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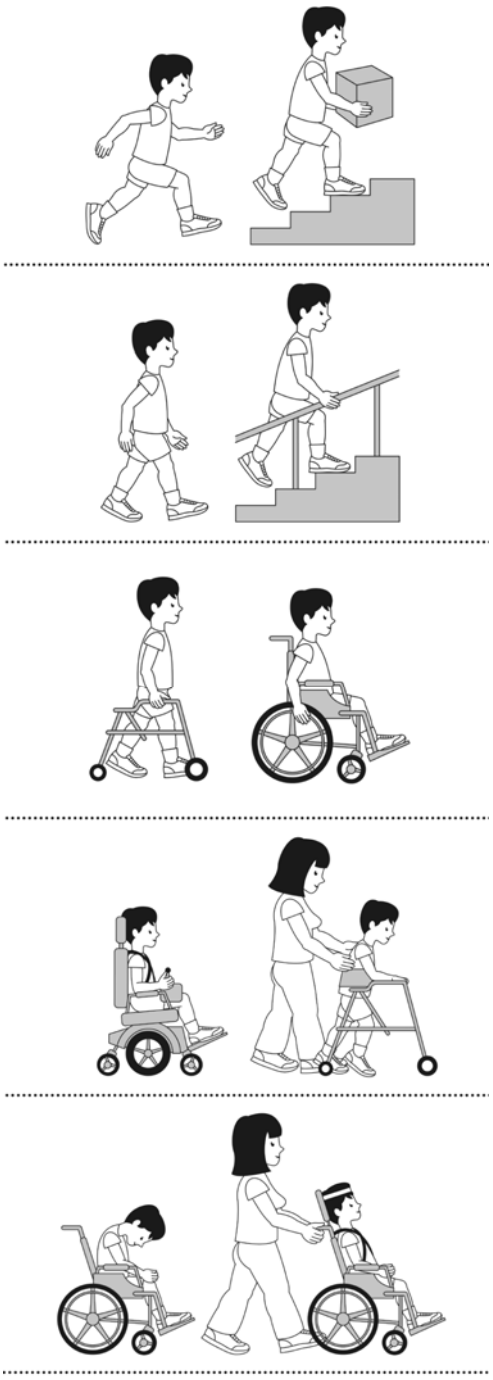

Recent Developments in Healthcare for Cerebral Palsy: Implications and Opportunities for Orthotics

Edited by
Christopher Morris
& David Condie

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Report of a meeting held at

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Edited by

Christopher Morris & David Condie

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Report of an ISPO conference organised by

Mr David Condie
Consultant Clinical Engineer,
11, Lansdowne Crescent,
Glasgow G20 6NQ
Scotland, UK
david@mailcondie.me.uk

Dr John Fisk
Emeritus Professor in Orthopaedic Surgery
4 Key West Drive,
Saint Helena Island,
South Carolina 29920
sailingfisk@gmail.com

Dr Christopher Morris
Principal Orthotist, Nuffield Orthopaedic Centre;
Research Fellow, Department of Public Health,
& Wolfson College, University of Oxford
Oxford OX3 7LF
christopher.morris@dphpc.ox.ac.uk

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INTRODUCTION

We welcome you to this report and in this introduction describe the background and context of the conference that led to the enclosed reviews and recommendations.

In 1994 ISPO convened a consensus conference on the lower limb orthotic management of cerebral palsy held at Duke University, USA. The conference considered the evidence for the use of lower limb orthoses in the physical management of children with cerebral palsy from the multidisciplinary perspective, which underlies the ethos of ISPO. The report of that conference, published the following year, became the society's most successful publication. The report has been used by ISPO as the basis for a series of nine instructional courses held around the world to disseminate the key treatment principles and clinical practices that were recommended.

There have been many developments in the understanding and management of cerebral palsy in recent years. The methods of assessment, classification and overall approach to treatment planning and coordination have become more sophisticated. New treatments have emerged; in addition research and experience with some established interventions has accumulated. This has resulted in changes in the ways in which orthoses are, and can be, used. New designs of orthoses have also become available. The need to measure the outcomes of all forms of treatment is now widely accepted.

In 2007 the Executive Board of ISPO, mindful of its responsibility to ensure that orthotic practice keeps apace with advances in healthcare, resolved to convene a follow-up to the previous consensus conference. This conference would consider relevant recent research and contemporary thinking in the healthcare of people with cerebral palsy identifying the implications for orthotic research and clinical practice. The scope of the conference was additionally expanded to include consideration of the evidence regarding the management of the spine, hips and upper limbs.

An international, multidisciplinary group of 24 health care professionals and research scientists, all of whom are recognised experts in this subject area, were invited to take part in the conference which was held in Wolfson College, Oxford in September 2008. Twelve of the participants were asked to prepare and present review papers. Their topics included a global health perspective, the definition and classification of cerebral palsy, the classification of gait, and physiotherapy, occupational therapy, medical, surgical and orthotic management of cerebral palsy. The reviewers of specific treatment areas were asked to synthesise the best available published evidence and where possible to grade their recommendations according to the level of evidence on which they were based using the Oxford Centre for Evidence-based Medicine Levels of Evidence (in Appendix).

At the conference, ten participants were nominated to each lead a plenary discussion session that followed the presentation of review papers. Participants were then separated into three smaller groups with the composition varying over the course of the conference. The small groups were used to explore gaps in the evidence and consider areas of controversy. In order to structure the discussion sessions, the organisers in consultation with the chairperson of each session devised a number of questions for the groups to address. Further plenary sessions were then held to hear the reports from each group and agree upon recommendations. The whole conference,

which lasted for three and a half days, was extremely rich in content and involved a great deal of hard work for everyone due to the huge amount of material which was covered.

Following the conference a first draft of conclusions and recommendations resulting from the small group reports was formulated by the organisers. This was first submitted to a previously agreed representative nominal group of the participants for comments. After modification to take account of their suggestions, this document was supplemented by the addition of the principal conclusions and recommendations contained in the reviews. The resulting composite Conclusions and Recommendations from the conference were then submitted to all participants for final comment and approval.

This Final Report contains further details of the organisation of the conference, the text of all the papers presented and the Conclusions and Recommendations. The presentations and plenary discussions were all video and/or audio recorded by our equally industrious media team. The material has been collated and made available by ISPO free of charge on the Internet. ISPO intend to use the report as an educational resource for further instructional courses and the information will be disseminated as widely as possible with the aim of improving the health care of people with cerebral palsy worldwide.

The organisers would like to thank all the participants for their sustained efforts and commitment to their tasks, and their contributions to this report.

David Condie
Glasgow, Scotland

John Fisk
S Carolina, USA

Chris Morris
Oxford, England

Copies of this Report can be obtained from
International Society for Prosthetics & Orthotics
Hans Knudsens Plads 1A, 2100 Copenhagen Ø, Denmark
Telephone: +45 39 20 72 60 Fax: +45 39 20 75 01
Email: ispo@ispoint.org
Web: www.ispoint.org

The Report can be downloaded free of charge from
www.ispoint.org

The presentations can be viewed at
http://homepage.mac.com/eaglesmoon/CP_Consensus_Conference/Home.html

PARTICIPANTS

Organisers

John Fisk MD
Emeritus Professor in Orthopaedic Surgery,
Southern Illinois University School of Medicine
USA.

David Condie BSc CEng FISPO
Consultant Clinical Engineer,
Glasgow, UK.

Christopher Morris MSc DPhil
Principal Orthotist, Nuffield Orthopaedic Centre;
Research Fellow, Department of Public Health
& Wolfson College, University of Oxford, UK

Reviewers

Peter Rosenbaum MD FRCP(C)
Professor of Paediatrics, Canada Research Chair In Childhood Disability
& Director, McMaster Child Health Research Institute,
McMaster University, Hamilton, Canada

Roy Davis III, PhD PE
Director, Motion Analysis Laboratory, Shriners Hospitals for Children Greenville
Professor of Bioengineering
Clemson University, Greenville, USA

Jules Becher MD PhD
Professor of Paediatric Rehabilitation Medicine
VU University Medical Center
Amsterdam, Netherlands

Deborah Gaebler-Spira MD
Professor, Departments of Physical Medicine and Rehabilitation & Pediatrics
Rehabilitation Institute of Chicago & Northwestern Feinberg School of Medicine
Chicago, USA

Professor Annette Majnemer BSc(OT) MSc PhD
Professor, School of Physical & Occupational Therapy,
McGill University, Montreal, Canada

Ann Christin Eliasson PhD
Associate Professor, Neuropediatric Research Unit,
Department of Woman and Child Health, Karolinska Institute
Stockholm, Sweden

Professor Kerr Graham MD FRCS (Ed) FRACS
Professor of Orthopaedic Surgery,
University of Melbourne, Murdoch Childrens Research Institute
Victoria, Australia

Jeremy Fairbank MA, MD, FRCS
Professor of Spinal Surgery
University of Oxford
Oxford, UK

Wayne Stuberg PT PhD PCS
Professor of Physiotherapy
University of Nebraska Medical Center
Nebraska, USA

Adrienne Harvey BAppSc MPhysio PhD
Physiotherapist & Postdoctoral Research Fellow, McMaster Child Health Research Instutute & Hugh Williamson Gait Laboratory,
Victoria, Australia

Roy Bowers Dip Pros Orth
Senior Lecturer in Orthotics
National Centre for Prosthetics and Orthotics, University of Strathclyde
Glasgow, UK

Phil Stevens MEd, CPO, FAAOP
Prosthetist & Orthotist
Hanger Prosthetics and Orthotics,
Salt Lake City, USA

Discussants

Steffen Berweck MD
Paediatric Neurologist
Department of Paediatric Neurology and Developmental Neuroscience
University of Munich, Germany

Diane Damiano MS(PT) PhD
Director, Biomechanics Program, Rehabilitation Medicine Department
National Institutes of Health
Bethesda, USA

Dianne Russell BSc MSc PhD
Associate Professor, School of Rehabilitation Science
& Ontario Federation for Cerebral Palsy Research Scholar
McMaster University
Hamilton, Canada

Marjolein Ketelaar PhD
Senior Researcher
Rehabilitation Centre De Hoogstraat & Rudolf Magnus Institute of Neuroscience
Utrecht, Netherlands

Jan Willem Gorter MD PhD
Associate Professor, Paediatric Rehabilitation Medicine
McMaster University
Hamilton, Canada

Barry Meadows PhD
Head of Neurobiomechanics
Westmarc (West of Scotland Mobility and Rehabilitation Services)
Glasgow, UK

Josephine Durkin PhD
Head of Occupational Therapy
Chailey Heritage Clinical Services
Lewes, UK

Laurie Snider BSc(OT) MA PhD
Associate Professor
School of Physical & Occupational Therapy,
McGill University
Montreal, Canada

David Phillips B.P&O
Orthotist, Orthotic Innovations
Victoria, Australia

Media

Dan Blocka BSc CO(C) FCBC
President of International Society of Prosthetics and Orthotics
Copenhagen, Denmark

Rebecca Westover BSc
Clinical Intern, Clinical Orthotic Consultants Inc.
Mississauga, Canada

Participants (left to right): Chris Morris, David Phillips, Dan Blocka, Adrienne Harvey, Peter Rosenbaum, Deb Gaebler-Spira, Roy Davis, Jules Becher, Jan Willem Gorter, Laurie Snider, Barry Meadows, John Fisk, Phil Stevens, Roy Bowers, Josephine Durkin, Wayne Stuberg, Dianne Russell, David Condie, Marjolijn Ketelaar, Anki Eliasson, Diane Damiano, Steffen Berwick, Annette Majnemer, Liz Condie, Kerr Graham.

CONCLUSIONS AND RECOMMENDATIONS

The following Conclusions and Recommendations are are drawn from the Review papers presented at the meeting and the subsequent Discussion sessions. Further details relating to each topic can be found in the relevant Reviews.

Despite improvements in the way people with cerebral palsy can be defined, classified and described, and the better quality of research methodologies generally employed in recent studies, it is apparent that the evidence for most interventions remains at a weak level or is inconclusive. Therefore, when producing this list, we found it difficult it distinguish evidence-based conclusions from opinion and consensus views. A caveat to many of the statements is that, in the absence of solid evidence, the statements reflect the best current practice based on the participants' knowledge and experience.

1. OVERARCHING THEMES

1.1 An important overarching goal of improving the health of disabled people is to enable culturally-appropriate 'activities and participation' as defined in the World Health Organisation's International Classification of Functioning, Disability and Health (ICF). 'Participation' in this context can be considered synonymous with the term 'social inclusion' often used in policymaking. The ICF is referred to through the report and it is recommended that readers familiarise themselves with the basic concepts; for further information see www.who.int/classifications/icf/en

1.2 It is believed that there are significant unmet needs for disabled people worldwide, and particularly in low/middle income countries where health service infrastructure, education and public awareness are lacking.

1.3 From a global health perspective, the appropriate means of promoting the participation/social inclusion of disabled people is dependent on local needs (both country and region-specific) and the availability of resources.

1.4 From an individual perspective social inclusion will be influenced by child and family needs, priorities and preferences. A 'family-centred' approach in delivering health services is likely to improve the wellbeing of children and their parents, and parental wellbeing is believed to be associated with improved outcomes for the child.

1.5 Transition from childhood to adulthood for disabled people needs to be supported by appropriate planning and coordination by the health, education and social services (including attention to housing, vocational and recreational opportunities).

1.6 The aim of health care for people with cerebral palsy is to enable activities and participation by promoting efficient movement, limiting deformity, reducing pain, and employing cognitive and/or behavioural strategies. These objectives may sometimes conflict, for instance when strategies to limit deformity temporarily hinder activities. Thus, the rationale for any intervention should be shared with the individual with cerebral palsy and the family to enable them to make an informed decision with the professionals, to prioritise goals and to plan management regimens.

1.7 In addition to neurological and musculoskeletal problems, children with cerebral palsy may require care for multiple impairments of sensation, perception, cognition, communication, and behavioural problems, epilepsy, difficulties with sleeping, drooling and feeding.

1.8 Health care for people with cerebral palsy requires the skills and input from a variety of professions who must work efficiently and collaboratively with the family as a team. Each professional, and the teams involved in managing different issues (e.g. orthopaedic, epilepsy, feeding etc) must understand the range of problems associated with the condition as well as the family goals and priorities.

1.9 When a child needs to receive many health services there is the potential for stress and other undesirable impacts on the wellbeing of both the child and the family.

1.10 Efficient co-ordination of health services is therefore crucial to minimise such adverse effects on the family. This issue is particularly pertinent at the times of transition, for example between primary and senior school, and from teenager to young adult.

1.11 Differences exist across countries, settings and health care systems in efforts made to co-ordinate services. There are examples of good practice such as *Key Workers* and much can be learned and shared between models of good practice. (A key worker is a named person who liaises with the family and professionals about the disabled child, and who ensures access to services and coordination of care).

1.12 For children attending multiple services including hospital and community-based health professionals, and/or education and social services, it is desirable to have a designated health or other professional to ensure information is exchanged efficiently and disseminated appropriately.

1.13 Within each service or multidisciplinary team (e.g. musculoskeletal, feeding, epilepsy, etc.) one health professional (e.g., a therapist or nurse) should be the designated point of contact for families to provide information, and liaise with other team members as appropriate.

1.14 Where appropriate, copies of letters and reports should be sent to the family to facilitate communication, understanding of treatment goals and the need for interventions.

1.15 The KIT® and YouthKIT® (*Keeping it Together* and *KIT for Youth*, which are methods for organising such information have been shown to empower families (available from *CanChild* Centre for Childhood Disability Research www.canchild.ca).

2. DEFINITION AND CLASSIFICATION OF CEREBRAL PALSY

2.1 Although there are various definitions of 'cerebral palsy' used in epidemiology and clinical practice, for the purposes of generalising the conclusions and recommendations of this report, the consensus amongst participants was to adopt the definition reported by Rosenbaum et al. (2007):

> *"Cerebral palsy (CP) describes a group of permanent disorders of the development of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain. The motor disorders of cerebral palsy are often accompanied by disturbances of sensation, perception, cognition, communication, and behaviour, by epilepsy, and by secondary musculoskeletal problems."*

2.2 Further classification of such a heterogeneous condition (i.e., classification of subtypes) is fundamental in research and clinical practice for communicating clearly the type and severity of the condition.

2.3 Classification of the type and distribution of motor impairment is recommended using the system advocated by the Surveillance of Cerebral Palsy in Europe (SCPE). The predominant types of motor impairment are spastic, dyskinetic (dystonia and choreoathetosis) and ataxic. The spastic type can be further classified according to the distribution as either unilateral or bilateral.

2.4 The terms hemiplegia, diplegia and quadriplegia are also commonly used to describe distribution in spastic cerebral palsy for unilateral involvement, lower limbs more affected than upper limbs, and total body (4-limb) involvement, respectively. However, while these terms are conceptually useful they are clinically imprecise as they lack reliability among observers.

2.5 When describing individuals or groups it is important to record associated impairments such as difficulties with (or impairments in) sensation, perception (hearing or vision), cognition, communication, behavioural problems, epilepsy, and musculoskeletal problems. Limitations in feeding and sleeping should also be recorded.

2.6 It is recommended that function be classified using valid and reliable systems such as the Gross Motor Function Classification System (GMFCS), Manual Ability Classification System (MACS), cognitive function in terms of Intelligence Quotient (e.g. IQ </>70), and other such systems as they become available.

2.7 The classification of gait is useful in the education of health professionals, for research and as part of a comprehensive clinical evaluation for children who can walk. Gait classifications have focused on children with spastic cerebral palsy predominantly in the sagittal plane and, although acceptable reliability has been shown, not all children can be classified in this way.

2.8 The qualitative classifications of gait in the sagittal plane proposed by Winters/Gage/Hicks for hemiplegia, and the Sutherland/Davids and Melbourne classification of sagittal gait patterns in spastic diplegia, provide the best available

starting points for clinically relevant classification of sagittal gait patterns (for further details see Davis paper page).

2.9 Improving the utility of gait classification systems is likely to require inclusion of the coronal, transverse and sagittal plane kinematics and the incorporation of other assessed parameters such as kinetics, EMG, clinical examination etc.

2.10 Longitudinal analyses of gait are desirable because of the tendency for gait patterns to change over time, and especially during growth.

2.11 Instrumented gait analysis is an important tool for teaching health professionals about gait, and can be helpful both in determining treatment options and in explaining them to patients and families.

2.12 Subject to the availability of resources and skilled personnel, instrumented gait analysis is desirable for assessing complex clinical problems to aid decision-making, in particular when considering orthopaedic and/or neurosurgical intervention and to evaluate the outcomes of intervention.

2.13 Instrumented gait analysis requires trained experts to minimize the potential sources of error in the process of gathering, analysing and interpreting the data.

2.14 In clinical settings where instrumented gait analysis is unavailable, observational analysis, with or without the use of video recording, is an acceptable alternative for the assessment and understanding of gait problems.

2.15 Observational gait analysis should be structured to consider the stages of the gait cycle, each of the hip, knee and ankle joints, and all three planes. An appreciation and understanding of basic biomechanical principles is essential.

3. RESEARCH DESIGN

3.1 There are key generic issues that are relevant to designing research studies to evaluate the effectiveness of interventions for people with cerebral palsy.

3.2 Randomised controlled trials (RCTs) are the gold standard research methodology for evaluating discrete interventions for both their efficacy and effectiveness.

