) and 40 healthy, age- and sex-matched children completed a test battery that consisted of 8 tests and 28

measures that assessed cardio-respiratory fitness, energy expenditure, anaerobic endurance, muscle strength, agility, stability and flexibility. Children with CP had significantly lower performance (P<0.05) on most cardiorespiratory and metabolic tests than those of healthy children, Differences in neuromuscular measures of muscular strength, speed, agility, anaerobic endurance, and flexibility between groups were most apparent. Grouped differences in cardiorespiratory variables revealed a 25% difference in performance, whereas grouped differences in metabolic and neuromuscular measures were 43% and 60%, respectively. The physical fitness of contemporary children with CP is significantly less than healthy, age-matched children. Significant differences in

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neuromuscular measures between groups can aid in the identification of specific fitness abilities in need of improvement in this population.

Free PMC Article PMCID: PMC4849492 PMID: 27162775 [PubMed]

Reliability and Validity of Objective Measures of Physical Activity in Youth With Cerebral Palsy Who Are Ambulatory. O'Neil ME, Fragala-Pinkham M, Lennon N, George A, Forman J, Trost SG Phys Ther. 2016 Jan;96(1):37-45. doi: 10.2522/ptj.20140201. Epub 2015 Jun 18.

BACKGROUND: Physical therapy for youth with cerebral palsy (CP) who are ambulatory includes interventions to increase functional mobility and participation in physical activity (PA). Thus, reliable and valid measures are needed to document PA in youth with CP.

OBJECTIVE: The purpose of this study was to evaluate the inter-instrument reliability and concurrent validity of 3 accelerometer-based motion sensors with indirect calorimetry as the criterion for measuring PA intensity in youth with CP.

METHODS: Fifty-seven youth with CP (mean age=12.5 years, SD=3.3; 51% female; 49.1% with spastic hemiplegia) participated. Inclusion criteria were: aged 6 to 20 years, ambulatory, Gross Motor Function Classification System (GMFCS) levels I through III, able to follow directions, and able to complete the full PA protocol. Protocol activities included standardized activity trials with increasing PA intensity (resting, writing, household chores, active video games, and walking at 3 self-selected speeds), as measured by weight-relative oxygen uptake (in mL/kg/min). During each trial, participants wore bilateral accelerometers on the upper arms, waist/hip, and ankle and a portable indirect calorimeter. Intraclass coefficient correlations (ICCs) were calculated to evaluate inter-instrument reliability (left-to-right accelerometer placement). Spearman correlations were used to examine concurrent validity between accelerometer output (activity and step counts) and indirect calorimetry. Friedman analyses of variance with post hoc pair-wise analyses were conducted to examine the validity of accelerometers to discriminate PA intensity across activity trials.

RESULTS: All accelerometers exhibited excellent inter-instrument reliability (ICC=.94-.99) and good concurrent validity (rho=.70-.85). All accelerometers discriminated PA intensity across most activity trials.

LIMITATIONS: This PA protocol consisted of controlled activity trials.

CONCLUSIONS: Accelerometers provide valid and reliable measures of PA intensity among youth with CP.

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PMID: 26089043 [PubMed - indexed for MEDLINE]

## **Vieillissement** - Passage à l'âge adulte

#### **Extrapolating published survival curves to obtain evidence-based estimates of life expectancy in cerebral palsy.** Day SM, Reynolds RJ, Kush SJ.

Dev Med Child Neurol. 2015 Dec;57(12):1105-18. doi: 10.1111/dmcn.12849. Epub 2015 Jul 15.

Studies reporting long-term survival probabilities for cohorts of persons with cerebral palsy provide evidence-based information on the life expectancy of those cohorts. Some studies have provided estimates of life expectancy based on extrapolation of such evidence, whereas many others have opted not to do so. Here we review the basic methods of life table analysis necessary for performing such extrapolations, and apply these methods to obtain evidence-based estimates of life expectancy from several studies that do not report such estimates themselves.

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## **4** Prise en charge et Accompagnement

#### Musculoskeletal system pain and related factors in mothers of children with cerebral palsy.

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Terzi R, Tan G. Agri. 2016 Jan;28(1):18-24. doi: 10.5505/agri.2015.74436.

OBJECTIVES: The aim of the present study was to identify prevalence of musculoskeletal system diseases and related factors among mothers of children with cerebral palsy.

METHODS: Eighty-five mothers of children with cerebral palsy were included as the treatment group, and 42 mothers of healthy children were included as the control group. Sociodemographic characteristics of all subjects were recorded. Musculoskeletal system pain was evaluated by the standardized Nordic Musculoskeletal Questionnaire, and level of depression was evaluated according to Beck's Depression Scale.

RESULTS: Musculoskeletal system pain and depression scores of the treatment group were significantly higher than those of the control group. Most frequently reported by mothers in the treatment group was low back pain (44.7%). In multiple regression analysis, number of children, age, and functional level of the child with cerebral palsy, as well as depression level of the mother were identified as independent risk factors for musculoskeletal system pain.

CONCLUSION: Mothers of children with cerebral palsy are at higher risk for musculoskeletal system pain and depression. Prevalence of musculoskeletal system pain in these mothers, especially those with older children who have lower functional statuses, should be kept in mind.

#### **Free Article**

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