3.3 RCTs are feasible for cerebral palsy research to evaluate discrete specific interventions provided the study uses a valid primary outcome measure in a well defined group of children and the appropriate sample size calculations have been performed and are realistically achievable. The intervention must be described in sufficient detail to allow replication.

3.4 RCTs are ethical if there is consensual equipoise (uncertainty) about the effect of the intervention and the study design will substantially address that uncertainty.

3.5 Blinding of researchers measuring outcomes to the mode of intervention is necessary whenever feasible; blinding of participants to the intervention is also desirable but may not be possible depending on the intervention being evaluated.

3.6 Well designed single-case studies, within-subject crossover studies, and 'Clinical Practice Improvement' studies (i.e. comprehensive data collection and analyses of patients, processes and outcomes in routine clinical practice) are credible alternatives when RCTs are not feasible, or as pilot studies to inform the design of a subsequent RCT.

3.7 The primary outcome measure should be directly related to the hypothesised benefits of the intervention. Any measure chosen must be known to be reliable and valid, and be able to detect relevant change in the phenomenon being evaluated.

3.8 There is increasing interest in finding out which interventions improve activities and participation (as defined in the ICF), in addition to addressing impairments at the body functions and structures level, such as range of joint motion and muscle strength.

4. PHYSIOTHERAPY

Physiotherapy interventions

4.1 While there is some evidence that particular physiotherapy techniques for children with cerebral palsy are effective, evidence for other techniques is not consistent.

4.2 There is consensus that the following interventions are not recommended:

- An exercise program comprised primarily of passive stretching delivered by a therapist. Parents or patients can be instructed to carry out these exercises themselves.
- Passive-reflexive (massage) techniques.
- Therapeutic Electrical Stimulation (TES) to increase function.
- Classical Bobath/neuro-developmental therapy (NDT) where the emphasis is on "normalization" of muscle tone through passive handling techniques such as the use of reflex inhibiting patterns.

4.3 Emerging scientific evidence suggests that the following interventions can be recommended for clinical practice:

- Exercise activities that include active participation of the child to attain functional goals.
- Progressive Resistance Strength Training (PRST).
- Use of assistive technology to promote mobility such as orthoses, wheelchairs, walkers or crutches.

4.4 There is uncertainty about the efficacy and effectiveness of the following interventions which require systematic evaluation, preferably in a research context:

- Partial Body Weight Supported Treadmill training
- Robotic assisted walking (e.g. Lokomat®)
- Night splinting

Dosage

4.5 The 'dose' of physiotherapy intervention (e.g., frequency, duration, etc.) is often decided following tradition and modified by economic considerations; the dose is seldom evidence-based and therefore the optimal dosage not known.

4.6 Where possible, the dose should be based on known physiological principles and modified by the initial evidence of efficacy established in practice.

4.7 Research is required to establish the optimum frequency, duration, intensity and timing of physiotherapy interventions.

4.8 Despite the lack of evidence to support many interventions, there was broad consensus that parents generally value regular contact with therapists and the educational, emotional and technical or "hands on" support they provide.

5. OCCUPATIONAL THERAPY

Seating/positional devices

5.1 The Functional Sitting Position (FSP) is the optimum posture that a seating system should provide; the seat should be combined with foot rests, hip belt, and a cut-out tray as appropriate. The FSP is recommended to be determined for each individual.

5.2 There is insufficient evidence that posterior tilting of the seat helps prevent deformity. In practice many children benefit of an upright/anteriorly tilted sitting positions for seeing, eating and using their upper limbs.

5.3 Factors which must be considered when assessing the effectiveness of seating devices include the characteristics of the users (i.e. type of CP, GMFCS level, spinal alignment, hip status, Body Mass Index (BMI) and age).

5.4 Outcomes assessed should address comfort/tolerance, ease of use and care-giving, achievement of posture appropriate for education, social function and use of upper limbs, and adverse outcomes such as hip subluxation/dislocation and scoliosis.

5.5 Although the biomechanical effect of the device may be immediately apparent, follow-up at several intervals after provision is necessary to assess posture and function.

Constraint-Induced Movement Therapy (CIMT)

5.6 There is a growing body of evidence that CIMT can improve manual activity in children with unilateral involvement (hemiplegia) in MACS (Manual Ability Classification System) levels I & II; however the long-term effects have not yet been studied.

5.7 For such children, CIMT is indicated when the child is motivated and the family is supportive of the intervention.

5.8 The type of constraint (device or discouragement) seems not to influence the results.

5.9 The intervention is probably dose-dependent in that its effectiveness might increases with the frequency and duration of therapy.

5.10 Further research is required to understand how CIMT works (e.g. using brain function imaging) and for whom it works best (e.g., for what age, dosage of training other factors that might influence the result).

Upper limb (including orthoses, splinting & casting)

5.11 Due to the relatively poor quality of studies carried out to date, there is no good evidence to support claims that wrist hand orthoses (WHOs) prevent or correct deformities and improve manual ability over time; the effectiveness of WHOs should be evaluated in research studies both in isolation and in combination with CIMT and Botulinum neurotoxin A (BoNT-A) .

5.12 Given the lack of an adequate range of sizes of off-the-shelf devices for children, WHOs should generally be custom-made to achieve acceptable comfort and fit, and to optimise biomechanical efficacy.

5.13 The results of studies that have evaluated periods of casting of the upper limb show some effect of increasing range of motion and decreasing spasticity in the short term. However it is not known whether these potential gains lead to improvements in activities and participation; or whether they can be sustained long term.

5.14 Training that takes a functional, task-specific approach, with or without WHOs, may help to overcome activity limitations; this should be evaluated in future studies.

Feeding Interventions

5.15 There is moderate to weak evidence of the effectiveness of occupational therapy interventions that aim to improve feeding. Specifically, sensorimotor interventions may improve oral-motor skills, and positioning techniques may enhance oral-motor control. Future studies should address multi-modal approaches to promote growth and development.

Virtual Reality (VR)

5.16 Training using Virtual Reality (VR) is a new and emerging intervention that has shown promising, if preliminary, results on motor and visual-spatial skills, using a range of simulated situations. Therefore, VR may be a useful therapeutic intervention for children with cerebral palsy, and one that is similar to the games in which many children like to participate.

6. MEDICAL MANAGEMENT

Life Expectancy

6.1 Severity of impairment (particularly motor, but also cognitive and visual) is associated with decreased life-expectancy in people with cerebral palsy. Nevertheless, nearly all children with cerebral palsy become adults; consequently health care for people with cerebral palsy should be considered from a life-span perspective.

Nutrition

6.2 Gastrostomy, or other forms of parenteral feeding, can improve nutrition, increase body weight and improve growth in children in GMFCS levels IV and V that are unable to gain or maintain weight through feeding orally.

Spasticity management

6.3 Orally administered anti-spasticity medications can be used to decrease spasticity and improve comfort and well-being. Given the paucity of evidence, and the amenability of such pharmacological intervention to rigorous research, large randomised controlled trials are warranted using appropriate outcome measures.

6.4 Focal management of spasticity with Botulinum neurotoxin A (BoNT-A) avoids the side effects associated with more global interventions for spasticity, although orally-administered medication may still be indicated.

6.5 Botulinum neurotoxin A (BoNT-A) is useful for overcoming activity limitations caused by spasticity; BoNT-A is typically used in conjunction with orthoses and physical and/or occupational therapy.

6.6 In GMFCS levels I, II & III treatment with BoNT-A can improve gait, limit progression of fixed deformity during growth, reduce intolerance to orthoses resulting from spasticity, and may delay the need for orthopaedic surgery.

6.7 BoNT-A may be used in the upper limb to improve function and/or appearance. However, there is no evidence that BoNT-A is effective in improving bilateral hand function. Nevertheless, in principle, the three months after injection would appear to be an ideal period for a therapist to provide training. It is recommended that the effectiveness of BoNT-A to improve manual ability be evaluated in research studies.

6.8 In GMFCS levels IV and V, BoNT-A may be useful to improve posture and positioning, reduce pain, reduce drooling, improve hygiene, and ease caregiver burden.

6.9 Intrathecal Baclofen (ITB) should be considered if hypertonia is severe and warrants aggressive management. Children using ITB should be followed closely for complications by an expert team. Functional gains and changes in quality of life and caregiver burden should be measured in controlled studies with consistent definitions and assessment tools.

6.10 Selective Dorsal Rhizotomy is a well-researched treatment option that has been shown to improve gait and mobility for a defined group of children with bilateral

spastic cerebral palsy. Consideration must be given to the published criteria for identifying children likely to benefit.

Weight-bearing programmes

6.11 Weight-bearing programmes for children in GMFCS levels IV and V utilise a range of equipment to facilitate more upright posture in a variety of positions; some of these provide limited weight-bearing but encourage activity and experience in postures other than lying and sitting.

6.12 There are many potential benefits from using weight-bearing programmes such as increasing bone density (and reducing the potential for osteopenia and fractures), and delaying progression of lower limb deformities and scoliosis, also possibly improving respiratory function (preventing aspiration) and bowel/bladder function (urinary drainage and reducing constipation). There are also potential social and behavioural benefits if the experience is enjoyed, and if the posture facilitates improved social interaction.

6.13 Unfortunately, despite the wide range of potentially important benefits, no evidence is yet available from research studies to substantiate these outcomes and this is an important topic requiring further research to inform guidelines for clinical practice.

6.14 There are potential harms from weight-bearing programmes including pain and fractures; there is also the physical burden of using and storing the equipment at home.

Other forms of Medical Management

6.15 There are many new and emerging medications and surgical procedures for the management of spasticity, movement disorders and posture; these require further research to better define their effectiveness and roles in physical management programmes.

7. ORTHOPAEDIC SURGERY

Single event multilevel surgery

7.1 A key development in strategic thinking of how to manage fixed musculoskeletal deformity in cerebral palsy has been the move from single level surgery, repeated at frequent intervals throughout childhood (sometimes referred to as the 'birthday syndrome'), to single event multilevel surgery (SEMLS).

Equinus

7.2 Equinus is the most common musculoskeletal deformity in cerebral palsy and may adversely affect standing, posture and gait; specifically stability in stance and clearance in swing.

7.3 Although the foot and ankle cannot be considered in isolation from the rest of the lower limb, the following strategies can be used to manage equinus in children in GMFCS levels I, II & III, subject to availability of resources.

7.3.1 Intervention for equinus should start as soon as the problem becomes evident, for instance, stretching, taping and strapping can be used in infants.

7.3.2 Dynamic equinus (where there is a good range of passive dorsiflexion) can be managed by focal injections of BoNT-A to the gastroc-soleus, stretching and ankle- foot orthoses.

7.3.3 BoNT-A may also be indicated to treat gastrocnemius spasticity when it causes the knee to flex excessively during stance, or when ankle foot orthoses are not tolerated despite an adequate range of dorsiflexion being available.

7.3.4 Serial casting can be used in an attempt to increase range of motion where there is a reduced range of dorsiflexion (fixed equinus); ankle foot orthoses should be used to maintain any gain in dorsiflexion range.

7.3.5 Orthopaedic surgery is indicated to correct equinus if the deformity progresses to a stage that interferes with physical function, and should be followed by casting and ankle foot orthoses. The timing and amount of lengthening (dosage) are crucial to minimise the risk of over-lengthening, joint instability and likelihood of recurrence.

7.3.6 If surgical correction is not possible then the deformity must be accommodated in ankle foot orthoses to promote stability and efficient gait.

Upper limb surgery

7.4 The development of the MACS (Manual Ability Classification System) is a major step forward in description of upper limb function. Assessments such as the Melbourne Assessment of Unilateral Limb Function are also useful outcome measures to evaluate upper limb surgery.

7.5 As with lower limb surgery, increasing numbers of surgeons are moving from a single level approach to a "single event multilevel surgery" approach in reconstructive surgery of the hemiplegic upper limb

Hip subluxation/dislocation

7.6 The risk of hip subluxation/dislocation in cerebral palsy increases linearly by GMFCS level from negligible in level I to almost universal in V. However, there is an increased risk for children with Winters' type IV hemiplegia to develop late onset hip subluxation which may be extremely disabling.

7.7 All health professionals involved in the care of children with cerebral palsy should be aware of the risk factors for hip subluxation/dislocation. A designated member of the multidisciplinary team must take responsibility for each child's hip surveillance.

7.8 The Australian Hip Surveillance Guidelines provide a carefully constructed framework for monitoring hip status in children with cerebral palsy. It is recommended that each centre should have such a guideline for follow-up and management of the hips in children with cerebral palsy.

7.9 Surgical management of the hip should be considered when the migration index exceeds 30 degrees. Outcomes of hip surgery are better when performed as soon as indicated rather than being delayed while using alternative interventions.

7.10 BoNT-A or phenol injections (obturator nerve only) can be used to reduce the deforming forces caused by spastic muscles when surgery is not possible, to delay the need for surgical intervention, or to treat pain and discomfort.

Scoliosis

7.11 The risk of scoliosis appears to be higher for non-ambulant children; therefore those in GMFCS levels IV and V are like to be at most risk for developing scoliosis.

7.12 Surgical correction of scoliosis aims to improve posture, facilitate better seating positioning, reduce pain and discomfort, and reduce caregiver burden.

7.13 Secondary benefits of surgery to correct scoliosis may include improved respiratory function, feeding and gastrointestinal motility, and decreased reflux. Other secondary benefits may include improved use of the upper limbs, improved head control enabling use of head switches and improved view.

7.14 The results of scoliosis surgery are primarily measured on radiographs by a Cobb angle, however outcome measures such as the Spinal Alignment and Range of Motion Measure (SAROMM) or family-completed questionnaires such as the Caregiver Priorities and Child Health Index of Life with Disabilities (CPCHILD), Paediatric Pain Profile, Pediatric Outcomes Data Collection Instruments (PODCI), or the 'caregiver assistance' scale of the Pediatric Evaluation of Disability Inventory (PEDI) are also available to evaluate various possible outcomes of scoliosis surgery.

7.15 Corrective scoliosis surgery may be beneficial for patients with total-body-involvement (GMFCS Level IV and V) cerebral palsy and scoliosis. However the

benefits and risks of surgery should be further evaluated in appropriate research studies.

7.16 Segmental, third-generation instrumented spinal fusion appears to provide lasting correction of spinal deformity and improve quality of life in patients with cerebral palsy and scoliosis with a lower rate of pseudarthrosis than second-generation instrumented spinal fusion.

7.17 Modern instrumentation has allowed more surgery to be completed from a posterior approach alone. In children with severe deformity, especially when there is significant growth potential, combined anterior and posterior surgery is indicated. There is discussion as to do this in a single or staged sitting. Some investigators believe that a two-stage procedure appears to be safer and associated with fewer complications than when conducted in a single operation, especially in individuals with very large curves and concomitant medical illness.

7.18 Severe preoperative thoracic hyper-kyphosis appears to affect survival adversely and the parents of these high-risk children should be counselled about that likely prognosis.

Orthopaedic Surgery Research

7.19 The rigor of research in orthopaedic surgery in cerebral palsy will improve with well constructed research questions, prospective longitudinal design, well defined groups of patients (described by type and distribution of CP and GMFCS level), appropriate primary and secondary outcome measures, and long-term follow-up where possible.

7.20 The clarity and transparency of papers describing orthopaedic surgery research will improve if the reporting guidelines advocated by The EQUATOR Network (Enhancing the Quality and Transparency of Health Research) are utilised.

7.21 Orthopaedic surgeons and trainees need to be better educated consumers of the research literature to identify and reduce the number of poorly designed studies that are conducted and published.

8. ORTHOTICS

Prescription of orthoses

8.1 Dialogue between the orthotist and other members of the team (therapist, physician, surgeon, bioengineer etc.) is essential when deciding upon treatment goals and the biomechanical requirements to achieve these goals.

8.2 The role of the orthotist is to formulate the design, fit, align, deliver and review the orthosis which will, in theory, achieve the biomechanical requirements agreed by the team.

8.3 When an orthosis is prescribed consideration must be given as to when, and for how long in each twenty-four hour period it should be worn. The optimum duration of use (dose) and occurrence of side-effects (pressure sores, disturbed sleep, etc.) should be evaluated in future research studies.

8.4 Adherence is likely to be higher when there is clear agreement between the physiotherapist and the orthotist regarding usage, and the family understands the rationale for the prescription.

Lower limb orthotics

8.5 For ambulant children, the available evidence from gait laboratory studies suggests that ankle foot orthoses (AFOs) that prevent plantarflexion can improve gait efficiency improving temporal and spatial parameters of gait, (i.e., velocity, cadence, step length, stride length, single and double support) and ankle kinematics.

8.6 The indirect effects of AFOs on the kinetics and kinematics of the knee and hip joints have been demonstrated in gait laboratory studies; and these effects can be optimised by tuning the orthoses.

8.7 The energy cost of walking may be decreased by the use of AFOs.

8.8 It is unclear whether and how AFOs influence phasic activity of lower limb muscles.

8.9 It is unclear how the assessment of the efficacy of AFOs in the gait laboratory can be used to predict children's mobility and gait efficiency in their usual environments.

8.10 There is inconclusive evidence of the effect of AFOs on children's motor function. The Gross Motor Function Measure has been shown to be sensitive to the changes associated with use of orthoses and mobility aids and could be used to evaluate this outcome.

8.11 It unclear whether AFOs can maintain/increases muscle length and hence prevent/reduce deformity developing over time.

8.12 Long term effects of orthoses on muscle strength are unknown. Given recent recognition of the importance of muscle strength and endurance for maintaining mobility into adulthood, further research on this topic is desirable.

Upper limb orthoses

8.13 Thumb abduction orthoses for children with hemiplegia are proposed to improve hand function by facilitating grasping and preventing fixed deformity; however there is no scientific evidence demonstrating these effects.

8.14 A rigorously constructed study is required to address the research question 'Does a thumb abduction orthosis improve use and manual function of the affected hand in children with hemiplegia?'

> 8.14.1 The subjects would be children with spastic hemiplegia, MACS level I or II, in a defined age group, perhaps focusing initially on preschool children. The Zancolli and House classifications of grasping and hand function may also be useful.
>
> 8.14.2 The orthosis would be a custom-made thumb abduction orthosis with appropriate strapping; the comparison condition would be no orthosis.
>
> 8.14.3 The research design should include a cross-sectional element to assess whether the orthosis achieves the biomechanical objective (maintaining abduction etc.) and a longitudinal study of whether it actually improves hand use and functioning with training.
>
> 8.14.4 The outcomes assessed could comprise both unilateral and bilateral manual ability; measures could include the Assisting Hand Assessment (AHA), Quality of Upper Extremity Skills Test (QUEST), Melbourne Assessment of Unilateral Upper Limb Function, and the Canadian Occupational Performance Measure (COPM).

8.15 There is the potential for a 'factorial' randomised controlled trial evaluating the individual and combined effectiveness of thumb abduction orthoses, constraint-induced movement therapy and/or BoNT-A in the same study.

Spinal orthoses

8.16 It is generally accepted that spinal orthoses do not appear to prevent progression of scoliosis in children with cerebral palsy; however, by stabilising trunk posture, they may provide other substantive benefits.

8.17 Although progression may not be arrested, in theory, spinal orthoses may reduce the rate of progression of scoliosis by reducing the deforming forces on the spine; hence delaying the timing of surgery until cessation of growth, or in some cases, avoiding the need for surgery.

8.18 The benefits of spinal orthoses may include improved head control (perhaps enabling the use of head switches and improving view perspective) and/or upper limb function, and social interaction. Spinal orthoses can be used in conjunction with a seating system.

8.19 A spinal orthosis may be beneficial for children for whom surgery is not feasible.

8.20 Spinal orthoses are particularly recommended for small curves, and to assure the family that all alternative interventions have been tried before surgery is considered.

8.21 The primary measure of the effectiveness of spinal orthoses is the Cobb angle measured from X-ray; secondary outcomes could be the incidence of pressure sores or respiratory infections, improved head control and/or upper limb function, and social interaction.

8.22 There may be an increased risk of rapidly progressing scoliosis following intervention using intrathecal Baclofen (ITB); a potential research study could compare rates of curve progression in children with and without a spinal orthosis following this intervention.

Hip orthoses

8.23 There is no evidence that hip orthoses (for example the SWASH orthosis) prevent progressive hip subluxation/dislocation over time.

8.24 However, it is suggested that hip positioning devices (orthoses, seating, postural management devices) may produce other benefits.

8.25 For non-ambulant children hip orthoses may improve symmetry of sitting posture and comfort; outcome measures that could be assessed include range of hip motion, seated pressure mapping, and/or timed sitting tolerance.

8.26 For ambulant children hip orthoses may control scissoring of the lower limb during gait and thereby improve stability, acquisition of skills and gait efficiency.

8.27 Further research is required to substantiate these potential benefits of hip orthoses.

8.28 Hip orthoses are useful for positioning in postoperative management regimens.

Orthotic research

8.29 Research studies evaluating the effect of orthoses often lack sufficient detail of the research methodology, the subjects and/or the intervention, thereby failing to inform future research, and preventing generalisation or replication in the clinical setting.

8.30 Papers describing orthotic research studies should include a minimum dataset of details of the subjects and the design of the orthosis being used by the subjects in the investigation. Details of this proposed minimum dataset should be disseminated to relevant journal editors to aid them in consideration of papers submitted for publication.

8.31 The minimum dataset of details of the subjects should include age, sex, type and distribution of cerebral palsy, GMFCS level and/or MACS level (for studies of the upper limb) and whether subjects have had recent surgery or pharmacological intervention.

8.32 Other requirements include a well constructed research question stating the hypothesis being investigated, the research methodology, and the outcome measures being used.

8.33 There is often confusion about what is evaluated in orthotic research studies, between whether an orthosis achieves its biomechanical effect (e.g., improving gait pattern in the gait laboratory) and whether it improves function in daily life.

8.34 It is acknowledged that comprehensive reporting of such details will increase the length of research papers, but is considered necessary for appreciation of what was done in the study, and supplementary material is now commonly published by journals online.

8.35 Most studies of orthoses are cross-sectional in design; longitudinal studies are recommended to assess the long-term impact of orthoses.

8.36 It is recommended that for any project investigating the use of orthoses an orthotist be an integral member of the research team.

Research involving ankle foot orthoses

8.37 Description of the subjects should include range of motion (ROM) of all lower limb joints, highlighting whether range is attained with ease or difficulty. Particular reference should be paid to dorsiflexion range with knee extended, which is an indication of gastrocnemius length. Any fixed deformities (including rotational deformity) should be reported, together with an assessment of muscle spasticity and strength.

8.38 A description of gait, both with and without orthoses if appropriate, should be provided, stating whether or not mobility aids are used.

8.39 A minimum description of AFOs used to improve gait should included the design and construction of the orthosis, alignment of the leg in the orthosis, alignment of the orthosis to the ground in footwear, and the footwear design.

8.39.1 The design and construction description should include whether it is custom-made or prefabricated, materials (type and thickness), trimlines including foot plate length, type of any ankle articulations and range of motion permitted by these (or by flexible AFO designs), and the type and location of straps and fastenings.

8.39.2 The foot and ankle alignment imposed by the AFO should be described in the sagittal, coronal and transverse planes.

8.39.3 The alignment of the AFO and leg in the sagittal plane when the subject is standing wearing the orthosis with footwear should be stated.

8.39.4 Details of the footwear should include the pitch (heel-sole differential), the design of the heel, the stiffness of the sole, the sole profile, and any additional footwear modifications.

Lycra garments

8.40 There is considerable variation in the extent of body areas covered by garments ranging from gloves to body suits which may confound interpretations of effectiveness.

8.41 Some subjects appear to experience functional gains using Lycra garments; however others experience difficulties with donning and doffing the garments, feeling hot and restricted, difficulties with toileting and incontinence, and respiratory compromise.

8.42 The characteristics of people who benefit from using Lycra garments are not well defined; from the literature.

8.43 The effectiveness of Lycra and similar garments should be evaluated carefully, preferably in a research context.

CEREBRAL PALSY - A GLOBAL PERSPECTIVE

John R. Fisk, MD
Emeritus Professor
Southern Illinois University School of Medicine, USA

The sponsoring body for this Consensus Conference Update is the International Society of Prosthetics and Orthotics. ISPO is a professional society familiar to nearly all of the participants but for those new to its activities let me say a few words. The International Society for Prosthetics and Orthotics (ISPO) is a multi-disciplinary organization comprised of persons who have a professional interest in the clinical, educational and research aspects of prosthetics, orthotics, rehabilitation engineering and related areas.

ISPO was founded in Copenhagen, Denmark in 1970 as an international society by a group of surgeons, prosthetists, orthotists, physiotherapists, occupational therapists and engineers to promote improvements in the care of all persons with neuromuscular and skeletal impairments. ISPO is a Non-Governmental Organization (NGO) in Special Consultative Status with the Economic and Social Council of the United Nations, and is in Official Relations with the World Health Organization.

ISPO presently has 3000 members worldwide in 84 countries. The image of ISPO is that of a professional society which has taken responsibility for setting the standards for and supervising the education of Orthotists and Prosthetists primarily in the developing world, however increasingly it is being called upon to do the same in the developed world. It also promotes research and is a forum for the investigation and exchange of information on services for the physically disabled.

One of its recurring activities is the sponsoring and organization of Consensus Conferences. Since1960 it has had held eight such conferences.

- 1990 Amputation Surgery, Glasgow, Scotland
- 1994 Lower Limb Orthotics Management of Cerebral Palsy, Durham, NC, USA
- 1995 Appropriate Prosthetic Technology, Phnom Penh, Cambodia
- 1997 Poliomyelitis, Hammamet, Tunisia
- 2000 Appropriate Orthopaedic Technology Up-date, Moshi, Tanzania
- 2003 Orthotic Management of Stroke Patients, Ellecom, the Netherlands
- 2006 Appropriate Lower Limb Orthotics for Developing Countries, Hanoi, Vietnam
- 2006 Wheelchair Technology for Developing Countries, Bangalore, India

The purpose of each conference has been to review the pertinent body of literature and derive best practices reports. Some topics clearly emphasized the needs of developing countries but none sought to set a different standard for high income and low income nations. Many led to the development of Short Courses based on the recommendations derived from the consensus. The Orthotic Management of Cerebral Palsy has been one such course. Under the able leadership of David Condie it has been given in seven different countries. It has become increasingly obvious that the material from 1994 on which the course has been based is out dated and needs to be revisited. That is what we are gathered here to consider this week. David has had a nice run. For this and other contributions to his professional field and ISPO he was

awarded the Knud Janssen Lectureship at the 2007 ISPO World Congress. It is never too late to congratulate him publicly. Jules Becher currently and for the next two triennia has the responsibility for being the Task Officer for Cerebral Palsy Courses.

Since ISPO maintains a strong international perspective I felt this workshop should begin with just such an international view point. Most of what we will be discussing is information that comes from work and publications conducted in the developed world. What is known about cerebral palsy in the developing world? In a few words - not very much. I would like to review the literature from low income countries, report on some interesting statistics we have available from those countries and report on some personal observations made after working in developing nations.

LITERATURE REVIEW

The literature review was not an arduous task. It is almost non existent. I found four clinically based papers, fewer epidemiological papers and none having to do with orthotic management. I enlisted the reference librarians at my medical school library and they assured me that they went to non-standard sources.

According to Msall and Hogan (1), 4 million of the 130 million infants born each year around the world die during the first 4 weeks of life. Major associations with these early deaths include preterm birth, sever infections, and asphyxia, which in aggregate contribute to 80% of these deaths. They note that the historic assumption has been that if one sequentially applies advances in preventive health, then the presence of child disability will be substantially reduced. In the preschool years, this has happened with reduction of motor disability from polio and cerebral palsy from iodine deficiency; and other infectious diseases from vaccination. In low income countries in south Asia and sub-Saharan Africa, 200 million children under 5 years of age fail to reach their cognitive potential because of poverty, poor health and nutrition and the suboptimal home environment. (2)

The current knowledge on child disability in low-income (less than $875 gross national income) and middle-income countries ($875-$3465 gross national income) is woefully inadequate (3). Two diseases causing an increase in childhood encephalitis and resultant upper motor neuron dysfunction about which there is some data are HIV-AIDS and Malaria (14). In 2006, the estimated number of children younger than 15 years infected with AIDS was 2.3 million (8). In Zimbabwe 150% of children, in as much as many have two infections, contract Malaria.

Cerebral palsy is 5 to 10 times more common in poorer countries (4). Disorders of the nervous system account for at least 15% of the global burden of disease and at least 27% of average years lived with disability (5). These numbers refer only to disorders that arise within the brain. If we add the impact of the many conditions that damage the brain as part of their overall effect such as noted previously, the numbers become much larger approaching 30% of the global burden of disease (6). Brain health and disease may be the best overall indicator of a nation's success in promoting health.

It is often noted that the majority of related causes for cerebral palsy in developed countries is idiopathic. The incidence of CP in very low birth weight children is consistent at 7.7% for children born at less than 1000 grams. Most of the identified pathologies in these children are periventricular leukomalacia and proencephaly. Increased survival rates as a result of improved neonatal intensive care units have not

increased the numbers of children thus affected. In a study from Soweto similar findings have been reported (7). Survivability was increased from 24% to 66% for children under 1500 grams. They did not have sufficient respirator facilities for children under 1000 grams. Similar survival figures in the United States between 1960 and 1983 rose from 43% to 85%. Rates for cerebral palsy in both populations remained similar.

Hagberg reported that "the prevalence of cerebral palsy in preterms is no longer increasing, in spite of additional improvements in survival."(12) In a similar manner, Fanaroff et al. (13), reporting on prospective observations from 12 US centers, in 1995 concluded that the increase in survival of very-low-birth-weight infants seen after the introduction of surfactant therapy in the 1080's "was not accompanied by an increase in medical morbidity."

Over all global rates of Cerebral Palsy are between 2 and 3 per 1000 life births.(9) Although these figures are not available in low income countries there is data on the types of involvement in children with spasticity. Ozmen reported from Turkey that the types encountered according to Hagberg's categorization (10) were similar to developed countries.(11) My own unscientific observations in southern Asia and sub-Saharan-Africa is that there are more dyskenitic types perhaps from untreated ABO incompatibility and resultant kernicterus.

In a clinical study, M.A. Khan reported in 2007 on 85 children treated with single event multiple site surgery (15). He referenced the suggestions of Eugene Bleck and later of R.S. Paine that a child having spastic diplegia if unable to walk by age seven or eight probably would not walk. Reporting on his work done in Karachi, Pakistan he noted that there is a difference between western nations and his own. The children that the earlier authors were working with received treatment from the age of diagnosis. Khan's patients on the other had presented on average at age 8.5 years. He was able to affect ambulation in all 85 who had not been walking at the time of presentation. 18(21.2%) attained exercise ambulation, 39(45.9%) attained household ambulation and 28(33%) attained community ambulation.

This literature review illustrates the need for improved data but more importantly the desire to identify problems unique to low income countries. As we send our foreign students back to their home countries we must encourage them to repeat many of the studies performed in our high income countries in their low and middle income countries.

SURVEY RESULTS

Early in 2008 a brief questionnaire was sent to a number of treatment facilities in low income countries. The address list of ISPO which consists of all the certified P&O schools, non certified schools and their related workshops was used. The same questioner was also sent to all of the International Committee of the Red Cross P&O workshops. Seven questions were submitted for response. They dealt with requests for data on population incidence, numbers of patients served, types of services and needs for improved services.

Responses were received from eighteen low income countries and twenty-five work treatment facilities. Very few ISPO associated centers responded where as most ICRC workshops sent in their information.

Question 1: Do you have any data on the incidence of cerebral palsy in your region?

Fourteen facilities reported that no data is kept. One country kept records but would not release it to the public.

Question 2: Do you provide services to persons having cerebral palsy?

Twenty reported that they do, but that most are very limited.

Question 3: Which services do you or your facility provide?

Physical therapy 16, Orthotics 11, Surgery 5. One said that since "CP is not correctable so they did not provide services". Another facility said they have examples of equipment available for families to see so that they can construct them at home.

Question 4: What volume of services for CP are you or your facility providing?

Few had any data. One clinic said they saw as many 135 children with CP per month. One clinic fit 168 orthoses in 2007.

Question 5: What age range are you patients/clients?

Most said all of their CP patients were children, some worked with adults.

Question 6: What is your ability to address the level of need in your region?

"The ability will be defined according to the parents' attitude to CP and with reference to social stigma which is a problem in my region. It is imperative that the disabled view themselves as functional beings and it is critical that parents do not shy away from seeking professional help for their child."

There are insufficient personnel to do home visits. Unrealistic expectations of families lead to discouragement. In Kashmir there are only two facilities for 12 million people. There is need for better awareness.

Question 7: What are your needs, what are you lacking to provide services?

There are needs in all areas, infrastructure, manpower, therapy equipment, social workers, CBR, education, transportation, and food. "Lacking is specific training in the care of persons with CP for physicians and therapists." Lacking is trust to dispel stigma and money for home services.

One country acknowledged little interest or willingness in the care of people with cerebral palsy.

CONCLUSION

With a paucity of scientific literature concerning the adequacy of services for the care of persons with cerebral palsy in low and middle income countries few recommendations on best practices can be made. The World Health Organization

Task Force on Medical Rehabilitation Guidelines met in Geneva in October of 2005. One of the clear recommendations coming from that session was the need for accurate country by country data on the incidence and types of disabilities as a starting point in stimulating local governmental organizations to address service needs. Few Ministries of Health will act without identified needs and top down guidelines.

All of you who have had the privilege to work in low income countries as I have know of the inadequacy of the orthotic services seen there. The fabricator is rarely a consultant and only a technician. Materials are clumsy. Design is with poor biomechanical understanding and there is very poor understanding of cerebral palsy as a clinical entity. Most of the time treatment is aimed at deformity rather than function.

This paper has attempted to identify available resources on these issues. Clearly they are lacking. A nonscientific survey was added as a means of reporting on some current available services. The primary conclusion that can be drawn from it is that the needs are great. There is need of infrastructure, education, and public awareness. All of these take money. The most concerning need however was the expressed need for a societal change in attitude to over come stigma of cerebral palsy for specific and disabled persons in general. We must not lose sight of the goal for this workshop as we consider the medical needs of persons with cerebral palsy, the importance of inclusion of those persons in their society as a whole.

WHO Definition of Disability
"Any limitation in performing tasks, activities and roles to levels expected within physical and social context"

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CEREBRAL PALSY IN THE 21ST CENTURY: WHAT'S NEW?

Peter Rosenbaum MD FRCP(C)
Professor of Paediatrics, McMaster University, Co-Founder, *CanChild* Centre for Childhood Disability Research, Canada Research Chair in Childhood Disability, Director, McMaster Child Health Research Institute

Cerebral palsy (CP) has often been considered the prototype childhood 'neurodisability' [1] (Rosenbaum 2003). It is usually identified as the most common physically disabling condition seen and managed by child health professionals, with a prevalence of 2.0-2.5 per 1000 live births that has remained relatively stable over many decades [2]. In the past 40 years a number of thoughtful clinicians, researchers and epidemiologists have contributed to an ever-improving understanding of CP, in part through collaborative efforts among CP registers that have enabled researchers to discern epidemiological patterns using the very large databases that such registers make possible. Causal pathways that lead to the development of CP are increasingly being explicated. There have been important advances in understanding what CP looks like clinically, and how it can be defined and characterized. At the level of 'treatment', as will be described in this Workshop, research developments are helping to identify the benefits (and the limitations) of a host of specific interventions, using increasingly thoughtful and methodologically sound study designs and tailor-made outcome measures. In addition, a number of secular influences have encouraged both service providers and researchers to expand our conceptual horizons to consider and accommodate new thinking.

In this paper I shall offer a somewhat idiosyncratic perspective on what I see as some of the most important developments in the field of CP at the clinical and epidemiological levels. In doing so I will discuss several significant conceptual and technical factors that have allowed (or forced) us to consider CP in a new light. Thus, rather than attempt to undertake a comprehensive review of the literature and risk missing specific studies, let me offer instead a 50,000 foot overview as an opening perspective on 'cerebral palsy'.

Many of the fundamental issues and concerns about CP have been thoughtfully presented and discussed in an excellent series of papers in the Supplement to Developmental Medicine and Child Neurology (Definition and Classification of Cerebral Palsy, 2007) [3] by a number of the world leaders in CP. Several of these papers will be alluded to throughout, because any attempt by this author to improve on what was written in that excellent series of papers would be inappropriate.

DEFINITIONS OF CP – AND WHY THEY MATTER

An excellent historical perspective on cerebral palsy was provided by Morris in his 2007(a) introduction to the Supplement to DMCN referred to above [4]. There he offers readers a brief review of thinking during the less than 200 years during which this constellation of findings ('cerebral palsy', by many names) has been identified and considered as a specific clinical entity.

Moving ahead in time very quickly, we arrive at what might be considered the 'modern' era of CP, in which the Bax definition has usually been considered the classic: simply put, CP is "a disorder of posture and movement due to a defect or

lesion in the immature brain" [5]. The emphasis that was taken from this brief statement and repeated regularly by others to the present time was that CP was a 'motor disorder' (though in fairness to Bax and his colleagues there were more words in the full account of the consensus meeting he reported).

Subsequent efforts by others to refine or improve the definition have generally been subtle. These included the Mutch et al. definition of CP as "an umbrella term covering a group of non-progressive, but often changing, motor impairment syndromes secondary to lesions or anomalies of the brain arising in the early stages of development" [6]. This definition emphasized the ideas of the condition as a collection of clinical pictures, non-progressive at the level of CNS pathology but potentially evolving clinically. During this period leaders in CP research in the UK [7,8] and Western Australia [2] strove to consider how best to classify and describe the clinical dimensions of this neurodevelopmental disorder.

Building on these and related ideas, the Surveillance of Cerebral Palsy in Europe (SCPE, 2000) group endorsed the notion of CP as an umbrella term that describes permanent but not unchanging functional impairments involving a disorder of movement and/or posture and of motor function, due to a non-progressive interference, lesion, or abnormality in the immature brain [9]. They have also promoted the use of a simple classification of CP into three broad groupings based on the predominant motor manifestations (spastic, dyskinetic, and ataxic [along with the term 'mixed' when it is difficult to identify a single predominant motor pattern]). They further separated spastic CP into bilateral and unilateral, and discourage the use of the classic topographical terms 'diplegia' and 'quadriplegia'.

Before turning to the most recent effort at defining CP, it is important to emphasize a point identified by Rosenbloom [10] and Cans et al. [11] regarding the purposes for which any definition or classification might be created. It seems likely that at least some of the arguments surrounding attempts to define and classify CP concern what function(s) the definition and categorization are meant to serve. From an epidemiological (population) perspective it is essential to identify all legitimate cases, of all 'severities', but to exclude those that do not belong in the defined entity [12]. Accurate data may help us to identify temporal patterns in the appearance of the condition, perhaps associated with changes in the availability and use of health services, as Robertson et al. have so clearly shown about CP with a 30-year regional perspective [13]. Similarly, data from epidemiological studies may help us to recognize possible causal pathways [2] related to factors such as birth-weight and gestational age or the presence of multiple births as a risk factor for CP [14]. Policy-makers need to know about both incidence (new cases per year) and prevalence (how many people have 'it' at any specific time) in order to plan services for the population. On the other hand clinicians are concerned with individuals, and wish to know whether the specific person they are currently assessing may have 'it', how severe the condition is, what additional manifestations of the condition may be present, and what management strategies are important at any stage of that person's development.

It is therefore apparent, and problematic, that clinical and epidemiological definitions may be rather different to serve these different purposes. The difficulty arises when these perspectives collide. As discussed by Blair et al. [15] the epidemiological focus adopted in Western Australia includes every child who has "…presence of neurological signs of the motor impairment. These neurological signs may or may not be associated with limitations in normal activities of daily living but are likely to limit

the potential for physical performance. Those in whom the motor impairment is so minor that it may not be apparent to an untrained eye are classified as 'minimal CP'." Note that this definition makes a presumptive prognostic judgment ('…are likely to limit the potential for physical performance') despite apparently having no '…limitations in normal activities of daily living'. Many clinicians would not agree that children without any apparent functional limitations should be labeled with the term CP, even though their inclusion might serve an epidemiologist's needs. It thus becomes apparent that there can be an important mismatch between what these two groups are referring to when the term 'cerebral palsy' is discussed!

In 2007, following three years of meetings, commentary, feedback and discussion across the world, a proposed new, more detailed annotated definition of CP was published. The full definition reads: "Cerebral palsy (CP) describes a group of permanent disorders of the development of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain. The motor disorders of cerebral palsy are often accompanied by disturbances of sensation, perception, cognition, communication, and behaviour, by epilepsy, and by secondary musculoskeletal problems".[16]

As the lead author of the consensus definition I am aware of both the thinking that went into literally every word, and the areas of this statement that generated the most discussion within the committee and among critical colleagues who offered comments. Most of these latter ideas have been collated in the DMCN Supplement [3], and will not be discussed here. Rather, in this paper I wish to reflect on what I believe is, for clinicians at least, a new phase in our conception of CP. At the clinical level I hope to encourage people to see CP in a fresh and expanded light, because in so doing I believe that we will modify some of our traditional thinking and probably be able to serve children and families even more effectively than we have done until now.

Before turning to these modern perspectives it is important to comment briefly on the challenges of making a diagnosis of CP. From a clinical perspective is it obviously important to identify children whose motor (and other) development is in some way aberrant, and to offer appropriate developmental intervention as soon as problems are recognized. At the same time clinical investigations should be undertaken, and continued observation of the evolution of the child's development is essential to track the evolution of the developmental difficulties.

One challenge in making a formal 'diagnosis' of CP is the reality that, like most neurodevelopmental disorders, CP 'develops' over the early months and even years of the child's life. The early presentation of motor difficulties may include a combination of 'qualitative' and 'quantitative' signs (Rosenbaum 2006), the evolution of which may require long-term follow-up over the first few years [17]. The SCPE group supports the ascertainment of children with developmental motor difficulties at any age, but recommends that a formal assignment of the diagnosis 'cerebral palsy' only be given after the age of four years, when it is clear that the earlier signs and developmental difficulties have persisted but have not shown evidence of progression that would suggest another (usually more serious) diagnosis.[9]

Most people in the field would recognize the classic forms of CP – predominantly (80%) associated with spasticity, impaired gross (and usually fine) motor control, variously affecting one side of the body ('unilateral' CP) or both sides ('bilateral' CP), with variations in the degree of functional limitations of lower and upper

extremities. The traditional topographical descriptions of CP include unilateral CP (described by the interchangeable terms 'hemiplegia/hemiparesis/hemisyndrome'); and bilateral CP ('diplegia', referring to CP affecting the lower extremities more that the upper; and 'quadriplegia/tetraplegia' in which all four limbs are equally (and by implication more 'severely') involved). Bleck has recommended calling this distribution of CP 'total body involvement' to acknowledge the reality that this form of CP usually affects trunk and bulbar musculature, such that it has a pervasive impact on many areas of function.

The problem with these terms is that, while the classic examples of each are easily recognizable, there is simply too much imprecision at the 'edges' to be able to be precise in how to define these patterns with these classic terms. At what point does 'diplegia' merge into 'quadriplegia'? How do we classify someone with predominantly unilateral CP who, on more careful examination, has 'minor' signs on the presumed 'non-affected' side? No clear guidelines exist by which these nuanced distinctions can be made. One of the few clinical studies of inter-observer reliability of the clinical classification of CP found relatively poor agreement [18]. On the other hand when classification is made on the basis of functional characteristics the reliability is somewhat stronger [19]. Gorter et al. [20] explored the relationships between topographical distributions of CP, or the predominant motor 'types', and functional classifications made with the valid and reliable Gross Motor Function Classification System (GMFCS) [21,22]. They showed clearly that traditional assumptions about these topographical-functional relationships were not nearly as sound as has usually been assumed. For example, children with 'diplegic' CP were found in GMFCS Levels I through IV; and while the majority of children with 'hemiplegia' (88%) were classified in GMFCS Level I there were 12% in Levels II and III. They also argued that in the absence of more reliable classifications of CP than are afforded by the GMFCS, GMFCS classification should be used to describe the function of people with CP, while the other topographical terms could not be expected to imply functional status.

TWENTY-FIRST CENTURY THEMES

CP as a 'Developmental' Disability

The first theme that I want to discuss is that of CP as a 'developmental' disability and not simply a 'motor' disorder. For me one of the significant points of emphasis in the 2007 revised definition of CP was the identification of CP as '…disorders of the *development* of movement and posture' (my emphasis). In highlighting the developmental element of this 'neurodevelopmental disability' more explicitly than had heretofore been done, the new definition encourages people to recognize that CP affects the trajectories of the lives of developing beings from a very young age.

I believe that this concept has at least two implications. The first is that it is the *development* of movement and postural that is affected by the underlying motor disorders, meaning that whatever motor learning young children with CP are doing is likely to be in some ways atypical (I personally discourage people from using the imprecise and value-laden term 'abnormal'). The corollary of this notion is more subtle, but concerns the potential impact of disordered motor development on other aspects of child development. One need only think of the amount and range of exploratory activity of a typical motor-minded two-year old to recognize how much a child with CP and motor restrictions may grow up 'deprived' as a consequence of limited motor (and by extension other early life) experience. Thinking of CP as a

potential cause of childhood 'deprivation' may seem odd to many people, but considered in the light the challenge it poses to typical developmental I believe that framing CP in this way is conceptually logical.

One of the therapeutic implications of this perspective on motor limitations is to consider the potential value and impact of the early and liberal use of 'augmented (powered) mobility', which both overcomes the mobility restrictions and can be associated with powerful changes in developmental function. Butler's [23] excellent studies illustrated how 'empowering' children 23 to 38 months of age led quickly to enormous changes in self-initiated interaction with objects, spatial exploration and communication with care-giver. She also noted that these children, who were experiencing very significant restrictions in mobility, seemed to 'discover' and attempt to use their limited capacity more actively.

In considering the formulation of CP as a developmental disorder, one of the significant changes to the classic definition of the condition is the addition of the second sentence: "The motor disorders of cerebral palsy are often accompanied by disturbances of sensation, perception, cognition, communication, and behaviour, by epilepsy, and by secondary musculoskeletal problems". The formal identification of this constellation of potential associated neurodevelopmental challenges serves to remind people that while the hallmark characteristic of CP is a "…permanent disorder of the development of movement and posture, causing activity limitation…" other aspects of neurological function may also be impaired and may create functional limitations. In fact one might easily argue that at times many of these so-called 'secondary' problems are more functionally important than the motor limitations by which CP has traditionally been characterized.

These issues are important both philosophically and in practical terms. From a developmental perspective the functional motor difficulties of a child with CP will almost always present at a younger age than problems in language, cognition or behaviour. It is then too easy to assume that these later-emerging developmental difficulties are 'secondary' to the motor problems, when in fact they may co-vary as additional manifestations of the underlying "nonprogressive disturbances that occurred in the developing fetal or infant brain". At the same time, as discussed above, some of these associated developmental challenges may indeed literally be 'secondary' to the deprivation and related challenges imposed by mobility restrictions, or to the consequences of functional impairments (e.g., contractures secondary to limited range of joint motion associated with spasticity). Thus from a clinical perspective it behooves thoughtful service providers to identify all the developmental challenges that a child may experience (including those that emerge over time) without automatically assuming that these are either inevitable or 'secondary' difficulties. In practical terms they all need to be addressed, though it is possible that some may indeed be preventable (as suggested by Butler's work [23]).

The International Classification of Functioning, Health and Disability (ICF)

In 2001 the World Health Organization published a revision and expansion of its 1980 International Classification of Impairment, Disability and Handicap [24]. The new framework [25] incorporates concepts that include the structural underpinnings of a condition (originally 'impairments', now collectively labelled 'body structure and function'); the impact of these impairments on 'activity' (what people can do and restrictions thereof); and the idea of 'participation' (engagement in life, and possible restrictions in this aspect of life). In addition, the contextual elements of 'personal

factors' and 'environment' have been added, to reflect the reality that within a social model of disability these latter factors, outside the person, can be important contributors to whether or not a problem with body structure and function becomes a 'disabling' challenge.

For people working with youngsters with CP these concepts give us 'permission' to think about 'treatment' outside the confines of impairments of body structure and function implied by the ICF framework [26,27]. In many therapeutic traditions it was assumed (usually implicitly rather than formally) that one needed to remediate the basic problems of CP – e.g., excess muscle tone, reflex abnormalities, obligatory motor patterns, etc. – if the person were to achieve 'normal' function. In some traditions children were actively discouraged from standing and waking 'abnormally', and as a consequence were perhaps restricted not only by their impairments by but our beliefs about how function should be achieved!

The 2007 revised definition of CP [16] speaks of 'activity limitation', with the clear identification of activity as a valued aspect of child development. Nowhere are words like 'normal' or 'typical' used or even implied. Rather, this version of the definition identifies the *impact* of the underlying impairments on 'activity'. This idea contrasts importantly with that developed by the Western Australia group [15], in which, for epidemiological purposes, even the presence of neurological signs without limitations in function is enough to qualify someone for inclusion under the CP label.

The ICF appears to have considerable heuristic value and seems to have caught on much more extensively, in a few years, than the ICIDH ever did. Among the challenges posed by thinking about 'activity' (what people can do) and 'performance' (what people actually do) is the need to recognize the essential difference between these related ideas. To these concepts has now been added the notion of 'capability' – blending capacity with opportunity and inclination to perform the activity [28]. Why are these ideas important? In clinical assessments we often try to elicit best abilities ('capacity') as an indication of what might be possible for that person, and what we might want to focus on in treatment programs. One need only think about how we assess the mobility of a child with CP in the clinic – removing physical obstacles, watching them move on smooth floors, providing lots of verbal encouragement, etc. However, we may not think to take account of whether the child actually wants (and is encouraged) to do those activities we see that they have the capacity to perform during an assessment. We can too easily assume that this is what the child ought to do 'out there' on crowded, slippery or uneven surfaces – and be disappointed by reports of less functional 'performance' than we know to be within the child's repertoire. These three related but distinct concepts regarding what people can and do achieve represent an important new way to think about how we evaluate children and what they actually do, and how we help them achieve their goals rather than ours.

Modern Developments in CP

In the past quarter century a number of technical as well as clinical and research developments have had a considerable impact on our emerging understanding of CP. These will be alluded to briefly to remind people of the fluid and still-developing nature of this fascinating story.

The traditional belief was that perinatal difficulties were the 'cause' of CP. One might be inclined (perhaps unfairly) to lay at least some of the responsibility for this idea at the feet of the 'father' of cerebral palsy, John Little, whose 1862 signal paper

on the subject [29] was entitled "On the incidence of abnormal parturition, difficult labour, premature birth and asphyxia neonatorum on the mental and physical condition of the child, especially in relation to deformities". While he may have been partly correct in ascribing CP to perinatal misadventure (especially before the era of modern obstetrical care), Little can, of course, be forgiven for making a classic error in logic by assuming that because the child's difficulties *followed* a difficult birth they were *caused* by it ('post hoc, ergo propter hoc'). Modern imaging techniques are making it possible to characterize the nature and location of the brain impairments that underlie CP, and with increasing precision to link the timing and location of specific impairments to particular phases of intrauterine development [30]. It has therefore become possible to recognize that many people with CP have impairments in brain structure (and almost certainly in brain function [31]) prior to their delivery. In fact it is likely that a significant proportion of people who have CP experience perinatal difficulties *secondary* to pre-existing CNS difficulties that make perinatal and immediate adaptations challenging. These new insights into the timing of the insults to brain formation are important to parents, to the accoucheurs who have traditionally been blamed for obstetrical failures, and to neurobiologists seeking to understand brain ontogeny in order to prevent the developmental problems that underlie CP and related disabilities from occurring.

Another important 'technical' development in the field has been the creation of purpose-built classification tools and standardized clinical assessments that enable people to speak the same language and evaluate our interventions using consistent and valid instruments [32,33]. Furthermore, these recent developments have been created to assess *function* (with 'criterion-referenced' measures) rather than deviations from 'normal' (the latter assessed with so-called 'normative' measures). Classifications of gross motor function (Gross Motor Function Classification System [21, 22]) and manual abilities (Manual Abilities Classification System [34]) among individuals with CP have enabled people to communicate with each other, and with parents, who appear to find these 'levels' of function both acceptable and useful in understanding their child's status [35-37]. Morris recommends using the GMFCS levels as one of the frameworks for planning orthotic management of children and youth with CP [38,39]. Colleagues in Munich [40] (Heinen and Berwick) and Australia (Graham, personal communication 2007) have also begun to outline approaches to the management of motor function by GMFCS levels.

Without discussing specific measures in detail, it is important to note that the application of measurement science to the creation and validation of clinical measures has helped to move the field forward considerably. Developments have included the creation and appropriate validation of measures specifically developed to assess change (e.g., PEDI [41], GMFM [42], QUEST [43], and GMPM [44,45]). These tools are criterion-referenced instruments that assess function against preset criteria, rather than misapplying norm-referenced measures that almost always fail to detect (real) change by virtue of their structural and functional properties [46]. New measures have made it possible to assess whether, and to what extent, change happens 'naturally' [47] or as a function of interventions [48].

Another research advance has been the undertaking of prospective longitudinal studies in which it is possible to observe within-person change over time in various aspects of function and well-being (e.g., gross motor function [47,49] or quality of life [50,51]). Unlike cross-sectional studies which, in effect, 'join the dots' of the functional status of different individuals at different times ('between-subject' variability), this

research design makes it possible to observe how the same individuals develop and change over time ('within-person' variability). While more expensive in time and effort, these perspectives are essential and are adding new levels of understanding to the field.

As but one illustration of how prospective longitudinal studies can be useful, the extensive research from *CanChild's* Motor Measures Group has enabled us to learn about patterns of gross motor development in children and youth with CP by GMFCS levels, and to offer an evidence-informed prognosis for gross motor development into the early adult years [47,49]. This was made possible by studies that followed and assessed the same large number of children and youth over almost 10 years, plotting their individual 'motor growth curves' and aggregating these curves with appropriate statistical modelling techniques. The findings represent patterns of motor development associated with current mixed modalities of functional intervention, and may well change (and hopefully improve) as our therapeutic techniques become more refined. However, at a minimum we are in a position to offer parents evidence-based answers to questions such as "Will our child walk?".

CP as a life-long condition – transition to adulthood and what it means for child health

One concern about CP that has been discussed for years but seems finally to be gaining serious traction is the notion of 'transition to adulthood', and the recognition that there are increasing numbers of adults with this 'childhood' disorder [52]. Like so many neurodevelopmental conditions CP presents very early in life, and has traditionally been managed within the child health orbit. Parents report that when their children reach 18 years they and their youth 'fall off the cliff' – a metaphor that has been heard in many countries. We are at last starting to acknowledge and address the reality that CP does not disappear when children outgrow children's services. People have begun to explore what happens to the lives of adults with CP. (One should add as well that mortality in CP is relatively low in all but those people most complexly affected, and even in this group mortality is decreasing [53]. This means that premature death in childhood and adolescence is not the anticipated end for most children with CP born in the past two decades.)

A detailed discussion of these important issues is beyond the scope of this report. Suffice to say that there are enormous opportunities to develop effective transition services, and to undertake research and careful delineation of the life circumstances and life quality of adults with CP who may too easily be ignored by the world of adult services to whom the life experiences of people with CP are alien. At the same time it is very important to note that opportunities for gainful employment of adults with CP remain significantly limited. There also appears to be evidence of 'premature' loss of motor function in adults who have previously been more capable [54]. Of course, findings like these latter observations may, to some extent, represent self-fulfilling prophecies about what has always been assumed was the fate of adults with CP. It is quite reasonable to ask whether, with more effective attention and preparation early in the transition years (e.g., focusing on fitness and 'participation'), careful planning toward adult independence, and more community-based opportunities and services the current picture can be enhanced considerably.

The reason for raising these issues in this essay is simply to argue that those of us who work in the child and youth field need to take a much longer view of CP than has been our tradition with our focus on childhood. There are almost certainly important

lessons to be learned from adults who have received our services as children, and equally important opportunities for 'feed-forward' of these experiences to parents of young children with CP, youth with CP, and professionals in our many fields regarding how best to offer advice, interventions and supports to young children starting on the CP 'journey' though life.

CP as a condition that affects a whole family

Another 'modern' concept about CP and other neurodevelopmental disabilities is the recognition that these conditions usually have a significant impact on families as well as children. This in turn leads to a focus on the needs and well-being of families in what is usually referred to as 'family-centred service' [55]. One of the many reasons to be concerned about the well-being of parents is that there is documented evidence that, as a group, their physical and mental health is compromised [56-58]. There is also evidence that links 'processes' of service delivery to parental 'outcomes' (satisfaction with services, stress and mental health) [59]. Thus, the way that services are organized and delivered to families of children with CP actually matters to parent well-being, with the obvious assumption that this in turn matters to children!

'Quality of Life' (QoL) and CP – what does this concept mean in CP?

Finally, it is important to comment briefly on a ubiquitous concept – 'quality of life' – that has begun to move from a popular buzz phrase used in the community to an evolving set of concepts in health care that have both philosophical and academic importance. Like many of the ideas discussed in this paper, it is challenging to define quality of life precisely. One definition that is often cited states that QoL is the "individual's perceptions of their position in life in the context of the culture and value systems in which they live, and in relation to their goals, expectations, and concerns" [60]. The idea of situating a person's quality of life status within their culture and values speaks to the importance of understanding people's realities from their internal vantage point rather than exclusively from the perspective of outside observers (judges).

There are many reasons why it is usually assumed that the QoL of people with disabilities *must* be poorer that that of able-bodied citizens [61]. These include the assumption, on the part of many outside observers that if they were in the predicament they perceive to be experienced by people with CP their lives would be diminished in some way. It is also the case that in many research studies 'quality of life' is measured with norm-referenced tools, developed to encompass the whole population, that ask about or assess functional status with able-bodied function as the standard (e.g., HUI3 [62], PedsQL [63]). Since by definition people with CP have "…permanent disorders of the development of movement and posture, causing activity limitation…" it should be obvious that by the usual functional yardsticks used in these evaluations people with CP will score lower than people without CP!

The reason to bring this concept into focus in a paper primarily about cerebral palsy is to remind us to consider the broad goals of our treatments, therapies, interventions, management strategies – whatever words we use for the roles and activities of professional service providers. At the end of the day I believe that our overarching goal in working with young people with cerebral palsy ought to be to enhance their life quality. This of course includes efforts to promote functional well-being and capability using the best of our impairment-directed interventions, and to prevent the secondary consequences of functional limitations. These secondary consequences may include physical challenges associated with issues such as limitations in strength

and fitness; psychological difficulties associated with challenges in self-actualization; and social difficulties related to limited opportunities for meaningful employment and independence in the broad community. Thus, perhaps one guiding principle in all our therapeutic initiatives ought to be to ask whether, how, and in what ways these efforts will contribute to supporting or enhancing the quality of life of the people with whom we work.

It is exciting to realize that we are beginning to learn that the 'quality of life' of people with 'disabilities' is not as dismal as has often been assumed [64], nor is it inversely proportional to the level of externally visible disability. Both Dickenson et al. [65] and Rosenbaum et al. [66] explored 'quality of life' using measures that ask about aspects of people's inner lives rather than their functional status. When this lens is used to explore how people view their life situation, the findings are very different from the way that QoL has usually been assessed and reported [61]. This reframing of the nature and quality of the lives of people with 'disabilities' is encouraging in itself; it also challenges those of us privileged to work with these people and their families to consider how we think and act in offering advice about the development of children and youth with CP.

TO SUMMARIZE, in the 21st century there are increasing opportunities to (re)consider cerebral palsy as a developmental condition with clear biomedical underpinnings, affecting children and their families across their life course, requiring the best of both our biomedical therapies and our guidance to families and young people to make the most of their lives with whatever abilities they have and can develop. I strongly believe that the glass is currently at least half full, and that these new perspectives, technical advances and conceptual thinking are empowering us to continue the process of filling the glass completely!

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CLASSIFICATION OF GAIT IN PERSONS WITH CEREBRAL PALSY

Roy B. Davis, III, Ph.D.
Director, Motion Analysis Laboratory, Shriners Hospitals for Children
& Professor of Bioengineering, Clemson University, Greenville, USA

"Gait Classification" refers to the assignment of a subject's overall walking pattern to one of several defined categories or types of gait. For persons with cerebral palsy (CP), sub groups of gait patterns are generally identified based on their type of cerebral palsy, e.g. spastic hemiplegia. The work in gait classification can be traced back to the 1980's when it was more-commonly referred to as "gait pattern recognition." Two contributions from that era illustrate the two approaches used in development of gait classification strategies today: groups formed qualitatively by experienced clinical practitioners [1] and groups identified based on statistical methods.[2] As these two early studies illustrate, gait classification approaches generally rely upon quantitative data provided through clinical gait analysis.[3]

Motivations to develop and use gait classification approaches in CP vary. At the very least, an effective gait classification approach can provide concise terminology to describe a patient's complex set of gait abnormalities. If used clearly and consistently, then this lexicon can facilitate communication between researchers and clinicians. Sutherland and Davids [4] concluded that an understanding of common pathological gait patterns can also "facilitate accurate and detailed analysis of the individual patient with gait abnormalities." Winters et al. [1], followed in more detail by Rodda and Graham [5], broadened significantly the clinical application of gait classification by explicitly connecting gait patterns with specific treatment recommendations. McDowell and her colleagues at the Musgrave Park Hospital (Belfast, Ireland) sidestepped this controversy with the opening statement in their recent article, "While there has been much debate as to the usefulness of classification systems for interpreting pathological gait, many clinicians still find them useful for minimising the complexity of kinematic gait data."[6]

The purpose of this report is twofold. First, the pertinent literature associated with gait classification in CP will be examined and critiqued. This will focus on the comprehensive review published by Dobson et al. [7] in 2007. Several contributions associated with gait classification in CP, not included in or published since the Dobson review, will be considered. In the second part of this report, gait abnormalities commonly seen in persons with CP will be described. This synopsis is intended as foundation to subsequent reviews and discussions pertaining to the clinical management of ambulatory persons with CP. This manuscript will conclude with a discussion regarding the challenge of gait classification in the context of a disease that presents a continuum of gait deviation and propose recommendations that might yield a more comprehensive gait classification approach.

REVIEW OF THE GAIT CLASSIFICATION LITERATURE

In 2007, Fiona Dobson along with colleagues Meg Morris, Richard Baker and Kerr Graham from the Hugh Williamson Gait Laboratory & Murdoch Children's Research Institute at the Royal Children's Hospital (Parkville, Australia) published a systematic, carefully-designed review [7] of 18 published works [1, 2, 4, 8-22] that described strategies to classify the gait of persons with CP. The studies were identified from a search of nine literature databases and included abstracts and full manuscripts published from 1966 through March, 2005. For the most part, the identified studies focused on the gait of persons with CP aged 0–18 years. Dobson's review qualified the validity of the bases of these classification schemes through an evaluation of the studies with respect to their subject selection, classification construction, psychometric properties (i.e. degree to which a particular gait classification approach can actually classify gait, for example, considerations of reliability, construct validity, discriminant validity, basis of the classification construction, criterion validity, and concurrent validity).

Adequate subject descriptions and inclusion criteria were provided in only four [1, 17, 20, 21] of the 18 studies, thereby immediately limiting the utility of the resulting gait classification algorithm. For example, it was unclear in a number of the studies if the research cohort included subjects with a history of previous orthopaedic surgery. A retrospective convenience sample method was used in five [8, 13, 16, 21, 22] of the 18 studies and not stated for the other 13 investigations. This sampling method leaves open the question of whether the study cohort adequately represented the entire population. For example, a study group comprised only of patients referred to a clinical laboratory for gait analysis and treatment planning may not adequately sample those subjects with milder CP-related gait abnormalities.

With respect to classification construction, the majority (12) of the gait classifications were based on sagittal plane kinematics (and/or kinetics) despite the availability of three-dimensional quantitative gait data in 15 studies. This simplification may limit the reliability of the classification scheme in patients with femoral and/or tibial torsion, i.e. transverse malalignment may impact sagittal plane motion.

Nine [1, 4, 9, 12, 15, 17, 18, 20, 21] of the gait classification methods relied on groups formed qualitatively by experienced clinical practitioners, described by Dobson as "qualitative pattern recognition using quantitative data." As Dobson pointed out, "A potential advantage of qualitative methods is enhanced clinical relevance as groups are partitioned using clinical reasoning and rely on identifiable patterns of movement observed by clinicians. Therefore, they may be more meaningful in terms of clinical diagnosis and treatment planning." This reviewer agrees fully with this assertion. The fundamental problem, however, with these nine "qualitative" classification approaches is the lack of quantitative information provided in each article pertaining to how the groups were determined, e.g. how were the number of groups settled upon, how were subjects assigned to groups, are the identified groups different statistically. For example, Winters et al. [1] found four gait patterns for a cohort of children with spastic hemiplegia. The magnitude of the first standard deviation associated with the mean kinematic curves (refer to figure 1 in reference [1]) suggests that additional sub groups exist within the four identified groups. Hullin et al. [9] investigated this same clinical population and found five gait patterns.

Nine of the gait classification approaches employed several different quantitative pattern recognition techniques: cluster analysis [2, 10, 11, 13, 14, 19], hidden Markov models [8], generalized dynamic neural network [22], and support vector machine [16]. A fundamental advantage of the cluster analysis technique is that it requires objective specification by the user in setting up the analysis. The challenge with cluster analyses is that they can yield different results depending on those *a priori* specifications and definitions. As Dobson et al. [7] succinctly characterized it, "Cluster solutions may impose a structure on the data set rather than finding naturally occurring groups." The other analytical techniques (hidden Markov models, neural networks, and support vector machines) are used widely in pattern recognition (e.g. analysis of speech) and machine learning (e.g. data mining, machine vision). These approaches allow the underlying pattern relationships to be established (supervised or unsupervised machine learning depending on technique) with relatively large, coherent training sets. Basically, the model is provided enough "answers" so that it can probabilistically identify a new "unknown" pattern. The utilization of these approaches in gait classification is promising, but challenged by the availability of the required training data, particularly as related to comprehensive gait patterns. The more focused effort by Zwick et al. [22] (i.e. the characterization of plantar flexor muscle function with a neural network) appears to be a more plausible approach at this time.

Information pertaining to psychometric properties provided in the studies included in the Dobson review was limited. None of the articles associated with the nine qualitative-based classification approaches provided sufficient detail in this area. Somewhat amazingly, only two of the studies [15, 18] reported on inter-rater and/or intra-rater reliability. Three of the quantitative studies [8, 14, 16] included classification cross-validation. Dobson also reported that seven studies [2, 8, 13, 14, 16, 19, 22] demonstrated construct validity and five studies [2, 8, 14, 16, 22] demonstrated discriminant validity. Her review also qualified seven of the articles [1, 10, 13-16, 22] as adequately describing clinical utility, with another eight studies [2, 4, 8, 11, 12, 17, 19, 20] providing a "limited description."

Dobson et al. [7] abstracted her analysis of the 18 classification approaches in her review as follows: "Many did not provide adequate guidelines or evidence of reliability and validity of the classification system. No single classification addressed the full magnitude or range of gait deviations in children with CP. Although gait classification in CP can be useful in clinical and research settings, the methodological limitations of many classifications restrict their clinical and research applicability."

Published subsequent to the Dobson review article, Toro et al. [23] described the application of hierarchical cluster analysis of sagittal (hip, knee, ankle) gait kinematics associated with 54 children with CP (hemiplegia (n = 25), diplegia (n = 24), quadriplegia (n = 4), monoplegia (n = 1)) and 11 typically-developing children. The investigators' goal was to objectively determine the optimal number of gait pattern clusters associated with this non-homogeneous study cohort. The investigators used the following criteria define optimal number of gait pattern clusters: 1) distinct identification of the gait pattern associated with the typically-developing children, 2) minimized hip, knee and ankle kinematic standard deviations within the clusters, and 3) for any given cluster, at least one kinematic curve (hip, knee or ankle) distinctly different from its neighboring cluster. The investigators found 13 gait pattern clusters, four that they qualified as crouch gait type, four as equinus gait type, four as an "other gait" type, and one as "normal gait" type. It is not

entirely clear that the objective analysis satisfied the first criteria as data for one typically-developing subject was associated with an "other gait" type cluster. Moreover, the investigators arrived at 13 clusters from a 14-cluster solution by eliminating one of two merged clusters, discarding what they qualified as an unstable cluster, and including a cluster identified in a 15-cluster solution. These adjustments illustrate the challenge associated with defining objectively the number of clusters within non-homogeneous collection of gait patterns.

Three publications were identified that exercised the Winters [1] classification approach. Riad et al. [24] considered 112 pediatric patients with spastic hemiplegia CP (without prior orthopaedic surgery). Twenty-six patients (23%) of 112 could not be classified in any of the four Winters classifications. The gait deviations in these patients were slight relative to a laboratory normative reference, but were still qualified by the investigators as abnormal. The authors proposed that a "Group 0" be added to the Winters 4-type classification approach to accommodate this additional, mildly-involved group.

McDowell et al. [6] also investigated the validity of the Winters [1] classification approach through the examination of 91 pediatric subjects with spastic hemiplegia CP recruited from the community (30/91 with prior orthopaedic surgery) and 49 typically-developing children. Consistent with Riad et al. [24], McDowell and her colleagues found that the Winters classification approach failed to classify the most mildly-impaired children with CP (38/91, 30/61 children with no previous surgical history, 8/30 children with previous surgical history). The classification rules utilized by McDowell differ from those employed by Riad, although both papers sought to employ the Winters classification system. McDowell spoke to the challenge of creating explicit quantitative classification rules based on the somewhat ambiguous description provided by Winters et al. [1], a criticism that Dobson et al. [7] had of all of the qualitative CP gait classification schemes.

The last evaluation of the Winters [1] CP gait classification method was provided by Dobson et al. [25]. In this study, 16 experienced clinicians (from six established gait laboratories) sought to classify 34 children with spastic hemiplegia CP into one of the four Winters types based on video and/or quantitative gait data and limited physical examination values (even though physical examination data were not incorporated into the Winters classification algorithm). Although weighted kappa scores were sufficiently high for the investigators to conclude that there was acceptable overall clinician agreement using the Winters classification tool, the authors also reported actual agreement values as low as 32% (when using quantitative gait data and limited physical examination values) and 35% (when using video records and limited physical examination values). Dobson and her colleagues recommend that the Winters gait classification approach be refined to define more explicitly the groups and to possibly include transverse and coronal gait kinematics.

COMMON GAIT ABNORMALITIES SEEN IN PERSONS WITH CEREBRAL PALSY

For detailed descriptions of the gait of the typically-developed individual, including terminology associated with gait events and phases of the gait cycle (e.g. initial contact, the three foot/ankle rockers) and definitions associated with segment and joint kinematics and intersegmental kinetics, readers are referred to the reference texts by Perry [26] and Rose and Gamble [27]. The Perry text is also an excellent resource

on pathological gait. Davids [28] provides a description of common foot deformity seen in individuals with CP and its consequence with respect to gait, a topic that is not included here.

Despite the significant limitations identified in the review by Dobson et al. [7], two of the "qualitative" CP gait classification approaches (Winters et al. [1], Sutherland and Davids [4]) cited in that review are commonly-referenced clinically and provide a starting point for describing routinely-seen gait deviations in CP. Colleagues from Melbourne (Rodda et al. [5] and later Rodda et al. [15]) build on these earlier classifications and provide a framework for this presentation. The gait patterns described in all of these articles include a consideration of simultaneous changes in ankle, knee, hip, and pelvic sagittal plane motion. This complete lower extremity presentation allows an opportunity to appreciate the dynamic relationships between joints and segments.

Spastic Hemiplegia CP (refer to Figure 1)

Type 0 (described by Riad et al. [24]) – milder than Winters Type 1, ankle dorsiflexion in mid swing with excessive ankle plantar flexion in terminal swing, increased knee flexion in terminal swing and at initial contact associated with a toe or foot flat landing at initial contact

Winters Type I – excessive ankle plantar flexion throughout swing (drop foot), increased knee flexion in terminal swing and at initial contact associated with a toe or foot flat landing at initial contact; commonly-seen compensations for foot drop include a vault (premature heel rise) on the sound side and/or lower limb circumduction on the involved side

Winters Type II – excessive ankle plantar flexion over the entire gait cycle, full knee extension in midstance (Melbourne Type 2a) or knee hyperextension (recurvatum) in midstance (Melbourne Type 2b), increased knee flexion in terminal swing and at initial contact associated with a toe or foot flat landing at initial contact (seen in the data presented by Riad et al. [24])

Winters Type III – excessive ankle plantar flexion over the entire gait cycle, increased knee flexion in terminal swing and at initial contact, full knee extension or recurvatum in midstance, reduced peak knee flexion in swing (resulting in reduced knee range of motion over the gait cycle)

Winters Type IV – excessive ankle plantar flexion over the entire gait cycle, increased knee flexion over entire gait cycle, reduced knee range of motion over the gait cycle, lack of full hip extension in terminal stance, increased pelvic lordosis in terminal stance and pre swing (resulting in increased pelvic range of motion over the gait cycle, "single bump" pattern [29])

Spastic Diplegia CP (refer to Figure 2)

Melbourne Group I, True Equinus – excessive ankle plantar flexion over the majority of the gait cycle (with possible "inverted second rocker," refer to the description presented below), increased knee flexion in terminal swing and at initial contact, full knee extension (or mild recurvatum) in mid stance, full hip extension in terminal stance, anterior pelvic tilt with low amplitude bi-modal ("double bump") pattern [29]

Melbourne Group II, Jump Gait – excessive ankle plantar flexion over the majority of the gait cycle (with possible "inverted second rocker"), significantly increased knee flexion in terminal swing and at initial contact, excessive knee flexion in mid stance, knee range of motion may be reduced, lack of full hip extension in terminal stance, hip flexion bias overall, anterior pelvic tilt with double bump pattern

Melbourne Group III, Apparent Equinus – ankle range of motion over the entire gait cycle within normal limits but with a "flat second rocker" indicative of limited ankle motion during single support, knee and hip pattern similar to Jump Gait, distinguished from True Equinus and Jump Gait by the neutral or dorsiflexed ankle angle, i.e. a toe-toe foot contact pattern misleads the observer to conclude ankle equinus, anterior pelvic tilt with double bump pattern

Melbourne Group IV, Crouch Gait – peak dorsiflexion in stance elevated and delayed until opposite initial contact (as described by Sutherland) or excessive ankle dorsiflexion over the entire gait cycle (shown in the Melbourne data, Rodda et al. [15]), excessive knee flexion over the entire gait cycle, reduced knee range of motion, lack of full hip extension in terminal stance, hip flexion bias overall, normal to posterior pelvic tilt with double bump pattern.

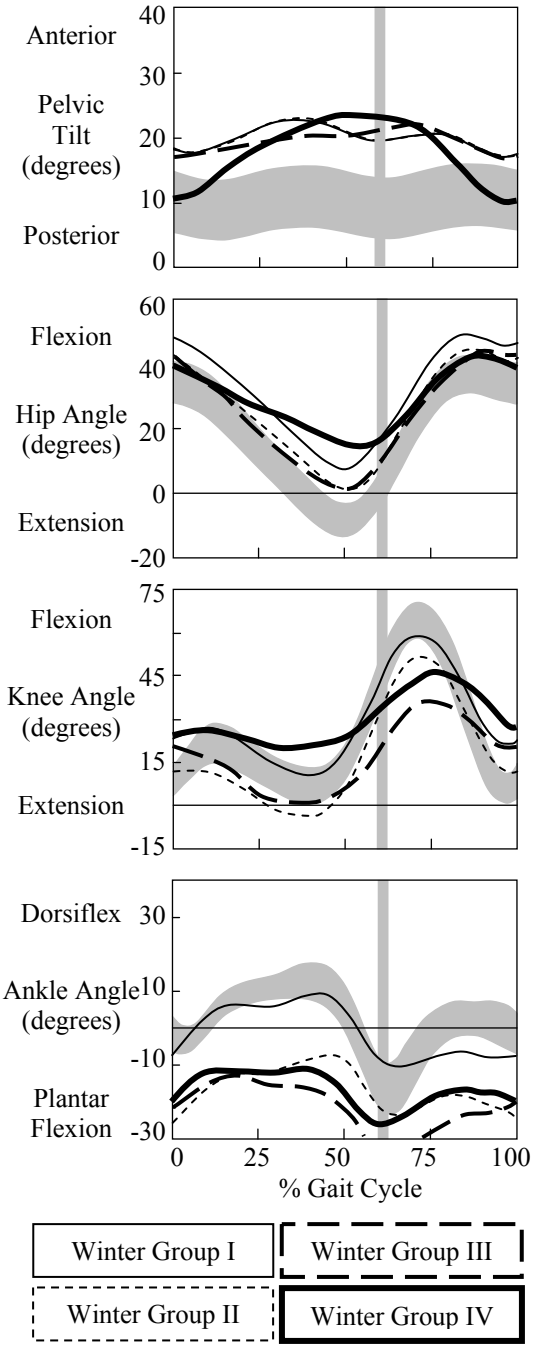

Figure 1. Mean kinematics associated with Winter Groups I-IV for spastic hemiplegia CP gait. Approximated from data presented in Winter et al. [1]. The gray band represents the first standard deviation about the mean for the laboratory normative reference (Shriners Hospitals for Children, Greenville).

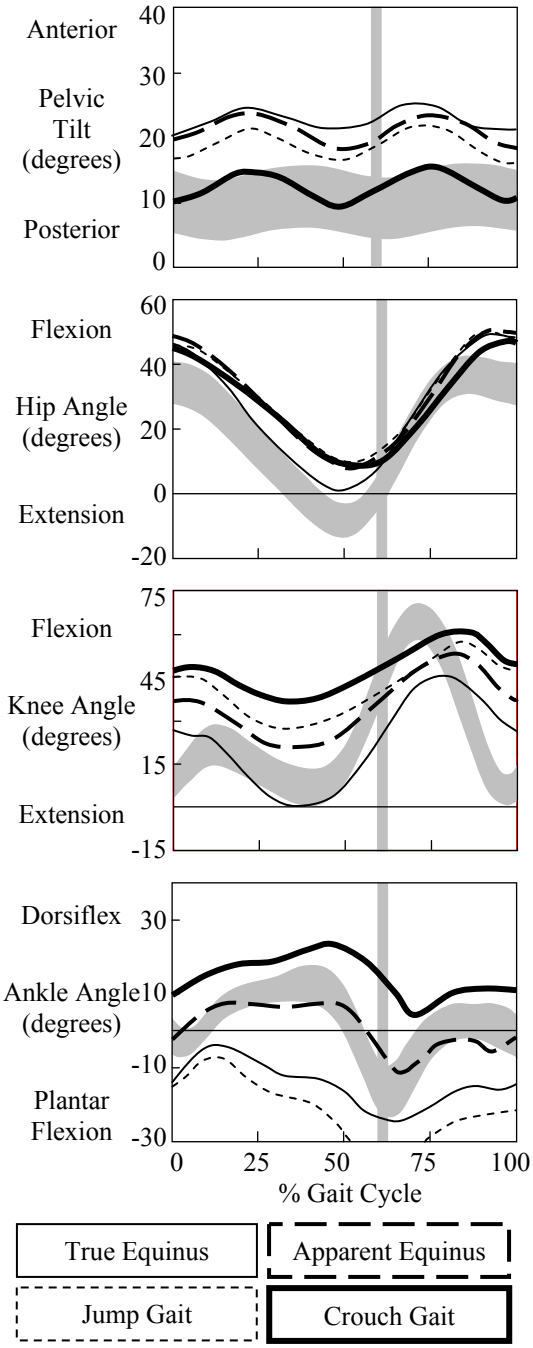

Figure 2. Mean kinematics associated with the Melbourne gait classes for spastic diplegia CP gait. Approximated from data presented in Rodda et al. [15].
The gray band represents the first standard deviation about the mean for the laboratory normative reference (Shriners Hospitals for Children, Greenville).

Additional Kinematic and Kinetic Details

First or heel rocker (heel contact at initial contact) is commonly absent in the gait of persons with CP. More common is a forefoot/toe or foot flat pattern at initial contact. With respect to sagittal plane ankle kinematics, immediately following initial contact (where the ankle can be excessively plantar flexed), the ankle begins to dorsiflex and continues to dorsiflex in loading response. This brief dorsiflexion wave is followed in mid stance by a plantar flexing motion that can continue for the remainder of stance. This is referred to variously as "inverted second rocker" or "disrupted second rocker." Alternatively, this plantar flexion wave may end, to be replaced by a dorsiflexing motion, creating a bimodal ankle kinematic curve (Figure 3).

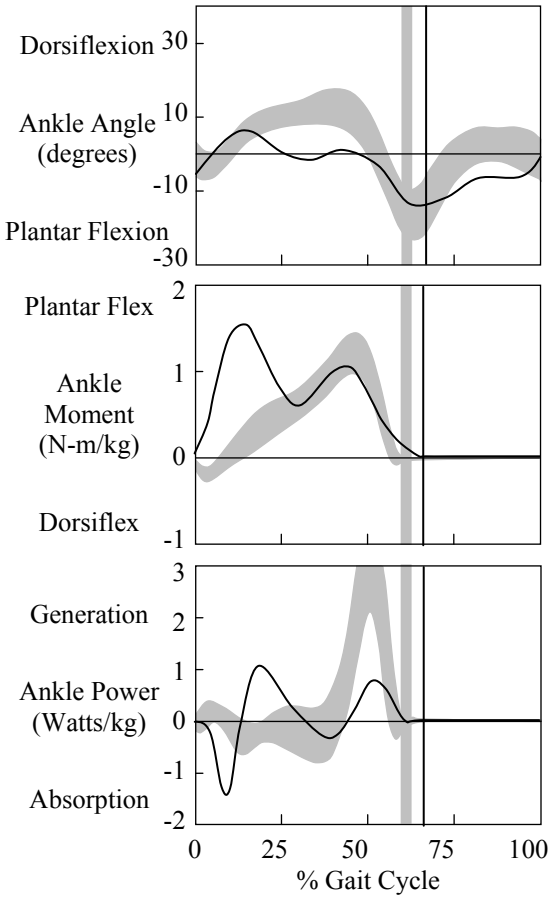

Figure 3. Illustration of inverted second rocker kinematics with associated deviations in ankle kinetics. The gray band represents the first standard deviation about the mean for the laboratory normative reference (Shriners Hospitals for Children, Greenville).

There are changes in ankle kinetics associated with the absence of first rocker as well. In cases of mild to moderate equinus, a forefoot landing at initial contact causes the ground reaction point of application (or center of pressure) to be shifted well in front of the ankle center. This results in an elevated internal ankle plantar flexor moment in mid stance with possibly a bimodal pattern (Figure 3).[29, 30] Premature ankle power generation is seen with this kinetic response as well as reduction in ankle power generation in terminal stance (Figure 3). In cases of severe equinus, the ankle position shortens the moment arm between the center of pressure and the ankle center, thereby reducing the internal ankle plantar flexor moment in mid stance.

In cases of excessive knee flexion in single support (i.e. crouch gait), an elevated internal knee extensor moment can result. If the subject's trunk is leaning forward, the internal knee extensor moment may be reduced and the internal hip extensor moment may be elevated to support the excessive knee flexion.

Commonly associated with pathological hip involvement (motor control, spasticity) is a reduction and possible delay in hip power generation during the stance-to-swing transition.

In addition to the sagittal plane kinematic deviations associated with spastic hemiplegia CP described above, transverse gait deviations may include pelvic retraction/protraction,

internal hip rotation secondary to femoral anteversion and internal foot progression secondary to femoral anteversion and/or tibial torsion. Note that internal hip rotation with simultaneous knee and hip flexion may present as hip adduction in a coronal plane view of the walking subject. Moreover, in a coronal plane view of the walking subject, pelvic retraction may "hide" internal hip rotation on the retracted side while "falsely" identifying or exaggerating internal hip rotation on the protracted side (i.e. misled by internal knee progression and/or internal foot progression). These transverse plane gait deviations are also seen bilaterally (often asymmetrically) in persons with spastic diplegia CP as well and are best sorted out through quantitative gait analysis.

CONCLUSION – GAIT CLASSIFICATION IN CP

Motivations in support of the development of an accurate and reliable gait classification system for persons with CP include: to assist in diagnosis, clinical decision-making and communication.[7] Perspectives on the use of gait classification in clinical decision-making in CP varies. Dr. James Gage [1] and Professor Kerr Graham [5, 15] and their respective colleagues offered explicit guidance as to how specific groups of patients with gait abnormalities associated with spastic hemiplegia CP and spastic diplegia CP could be managed clinically. Drs. David Sutherland and Jon Davids [4] qualify the clinical application of their observed patterns more conservatively, "It is not the intention of this paper to suggest that every patient with spastic diplegic-type cerebral palsy can be conveniently classified into one of the four groups described. Rather, it should be recognized that although there are common types of pathological gait, a patient may often exhibit combinations of the primary patterns. Each patient is an individual and should be evaluated as such. Careful attention to the detailed analysis of the patient's gait, in conjunction with an understanding of the most common pathologic patterns, should lead to an individualized treatment plan." At the Seventh Annual East Coast Clinical Gait Laboratory Conference in Richmond, Virginia, USA in 1991, Dr. Murali Kadaba (then at Helen Hayes Hospital, West Haverstraw, New York) presented pattern recognition research findings associated with the gait of 30 patients with CP. Following the presentation, Dr. Jacquelyn Perry (Rancho Los Amigos Hospital, Downey, California) came to the floor and commented, "30 patients, 30 patterns." Dr. Perry's succinct statement captures the fundamental challenge of placing a patient's gait pattern into one of several groups. The variance in neuromuscular deficits associated with cerebral palsy results in a continuum of ambulatory impairment associated with a broad range of primary gait abnormalities (type, magnitude) and secondary and compensatory deviations.

All of these clinicians have advocated the use of quantitative gait analysis in decision-making and treatment planning in this clinical population, e.g. Novacheck and Gage [31]. Quantitative gait analysis produces an abundance of gait-related information which can be extremely challenging to interpret, particularly in cases where the complex three-dimensional gait deviations are significantly different from normative references. McDowell et al. [6] pointed to the utility of gait classification as "minimising the complexity of kinematic gait data." In this sense, a "whole-body" gait classification strategy may provide a framework for the interpretation of this data, a way of

"organizing" the complexity of the gait data by promoting not only a joint-by-joint analysis, but also the examination of the relationships between anatomical levels.

But a consideration of gait kinematic patterns alone is not sufficient. Hullin and his colleagues in Edinburgh make this point in their proposed gait classification approach that extends the analysis of kinematic patterns by exploring the underlying kinetic relationships associated with the identified kinematics. A more comprehensive understanding of the underlying biomechanical relationships between particular kinematic and kinetic variables may be the key in more effective use of quantitative gait data for clinical decision-making and treatment planning.

For example, factors often included in consideration of the surgical transfer of the rectus femoris muscle in children with CP are the magnitude and timing of peak knee flexion in swing, shape of knee flexion wave in early swing, and the electromyographic (EMG) activity of the rectus femoris muscle in swing.[32-36] Drs. Perry, Sutherland and Gage appreciated the relationship between a pathological muscle force (thought to be associated with a spastic reflex to the quick stretch of the muscle) and a kinematic deficit (insufficient magnitude and/or timing of peak knee flexion in swing). These factors (magnitude and timing of peak knee flexion in swing, shape of knee flexion wave in early swing, and the EMG activity of the rectus femoris muscle in swing) represent a specific "pattern of treatment indicators" and serve to illustrate an approach that may prove more useful than the "whole limb" gait classification approaches included in the Dobson review. The clinical and biomechanical intuition of these orthopaedists provided a starting point in identifying the pattern of indicators associated with rectus femoris transfer. Biomechanical gait simulations point to other potential indicators as well. For example, the modeling work of Piazza and Delp [37] highlighted the possible influence of the hip flexors at toe-off on swing phase knee flexion as well as simulating the impediment to swing phase knee flexion associated with rectus femoris muscle overactivity.

A gait classification system can offer a framework for organizing the complex data associated with the walking impairment of a patient with CP. An understanding of the kinematic and kinetic relationships associated with the common patterns can augment the interpretation of an individual patient's data. Any future gait classification algorithm should integrate gait kinematics (including all three planes of motion and trunk kinematics as well as lower extremity kinematics), kinetics, and EMG with clinical examination measures and observational gait qualifiers. The investigator(s) must provide a complete description of the gait model used in processing the gait data, explicit group or type criteria, and demonstrated internal/external validity. Comprehensive gait analysis also requires that the underlying relationships between specific clinical examination, kinematic, kinetic, and EMG parameters be identified and considered in the context of reliable patterns of treatment indications and contraindications.

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RECENT DEVELOPMENTS IN PHYSIOTHERAPY FOR CHILDREN WITH CEREBRAL PALSY: WHAT IS THE EVIDENCE?

Adrienne Harvey PT PhD
McMaster Child Health Research Institute Post-Doctoral Fellow, Canada
Physiotherapist Hugh Williamson Gait Laboratory, Australia
&
Liz Martin PT
Physiotherapist Belmore School, Yooralla, Australia

Physiotherapy has been a mainstay of the rehabilitation management of children with cerebral palsy (CP) for decades[1]. There are many therapy approaches to the management of CP however despite some attempts to evaluate effectiveness of therapy interventions there are still many questions left unanswered. Many agree that therapy is beneficial for the child with CP, however there is still limited evidence on which to base clinical practice[2].

For the child with CP the main aim of therapy should be to improve their quality of life with three general aims of: 1) increasing or improving the skill repertoire, 2) maintaining functional level and 3) the general management and minimization of contractures and deformities[2]. Common interventions usually address; the reduction of the effects of abnormal muscle/postural tone, musculoskeletal problems (muscle weakness and muscle length), working for activity in a functional context and providing opportunities for practice[2].

Activity-based approaches aim to prevent secondary musculoskeletal impairments and maximise physical functioning, foster cognitive, social and emotional development of the child and develop, maintain and perhaps restore neural structures and pathways[1].

Two components of physical function are muscle strength and cardio-respiratory fitness and limitations in these are a major problem affecting the function and health of children with CP[3]. Impairments such as weakness, spasticity and balance problems make it difficult for children with CP to participate in sport and activities at a level of intensity sufficient to develop and maintain normal physical fitness levels[3].

There has been a resurgence in the late 1990's of treatments based on perspectives of motor control in physiotherapy management[4]. The concern in the past has been that performing strengthening exercises to address muscle weakness would increase spasticity and produce abnormal movement patterns, however this concern appears to be unfounded[3; 4]. Strengthening has been a popular treatment in more recent times and there has been an increase in the number of studies examining strengthening in the past 10 years. Another area that has increased in popularity is supported treadmill training which is designed to train walking.

A common treatment approach that has been used in the past and is still used is Neurodevelopmental Treatment (NDT), also known as the Bobath approach. This is a therapy approach originating from the work of Berta and Karel Bobath, and aims to facilitate normal motor development and function and to prevent development of secondary impairments due to contractures and deformities focusing on sensorimotor components[5]. The approach has changed over the years to incorporate carryover into activities of daily life and preparing for specific functional tasks. This makes it a little harder to define, especially when used in conjunction with other techniques[6].

Because there is a range of interventions that a physiotherapist might use to treat a child with CP, it is difficult to assess physiotherapy overall. Research is best directed to examining the effectiveness of individual specific types of therapy or exercise. Other recent reviews of the effectiveness of passive stretching[7], conductive education[8], aquatic interventions[9], casting[10] and static weight-bearing exercises[11] have all shown the low levels of evidence for these interventions and the need for further studies with improved methodology. As well as individual treatment interventions, the dosage of physiotherapy generally impacts its effectiveness. A small body of literature has attempted to answer the question of what is the optimal intensity and duration of physiotherapy intervention that is most beneficial for the child with CP.

A review of changes in the scientific quality of therapeutic motor interventions studies on children with CP over the decade 1990-2001 showed an increase in intensity of treatment, increase in number of home-based treatment and a definite improvement in methodological quality with higher levels of evidence seen[12]. This review was conducted to update the literature on physiotherapy interventions for children with CP.

The aim of this review was to identify the common conventional physiotherapy interventions utilised in the past 10 years and to critically appraise the evidence of these. The focus was on recent trends in physiotherapy management and on those interventions relevant to orthotic management for the child with CP.

The research question was: "what level of evidence exists for the effectiveness of common physiotherapy interventions for children with CP utilised in the past decade?"

METHOD

Search Strategy

The search was conducted in May 2008. An electronic search was performed of the following databases: MEDLINE (1996 – May 2008), Embase (1980-May 2008), CINAHL (1983-May 2008), PubMed, Database of Reviews of Effectiveness (DARE), the Physiotherapy Evidence Database (PEDro), and Cochrane Database of Systematic Reviews. Keywords used in the search included 'cerebral palsy', 'treatment outcome', 'physical therapy (specialty)/or physical therapy modalities/ or physiotherapy.mp'. Terms were exploded or searched as a keyword where appropriate. Targeted hand searching was also employed to minimize the chance of missing key studies, including searching reference lists in key studies and review articles.

Two reviewers were involved in the process of reading the abstracts independently to determine eligibility of inclusion into the review. Once the abstracts had been reviewed, the full text papers were retrieved and read. In some cases it wasn't clear until the full paper was read whether it was to be included.

Inclusion criteria

The focus of this review was on common physiotherapy interventions utilised in the past 10 years for children with CP aged 4-18 years. The inclusion criteria were focused and precise. The interventions included were; strengthening (progressive

resisted strength training, task-related training and strengthening through cycling), functional training, supported treadmill training, NDT and other general forms of physiotherapy. The inclusion criteria for the interventions were those considered to impact on orthotic intervention.

Studies examining the above interventions were included if they included children with CP aged between 4-18 years, if they assessed the effectiveness of these interventions using outcome measurement, were published from 1995 onwards and were full text papers in peer reviewed journals.

Exclusion criteria

The interventions excluded were those considered not "standard" physiotherapy including alternative and adjunct treatments. Excluded were; hydrotherapy, hippotherapy, electrical stimulation and other electrotherapy and biofeedback modalities, conductive education and other forms of educational based therapy, casting and splinting and taping. Also excluded was treatment focusing on early intervention because the focus of the review was treatment relevant for orthotic interventions. Studies were excluded if they; were descriptive studies not examining the outcomes of interventions, were single case studies (n=1), had a sample of children less than 4 years or older than 18 years, focused on the upper limb only, did not have children with CP in the sample and were published prior to 1995. Also excluded were abstracts and conference proceedings.

Studies were included if they were examining the effects of physiotherapy after interventions such as surgery or selective dorsal rhizotomy but were excluded if the aim was to examine the effect of the surgery rather than the physiotherapy.

Data extraction and quality assessment

An individualised data extraction form was devised for this review and was pilot tested on 3 articles prior to use. The themes of the form were study aims, study design, subject characteristics, recruitment and sampling, interventions and outcome measures used, and clinical significance and relevance. The same form contained quality assessment items that were matched to the corresponding thematic items of data extraction. Two reviewers reviewed all the included articles independently using the data extraction and quality assessment form. Following this a consensus meeting was held where any disagreement between reviewers was discussed using a third person until consensus was reached.

As well as the individualized quality assessment form, the Oxford Centre for Evidence-based Medicine Levels of Evidence (May 2001) were used to assign a level of evidence for each article. These levels were assigned for the different categories of interventions and an overall score.

For the purposes of this review:

1. A randomised controlled or clinical trial (RCT) involved randomly allocating participants to a treatment or control group
    a. a RCT was considered high quality if both groups had the same baseline treatment and the intervention group had the treatment in question, confidence intervals (CI's) were stated and the CI's were narrow

   b. a RCT was considered low quality if the 2 groups had 2 different treatments, there were no confidence intervals, the sample size was very small or extraneous factors were not well controlled for
2. A cohort study involved 2 groups matched as closely as possible where one group was exposed to the treatment and one was not (i.e. not randomised)[13]
3. A case series involved one group only, assessed at baseline and then reassessed following the intervention, i.e. observational studies.

For this review the question was the effects of treatments in children with CP, therefore studies that used normally developing children as a control group were not considered RCT's, they were considered case series of children with CP. Also for this review, within participant designs (including if children acted as their own controls) were considered to be case series because there was not two groups allocated into different interventions.

RESULTS

The initial electronic search yielded 786 studies. After reviewing the titles there were 42 articles thought to be relevant. From these nine studies were excluded because they did not fit the inclusion criteria. Another six articles were found through hand searching. This left a total of 39 articles included in the main review.

Of the 39 articles included, 34 were studies and 5 were reviews. The numbers of articles in each category were:

1. strengthening; 20 articles (18 studies, 2 reviews)
2. functional training; 1 study
3. treadmill training; 6 articles (all studies)
4. NDT; 4 articles (3 studies, 1 review)
5. frequency of physiotherapy; 4 articles (all studies)
6. general/mixed; 4 articles (2 studies, 2 reviews)

Table 1 displays the articles included in the review and summarises aspects of the study design and subject characteristics. This excludes the reviews.

Table 1 Studies on physiotherapy for children with CP (excluding systematic reviews)

Study	Study Type	Study PT category	Sample size	Power analysis	Age Gender M:F	Classification of CP	Recruitment	Sampling	Inclusion Exclusion
Damiano[14]	CP: case series	strengthening	CP: 14 ND: 25	N	9.1 (6-14) 10:4	All SD	NS	NS	Incomplete
Damiano[4]	Case series	strengthening	14	N	9.1 (6-14) 10:4	All SD	Rehabilitation centre	NS	Vague
Damiano[15]	Case series	strengthening	11	Y	8.81 (6-12) NS	SH 5 SD 6	Rehabilitation centre	convenience	Inclusion ✓ Exclusion NS
MacPhail[16]	Case series	strengthening	17	N	15.8 7:10	SQ 1 SD 7 SH 9	NS	NS	Inclusion ✓ Exclusion NS
Seniorou[17]	RCT	strengthening	Rx 1: 11 Rx 2: 9	N	13 (7-16) 10:10	I: 3, II: 13, III: 4	NS	Random allocation	Complete
Patikas[18]	RCT	strengthening	Rx: 19 C: 20	N	9.75 NS	NS	1 hospital	Random allocation	Inclusion ✓ Exclusion NS
Patikas[19]	RCT	strengthening	Rx: 19 C: 20	Y	6-16 27:12	I: 12, II: 18, III: 9	I hospital	Random allocation	Complete
Fowler[20]	Cohort	strengthening	CP: 24 ND: 12	N	11.5 (7-17) NS	SD	1 hospital	NS	Inclusion ✓ Exclusion NS
Lee[21]	RCT	strengthening	Rx: 9 C: 8	N	6.3 10:7	SD 9 SH 8	1 clinic	Random allocation	Complete
Engsberg[22]	RCT	strengthening	12: 4 Rx groups	N	9.9 3:9	I: 5, II: 5 III: 2	Local PT's	Random allocation	Complete
Crompton[23]	RCT	strengthening	Rx: 8 C: 7	N	10.6 (6-14) 6:9	I: 12, II: 2 III: 1	CP register	NS	Complete
Liao[24]	RCT	strengthening	Rx: 10 C: 10	Y	5-12 12:8	I 10, II 10	Numerous centres	Stratified & randomised	Complete
Eagleton[25]	Case series	strengthening	7	N	12-20 NS	NS	NS	NS	Complete
Williams[26]	Case series	strengthening	11	Y	12.6 (11-15) 1:10	SQ 10, SD 1	1 special school	NS	Complete
McBurney[27]	Case series	strengthening	11	N	12.7 (8-18) 4:7	I: 2, II: 2, III: 7	From another RCT	NS	Inclusion ✓ Exclusion NS
Dodd[28]	RCT	strengthening	Rx: 11 C: 10	Y	13.1 (8-18) 10:11	I: 7, II: 5, III: 9	1 hospital	Random allocation	Complete
Morton[29]	Case series	strengthening	8	N	8.5 (6-12) 4:4	All III	2 special schools	convenience	Complete
Blundell[30]	Case series	strengthening	8	N	6.3 (4-8) 7:1	SD 7 SQ 1	1 school	convenience	Complee

Table 1 Studies on physiotherapy for children with CP (continued)

Ketelaar[31]	RCT	functional training	Rx: 28 C: 27	N	55mo (24-87) 33:22	SH 32, SD 11 SQ 12	PT's	Stratified & randomised	Inclusion ✓ Exclusion NS
Cherng[32]	Within participant	treadmill	8	N	3.5-6.3yrs 6:2	II: 2, III: 6	Not fully stated	NS	Inclusion ✓ Exclusion NS
Phillips[33]	Case series	treadmill	6	N	10.6 (6-14yrs) 4:2	I: 6 SH 4, SD 2	rehabilitation centres	NS	Complete
Begnoche[34]	Case series	treadmill	5	N	2.3-9.7yrs 4:1	I: 2, III: 1, IV: 2 SD 4, SQ 1	Outpatient clinic	convenience	Incomplete
Dodd[35]	Matched pairs	treadmill	Rx: 7 C: 7	N	9rs 10:4	III: 4, IV: 10	2 schools	convenience	Complete
Provost[36]	Case series	treadmill	6	N	6-14yrs 4:2	All I SH 4, SD 2	Part of larger study	NS	Complete
Schindl[37]	Case series	treadmill	10	N	11.5 (6-18) 4:6	SD 3, SQ 4, Ataxia 3	Outpatient clinic	NS	Complete
Tsorlakis[38]	RCT	NDT	Rx 1: 17 Rx 2: 17	Y	7.3 (3-14) 22:12	I: 10, II: 10, III: 14	1 centre	Stratified & randomised	Inclusion ✓ Exclusion NS
Adams[39]	Case series	NDT	40	N	6 (2.6-10.2) 45%:55%	incomplete	1 centre	convenience	NS
Knox[40]	Case series	NDT	15	N	7.4 (2-12) 9:6	I: 1, II: 4, III: 5, IV: 4, V: 1	3 centres	convenience	Complete
Christiansen[41]	RCT	frequency	Rx 1: 10 Rx 2: 14	Y	1-9 16:9	NS	1 centre	Stratified & randomised	Incomplete
Bower[42]	RCT	frequency	44: 11 in 4 groups	Y	3-11 NS	All SQ	14 health districts	Stratified & randomised	Inclusion ✓ Exclusion NS
Bower[43]	RCT	frequency	56: 13-15 in 4 groups	Y	3-12 31:25	NS	33 health districts	Stratified & randomised	Inclusion ✓ Excusion NS
Steinbok[44]	RCT	frequency	Rx 1: 14 Rx 2: 14	N	NS	NS	NS – part of larger study	NS	Complete
Shumway-Cook[45]	Case series	Other (balance training)	6	N	9.2 (7-13) 4:2	I: 2, II: 4	Local schools	NS	Complete
Butler[46]	Case series	Other (trunk training)	6	N	4.7 (2-8) 4:2	SQ 5, SD 1	1 centre	consecutive	Complete

RCT: randomised controlled/clinical trial, CP: cerebral palsy, NDT: neurodevelopmental treatment, Rx: treatment, C: control, NS: not stated, Y: yes, N: no, ND: normally developing, SD: spastic diplegia, SH: spastic hemiplegia, SQ: spastic quadriplegia, I II III IV & V: GMFCS levels, PT: physiotherapist

Of the 34 studies, 11 had sample sizes of 10 or less. The largest sample sizes were in the NDT and frequency studies and the smallest were in the treadmill training and general studies. Fourteen of the 18 strengthening studies had adequate sample sizes. There were power calculations or justifications of sample size in only 9/34 studies (3 frequency studies, 5 strengthening and 1 NDT study). The recruitment and sampling procedures were inadequately or incompletely described in 21/34 studies. They were well described in 8/18 strengthening studies, 1/6 treadmill training, 1/3 NDT and 3/4 frequency studies. Subject characteristics were inadequately or incompletely described in 8 studies (4 strengthening and 4 frequency).

The inclusion and exclusion criteria were vague or inadequately described in 16 studies (4 frequency, 10 strengthening, 1 NDT and 1 treadmill). Often the inclusion criteria were stated but not the exclusion. There was blinding of assessors in only 16 of the studies[42; 43; 46; 32; 41; 23; 14; 28; 31; 24; 27; 29; 19; 37; 44; 38]. There were co-interventions (other treatment as well as that under study) in at least 18 studies (10 strengthening, 4 treadmill, 1 NDT, 2 frequency and 1 other), in 10 it was not stated and there was no co-intervention in 6 studies only. There was a follow-up assessment (ie to assess carryover effects) in 15 of the 34 studies (10 in strengthening, 2 in frequency studies and 1 each in NDT, treadmill and general). In 18 studies the limitations of the study were explored adequately.

Table 2 displays all studies in the review with the study type and the Oxford EBM level of evidence. This table shows that overall there were 14 RCTs; 3 were rated as high quality (level 1b)[24; 19; 18] and 11 rated as low quality (level 2b)[17; 21-23; 31; 28; 35; 38; 44; 41-43]. The study by Liao was particularly well designed and performed. The Dodd study [28] was also well designed with tight inclusion and exclusion criteria and reported clearly, but was graded down because there were no confidence intervals. There was one true cohort study [35] and 19 case series[4; 14-16; 20; 25-27; 29; 30; 34; 32; 33; 36; 37; 39; 40; 45; 46]. There were no case control studies. Overall there were 3 level 1b studies, 12 level 2b studies, 19 level 4 studies and the 5 reviews were classified as level 5 because they did not consist of all RCT's.

The level of evidence per physiotherapy category was:

1. strengthening; 8 RCT's (3 level 1b, 5 level 2b) and 10 case series (level 4)
2. functional training; 1RCT (level 2b)
3. treadmill training; 1 cohort study (level 2b) and 5 case series (level 4)
4. NDT; 1 RCT (level 2b) and 2 case series (level 4)
5. frequency; all 4 were RCT's (level 2b)
6. general other; 2 case series (level 4)

There were 2 studies in the strengthening category that were graded as case series[20; 14] despite the authors considering them to be RCT's. This decision was made based on the review question which was the effect of physiotherapy on children with CP. Both of these studies used typically developing children as the control group.

Overall there were 8 strengthening studies rated as level 2b or higher. The functional training study was also level 2b. All studies addressing intensity of PT were level 2b and one of the treadmill training studies was level 2b. Despite the body of evidence containing a number of observational case series, there was also a number of studies with higher levels of evidence (level 2b or higher).

Table 2 Strength of the evidence

Study	Study Type	PT category	Oxford EBM Level	Additional Information
Damiano[14]	Case series (CP)	strengthening	4	Used TD children for controls
Damiano[4]	Case series	strengthening	4	
Damiano[15]	Case series	strengthening	4	
MacPhail[16]	Case series	strengthening	4	
Seniorou[17]	RCT	strengthening	2b	2 different Rx's compared, no CI's
Patikas[18]	RCT	strengthening	1b	
Patikas[19]	RCT	strengthening	1b	
Fowler[20]	Cohort/case series	strengthening	4	Used TD for control
Lee[21]	RCT	strengthening	2b	2 different Rx's, small sample
Engsberg[22]	RCT	strengthening	2b	
Crompton[23]	RCT	strengthening	2b	
Liao[24]	RCT	strengthening	1b	
Eagleton[25]	Case series	strengthening	4	
Williams[26]	Case series	strengthening	4	
McBurney[27]	Case series	strengthening	4	
Dodd[28]	RCT	strengthening	2b	No CI's
Morton[29]	Case series	strengthening	4	
Blundell[30]	Case series	strengthening	4	
Dodd[47]	SR	strengthening	5	
Darrah[48]	review	strengthening	5	
Ketelaar[31]	RCT	functional training	2b	PT's randomised not participants
Cherng[32]	Case series	treadmill	4	
Phillips[33]	Case series	treadmill	4	
Begnoche[34]	Case series	treadmill	4	
Dodd[35]	Matched pairs/cohort	treadmill	2b	
Provost[36]	Case series	treadmill	4	
Schindl[37]	Case series	treadmill	4	
Tsorlakis[38]	RCT	NDT	2b	2 different Rx's compared
Adams[39]	Case series	NDT	4	
Knox[40]	Case series	NDT	4	
Butler[5]	SR	NDT	5	
Christiansen[41]	RCT	frequency	2b	2 different Rx's compared
Bower[42]	RCT	frequency	2b	No strict controls, wide CI's
Bower[43]	RCT	frequency	2b	No strict controls, wide CI's
Steinbok[44]	RCT	frequency	2b	
Shumway-Cook[45]	Case series	Other (balance)	4	
Butler[46]	Case series	Other (trunk)	4	
Lannin[49]	SR	Therapy post Botox	5	
Harris[50]	SR	Postural control	5	
Total PT			**B-C**	**1b=3 2b=12 4=19**

TD; typically developing Rx; treatment CI; confidence intervals
RCT; randomised controlled/clinical trial SR; systematic review

Table 3 summarises the various regimes of treatment within each physiotherapy intervention category, the outcome measurement used and overall results. For each category there were similarities and differences with the treatment regimes. The strengthening studies often used a frequency of 2-3 times per week for 6 weeks. The treadmill training studies used either a 2 week intensive program or a 6-12 week program of 2-3 times weekly. For the NDT studies the participants had more individualised treatments based on goal setting due to the nature of the intervention. The frequency studies examined the effects of different intensities of treatments.

There was a range of outcome measures used overall. The most common was the GMFM which was used as an outcome in all categories. Dynamometry was common in strengthening studies. Functional tests such as timed 10m walk, and 6 and 10 minute walk tests were used as outcome measures in the treadmill training and strengthening studies. The measures used covered various aspects of the ICF but more commonly activity and body structures and function. There were very few studies that used outcomes to measure changes in participation.

The study designs were strongest in the strengthening and frequency studies, i.e these categories had higher quality studies thus providing more evidence to accept their findings. For strengthening the results varied but overall the studies found that strength was improved in selected targeted muscle groups as measured by dynamometry, but not always significantly. Gross motor function also improved after strengthening in some studies. The three level 1b studies comparing strengthening to other treatments or standard treatment showed that both groups tended to improve with no significant differences between the groups[24; 19; 18]. Strengthening did not increase spasticity. The four frequency studies for general physiotherapy found no significant differences in GMFM scores for different intensities of treatment. This contradicts the Tsorlakis study[38] which found that both intensities of NDT improved gross motor function with significantly more improvement in the more intense program.

Both the treadmill training and NDT categories contained primarily case series studies. The treadmill training resulted in variable outcomes with no clear indication of significant gains, although improvements in standing and walking parameters were seen. Improvements on the GMFM were seen after various intensities of NDT.

Table 3. Interventions and outcomes

Category	Regimes	Outcome measures used and results
Strengthening	PRST programs: 4-5 reps, 3 x weekly for 6-8 weeks[4; 14-16; 22; 25; 27; 29; 28] usually starting at 65% maximum isometric strength. Functional based programs used a number of exercise stations, 1 hour, 2 x weekly for 4-6 weeks[23; 30] or repetitive practise[31] Post-operative strength studies used a set of exercises, 3 x weekly for 6 weeks[17] and 9 months[18; 19] Others were 6 week programs of loaded sit-stand[24], general strengthening[21] or exercise bike[26]	GMFM used in 10 studies with; significant improvements[24; 15], non-significant improvement[28; 29; 26], no difference between strengthening and control groups with both improving[17; 18; 31], and no changes seen on GMFM[21; 22]. Dynamometry showed; significant increases after strengthening quads[4; 16], combined muscle strengths[28], and other muscle groups[30; 29; 22; 21]. Non-significant increases[17; 15; 14; 23] and no differences between groups also seen[19; 24]. Gait parameters: no change in energy expenditure[15; 16], no significant change seen in velocity or cadence[16; 14; 22; 24], improvements in some gait parameters[25; 18]. Significant increases in timed walk tests[30; 25]. No increase in spasticity[29; 20; 19].
Treadmill training	All used motorised treadmill with support harness Amount of support given arbitrary in some studies, others began at 30% of body weight and reduced support to 0% over study period[33; 36]. Some were 20-30 mins, 2-3 x weekly for 6-12 weeks[37; 35; 32]. Others 2 x daily, 6 x weekly for 2 weeks[33; 36]	GMFM used in all studies except Dodd with variable results. Significant increases in 1 study only[32]. Other studies show improvements in dimension D & E. Timed 10m walk used in 4 studies[33-36] with improvements, but only 1 significant[36]. Endurance (10 and 6 minute walk tests) showed either no change[33], non significant improvement[36; 35; 34]. Other gait parameters showed no changes in velocity or cadence[32; 34]
NDT	Treatments individualised for each participant with; 1) 1 hour, 2 x weekly for 6 weeks[39], 2) 75 mins, 3 x weekly for 6 weeks[40] and 3) 50 mins 2 x weekly compared with 5 x weekly for 16 weeks[38]	GMFM used in 2 studies[38; 40]: 1) significant increases in dimension C and E and total score maintained at follow up[40] and 2) both intensities of treatment improved[38] PEDI – improvements in functional skills self-care and caregiver assistance[40] Pedographs – significant increases in stride & step length & velocity (large SD's)[39]
Frequency	2 assessed 4 groups combining aims or goal directed therapy with routine or intense frequency for 2 weeks[42] and 6 months[43]. One compared intermittent bursts with continuous PT[41]. One examined effects of pre-SDR intense PT program[44].	GMFM primary outcome measure in all studies. There were no significant differences in GMFM between different intensities in any study.
Other	One examined effect of individualised targeted trunk training using specialised equipment for 1-2 trunk joints at a time[46]. One assessed effects of 5 consecutive days of balance training using movable force plate system[45]	All children gained independent sitting within 12-25 weeks but other results not analysed clearly[46]. Significant improvements seen in centre of pressure and reduced time to recovery of balance perturbation[45].

There were 2 reviews on strengthening[48; 47], one on NDT[5], one on the effect of therapy after botulinum toxin injections[49] and one on postural control[50]. One of the strengthening reviews was performed in 1997[48] and included only 7 studies with 6 of them low quality studies (level V evidence). The difference between this review and the later one on strengthening[47] is encouraging with the more recent one involving 11 studies from 23 that fitted the inclusion criteria (1 RCT, 9 observational studies and 1 review). Despite overall the studies having low levels of evidence, there have been improvements in study design and quantity of studies over the time period. The review on NDT[5] included 21 articles (not restricted to PT) and found 4 were level I evidence, 10 were level II, 2 were level III, 4 were level IV and 1 was level V. However of the 14 that were level I-II, many had small sample sizes thus compromising power. The review on the effects of all types of therapy after botox injections included 9 studies; 1 RCT on electrical stimulation (level II), 3 on casting (level II and V) and 4 others level IV. None were high quality RCT's and thus the authors concluded that there was a paucity of evidence of the effect of therapy after botox injections. The review on the effects of postural control included 12 studies, most were level III-V (8 studies) and 4 were level II. More research is required in this area also.

DISCUSSION

This review found overall that in the past 10 years there has been an increased interest in using strengthening, functionally based programs and functional training for children with CP. There were more RCT's in the recent strengthening studies with some good evidence of effects of strengthening targeted muscles for children with CP as measured by dynamometry, however carryover into improvements in activity and participation are not clear.

More recently the use of supported treadmill training has been investigated. Clinically this might be an appropriate form of endurance training for children, particularly for those more severely involved who in the past have not been able to participate in this type of exercise. However, most of the studies examining this were case studies with low levels of evidence and small sample sizes and further work is required.

There have been advances in the field over the past 10 years with greater numbers of studies performed and improved study designs. Despite this, the strength of the evidence overall still remains lower than is desired for clinicians to accept that these interventions are positively impacting children with CP. A number of the studies were observational case studies with low levels of evidence. A number of the RCT's were low quality RCT's because of the study design or methodology. The strongest study designs were in the intensity studies; however these 4 studies showed no difference in intensity. Many of the strength studies used a follow-up assessment to determine if there was a carry-over effect and to examine the long term effect of the intervention, however many of the others did not. A number of studies in this review did not report all aspects of their trials or series and there was missing information, particularly in the recruitment and sampling procedures. This makes it difficult to generalise the results. This issue of insufficient reporting has been shown previously in a study that examined the quality of reporting trials in CP, whereby only a small number of trials included were sufficiently reported[51]. This poor reporting jeopardizes judgements on clinical implications.

Conducting research in children with CP is a challenging task due to the heterogeneity of the population and small sample sizes[12]. Physiotherapy is a complex intervention with a diversity of facets such as type and intensity of therapy, using standardised or individually tailored approaches and the skills and experience of the therapists, therefore the specific ingredient responsible for the effect is frequently hard to discern and difficult to measure[52]. The clinical heterogeneity of CP is a major challenge in the performance and interpretation of clinical trials, and clinical interpretation of observed change has been widely omitted in trials[52]. Good quality RCT's to assess effectiveness of physiotherapy in this population are both feasible and necessary, however well designed observational studies can also provide important information.

The most common confounder in many of the studies in this review was the impact of children having "usual" or "standard" physiotherapy either as a control or in conjunction with the intervention in question. It was often not clearly stated what and how much this was. This highlights the issue of what the ideal control group is in this population. The nature of clinical research often means that it is not ethical for children to have "no treatment" as a control group. In the studies in this review it was often difficult to determine whether both groups received similar baseline treatments. The impact of the "usual" physiotherapy on the findings remained unclear in many studies. In the NDT studies each participant had individualised therapy based on goal setting and it is very difficult to control the study conditions in such situations. It is difficult to evaluate effectiveness of treatments when there is no discrete dosage administered under specific invariable procedures in conditions that are held constant[5]. Evaluating the effects of NDT is confounded further because the skill level of the therapists is seldom clear[5]. In one study in this review the study took place during a NDT training course where the therapists are likely to be inexperienced and with instructors present it was not representative of usual therapy conditions[39].

Other influences that may have impacted the results were participant numbers and dosage of treatment employed. Many of the studies were pilot studies with small sample sizes, thus limiting power. Treatment dosage may not have been effective with many studies choosing a 6 week period of treatment, with little evidence to support this decision. It is possible that longer treatment periods are required before beneficial results become evident.

A focused review of systematic reviews on the effectiveness of physiotherapy and conductive education interventions in children with CP showed that the high quality reviews found evidence supporting strength training, constraint-induced movement therapy or hippotherapy, and insufficient evidence on comprehensive physiotherapy interventions[53]. Because of the poor quality of primary studies many reviews drew no conclusions on the evidence of effectiveness of particular interventions.

A recent systematic review of the effectiveness of physiotherapy interventions (both upper and lower extremity) for children with CP that included only RCT's found that of 22 trials only 4 were high methodological quality[54]. The evidence for upper limb therapy was better. The evidence was limited due to low methodological quality and the statistically insignificant results of the studies[54]. Further well-defined trials with clear clinical questions that control for extraneous factors are still required.

Clinical significance

The evidence for strengthening shows that targeted strength training can improve the strength of particular muscle groups as measured at the body function and structure level without provoking increases in spasticity. There is evidence of more improvements in muscle strength than gait or functional parameters, thus carryover into activity and participation has not been shown using the dosages employed in the studies in this review.

Treadmill training may be clinically applicable in this population, however the results were mixed and there was a lack of evidence of significant changes for the better. Further evidence is required before it is used as a routine treatment for children with CP. There was some evidence of NDT improving gross motor function in children with CP but once again further high level evidence is required. The literature for intensity of physiotherapy showed that there was no difference in outcome for different intensities of general physiotherapy treatment; however a more intense program of NDT did show more significant improvements in gross motor function.

Many of the treatments outcomes were quantified using measures based around the ICF domain of body functions and structures such as muscle strength, spasticity, gait parameters, or activities such as gross motor function and gait function. Not many studies considered the effect of the intervention on the participation of the child. Some of the studies considered the effect of the environmental setting such as home programs[27; 28] or group based programs[23; 30]. This highlights the issue of matching appropriate outcome measures for the stated research question. Covering all ICF domains with selected tools is often difficult and a particular focused intervention may not impact all domains. Some studies that used particular outcome tools may have missed changes in other domains not measured in their studies.

There are many areas of physiotherapy practice that are not routinely measured. This includes family advocacy, the organisation of assistive devices and other equipment and education of the child and family. These are very important roles of the physiotherapist and attempts to quantify them should be made.

Limitations of this review

In considering the results of this review, the limitations need to be also considered. The review focused on common treatments utilised in the past 10 years relevant to orthotic management. It did not cover all treatments used by physiotherapists and did not consider adjunct therapies such as hydrotherapy, hippotherapy, electrical stimulation and conductive education. The inclusion criteria were focused and did not include single case studies, studies not published in peer review journals or abstracts. There is a chance that some key studies were omitted from this review.

Implications for orthotics

Although none of the studies in this review directly examined the effect of physiotherapy on orthotic management of the child with CP, clinical experience suggests they work hand in hand. With the increased use of interventions including strengthening and treadmill training, physiotherapists need to make decisions with respect to the role orthoses play with each intervention. Many children with CP have improved stability in stance and better gait function and endurance with AFO's on.

This suggests they would play an important part in treadmill training and strengthening programs, except when strengthening the foot and ankle. Physiotherapy and orthoses are often used in conjunction and complement each other. With less focus on normal movement and more on functional improvements in recent years, orthoses play an integral role in most physiotherapy interventions.

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PHYSICAL THERAPY MANAGEMENT OF CEREBRAL PALSY

Wayne Stuberg PT, PhD, PCS
Professor and Director, Physical Therapy & Motion Lab, Coordinator of Interdisciplinary Education, Munroe-Meyer Institute for Genetics and Rehabilitation, Nebraska Medical Center, USA

Physical therapy or physiotherapy (PT) as a profession has a long history in providing habilitative and rehabilitative services to children and adults diagnosed with cerebral palsy going back to the early 1900's. The model practice act of the Federation of State Boards of Physical Therapy (www.fsbpt.org) describes practice of PT as follows:

1. Examining, evaluating and testing individuals with mechanical, physiological and developmental impairments, functional limitations, and disabilities or other health and movement-related conditions in order to determine a diagnosis, prognosis and plan of treatment intervention, and to assess the ongoing effects of intervention.

2. Alleviating impairments, functional limitations and disabilities by designing, implementing and modifying treatment interventions that may include, but are not limited to: therapeutic exercise, functional training in self-care and in home, community or work integration or reintegration, manual therapy including soft tissue and joint mobilization/manipulation, therapeutic massage, prescription, application and, as appropriate, fabrication of assistive, adaptive, orthotic, prosthetic, protective and supportive devices and equipment, airway clearance techniques, integumentary protection and repair techniques, debridement and wound care, physical agents or modalities, mechanical and electrotherapeutic modalities, and patient-related instruction.

3. Reducing the risk of injury, impairment, functional limitation and disability, including the promotion and maintenance of fitness, health and wellness in populations of all ages.

4. Engaging in administration, consultation, education and research.

During the past two decades, since the last Consensus Conference of the International Society of Prosthetics and Orthotics (ISPO), a significant expansion in research has seen the publication of randomized clinical trials and a number of systematic reviews on the topic of PT and the interventions commonly used by therapists. Highlights of these developments will be discussed in this paper.

This is one of two papers to be presented at the 2008 ISPO Consensus Conference on the topic of PT. The purposes of this paper are to provide; 1) an overview of efficacy literature on PT and 2) to focus on select interventions commonly used by therapists. The interventions to be reviewed include exercise interventions to promote motor control for gait including neurodevelopmental treatment (NDT), the use of electrical stimulation and biofeedback for gait training and lastly, to review the efficacy of casting and stretching to promote flexibility.

METHODS

The following electronic databases were searched for articles up through June of 2008; Pubmed, CINAHL, the American Physical Therapy Association database Hooked on Evidence, the Physiotherapy Evidence Database PEDro, the American Academy of Cerebral Palsy and Developmental Medicine on-line database of Evidence Reports and the Cochrane Controlled Trials Register. The reference lists of the identified studies and reviews were screened for additional references. Study type inclusion criteria included published, full-length articles or full written reports of randomized clinical trials (RCTs) or systematic reviews published since 2000. If no RCTs or systematic reviews were identified then additional references were reviewed. The Oxford Centre for Evidence-based Medicine Levels of Evidence [1] was used to grade the evidence that was reviewed and also used as a basis for the author recommendations.

RESULTS

Meta analyses are the accepted highest level of evidence in support or refutation of interventions. No meta analyses were found on the topic of PT or interventions commonly used by PT related to CP. The next highest level of support are systematic reviews and a number of reviews have been published in the past decade related to PT and interventions for CP.

Methodological Issues

Two recent systematic reviews have focused attention on methodological quality of research on the topic of PT [2] and motor intervention programs [3] for children or adults with cerebral palsy. Kunz and colleagues [2] identified issues in performing randomized clinical trials (RCTs) to assess how the trials handled study design, methodology and analysis in working with a very complex patient population. Fourteen (14) RCTs were reviewed and the authors found that good to fair methodology was used in the studies. The article by Kunz et al [2] more specifically outlines the methodological characteristics of each of the studies in regards to study size, setting, randomization method, use of stratification and concealment. The authors concluded that despite the challenges, "good quality RCTs in physiotherapy for children with CP are feasible and currently being done". Siebes et al, [3] similarly reported that a significant improvement in methodological design was seen in articles published from 1990 to 2001 in contrast to previously documented findings of Vermeer and Bakx [4] who reviewed the literature in the prior decade. Siebes and colleagues identified a sample of 49 studies, of which 13 were classified as Sackett level one [5], 19 as level two and 15 as level four with the remaining studies as case studies and level five. An increase in the percentage of studies at level one or two doubled, from 32 to 66%, in comparing the two time frames.

Physical Therapy Interventions – Global Consideration

A recently published systematic review of the effectiveness of PT interventions for children with CP provides an overview of the topic [6]. The authors reviewed RCTs published between 1990 and 2007. Twenty two (22) trials were identified that included eight intervention categories typically reported in the literature. Overall the studies represented PT services provided to children age seven months to 18 years on

an overall cohort of 255 children with spastic diplegia, 238 with hemiplegia, 180 with quadriplegia and 20 with ataxic or mixed involvement. The levels of disability were represented using the Gross Motor Function Classification System (GMFCS) [7] with 21% at level one, 20% level two, 33% level three, 21% level four and 5% in level five. Eight intervention categories were identified with 53 outcome measures across the studies and only eight of the measures being used in more than one study. No strong evidence was found for the reviewed interventions. Moderate evidence on effectiveness was found for upper extremity treatment and constraint induced therapy. Moderate evidence on ineffectiveness was found on strength training for walking velocity or stride length and conflicting evidence was reported for strength training with the Gross Motor Function Measure (GMFM) [8] as an outcome measure.

Two additional RCTs were identified in the literature, more recently published than the systematic reviews listed above [9,10]. Weindling et al [9] randomized children with spastic CP to a PT only, PT with additional therapy provided by a physical therapist assistant or PT with additional services provided by a family support worker. Seventy six (76) families completed the intervention period with 43 children available for reassessment six months following intervention and 34 at one year post intervention. While improvement in motor functioning was seen following the six month intervention period, no difference was found with addition of the either the physical therapist assistant nor family support worker services. Christiansen & Lange [10] studied the effect of two delivery schedules for PT services. Twenty five children were stratified by GMFCS level and then received intervention sessions of 45 minutes four times per week for six weeks followed by a six week, no intervention phase. This schedule was repeated three times for a total intervention period of 30 weeks and a total of 48 intervention sessions. The other group of children was received 45 minute sessions once or twice per week for 30 weeks, also receiving a total of 48 sessions. Both groups significantly increased their GMFM-66 scores post intervention without difference between the groups other than better compliance with the group receiving four time-per-week intervention scheduling. It should be noted that neither of these studies included a no intervention control group so the effect of only the PT service as not assessed. These two additional RCTs provide further evidence to physical therapy as a global intervention for children with CP.

Table 1 Listing of Randomized Clinical Trials from Kunz et al [2] & Siebes et al [3]

Author, year	Topic	Level of Evidence[1]
Bower et al, 1996 [11]	Goal type and PT intensity	1
Bower et al, 2001 [12]	Goal type and PT intensity	1
Bumin and Kayihan, 2001 [13]	Center based vs home PT	1
Chad, 1999 [14]	Weight bearing on bone density	1
Girolami and Campbell, 1994 [15]	NDT vs regular PT	1
Hazlewood et al, 1994 [16]	Electrical stimulation	1
Jonsdottir et al, 1997 [17]	NDT	1
Ketelaar et al, 2001 [18]	Functional vs standard PT	1
Kuczynsky and Slonka, 1999 [19]	Hippotherapy	1
Law et al, 1991 [20]	NDT and casting	1
Law et al, 1997 [21]	NDT and casting	1
MacKinnon, 1995 [22]	Hippotherapy	1
Mayo, 1991 [23]	NDT intensive vs home	1
O'Dwyer et al, 1994 [24]	Physiologic study on muscle tone	1
Palmer et al, 1990 [25]	NDT vs infant stimulation	1
Reddihough et al, 1998 [26]	Conductive education vs PT	1
Steinbok et al, 1997 [27]	Rhizotomy and PT vs PT alone	1
Steinbok and McLeod, 2002 [28]	Rhizotomy and PT vs PT alone	1

[1]Oxford Centre Criteria, 2001

Neurodevelopmental Treatment Approach

Two systematic reviews [29] and [30] have been published on the topic of neurodevelopmental treatment (NDT) used by therapists working with children with CP. Butler and Darrah [29] participated in an American Academy of Cerebral Palsy and Developmental Medicine evidence report project on the topic. Four level one studies [20, 21, 25, 31] and eight level two studies [32, 33, 34, 35, 36, 37, 38,39] were reviewed. The authors reported that inconsistent evidence was available for the effect of NDT on motoric responses, more consistent positive results on handling to active range of motion, spasticity and discomfort and no advantage favoring NDT in the area of motor development. There was no evidence regarding the effect of NDT on cellular structure or function. Results in favor of an infant stimulation program over NDT were reviewed in the area of functional limitation / activity and no difference in parental report of satisfaction between NDT versus other intervention programming that was offered when a contrast was available to families. The report does provide more detailed information regarding subgroups of children who have shown benefit from the intervention. Brown and Burns [30] reported on 17 articles on NDT as an intervention. The authors also reported largely inconclusive evidence in support of NDT.

Functional Approach Contrasted to NDT or Vojta

Ketelaar and colleagues [18] randomly assigned a group of 55 children with CP to either a functional group intervention program or a reference group that included continuation of current therapy which was predominately NDT or Vojta. The intervention lasted for six months with an average of 3-4 PT sessions per month for

both the intervention and reference group. The main features of the functional approach were functional goals, repetitive practice on the motor ability in context of the functional activity, active participation by the children in motor planning and execution and active participation of the parents in goal setting, decision making, implementation of the program and evaluation of the goals. The program was evaluated pre, post . Follow up assessment were completed at six and 18 months following pretesting. The authors reported a significant improvement in GMFM at post-test for both groups, but on the group receiving the functional approach intervention improved on the PEDI with post testing. The authors concluded that using this approach greater functional improvement is seen over more traditional approaches to PT intervention.

Balance Training

Harris and Roxborough [40] published a systematic review of 12 studies examining the efficacy and effectiveness of physical therapy postural control interventions for children with CP. The authors reported on limited evidence, Sacket levels three to five, for all studies reviewed. Individual studies have since been reported by Ledebt et al [41] & Woollacott et al [42] that provide further evidence. Ledebt and colleagues [41] demonstrated improved gait symmetry and balance in children with hemiplegic involvement following a six week intervention program of three sessions per week. A multiple baseline design was used with randomization to treatment versus control phase. Woollacott et al [42] and Shumway-Cook et al [43] also used a single-subject, multiple baseline experimental design to examine massed practice effects of balance training on muscle activity and stability in six children with hemiplegic and diplegic CP. A moveable foreplate system (NeuroCom) was used to deliver 100 pertubations per day over a five days of intensive training schedule. Improvements in the standing subsection of the GMFM and select measures of balance from the force platform were reported. providing additional support for balance training for children with CP.

Neuromuscular Electrical Stimulation

Kerr et al [44] published a systematic review to examine the efficacy of electrical stimulation for strengthening or to improve motor function of children with CP. Eighteen (18) articles were reviewed with six of the articles classified as RCTs. Twelve (12) of the articles examined the use of neuromuscular electrical stimulation (NMES) and six (6) threshold e-stim (TES). The differentiation between the two is that NMES is applied to obtain a palpable contraction where TES is applied subthreshold to a muscle contraction. The RCTS reported by Kerr are listed in Table 2. Kerr reported evidence from three RCTs that NMES has been shown to improve motor control through an increase in GMFM scores but that conflicting and inconclusive evidence from three RCTs is seen for TES for either improvement of muscle strength or motor function in children with CP. More two additional RCTs [45, 46] have been published on the use of either NMES or TES in CP. Kerr and colleagues [45] examined the efficacy of NMES and TES in strengthening of the quadriceps and did not find any significant findings. Ho and colleagues [46] examines NMES applied to the gastrocsoleus complex to reduce muscle stiffness and to study the effect on select gait temporal-distance measures. The authors reported significant results in improving select walking measures, but not muscle stiffness.

Table 2: Electrical Stimulation Reference Articles

Author, year	Topic	Level of Evidence[1]
Dali et al, 2002 [47]	TES and PT	1
Hazelwood et al, 1994 [16]	NMES and PT	1
Park et al, 2001 [48]	NMES and PT	1
Sommerfelt et al, 2001 [49]	TES, stretching and PT	1
Steinbok et al, 1997 [27]	TES and PT	1
Van der Linden et al, 2003 [50]	NMES and PT	1

[1]Oxford Centre Criteria, 2001

Biofeedback

No systematic reviews or RCTs were identified addressing the issue of biofeedback and gait training in children with CP. Table 3 lists eight articles that were found on the topic and the level of evidence of each. The use of auditory and visual surface EMG biofeedback has limited evidence with no studies identified who randomized the intervention to a non-intervention control group. Only the Dursun article [51] included randomization of treatment assignment in their study design and that was to contrast the use of biofeedback to the use of conventional exercise. Biofeedback was used by the authors to treat dynamic equinus with a cohort of 21 children receiving both exercise and biofeedback in comparison to an exercise only group. Both groups significantly improved gait function, but it was the biofeedback group that significantly improved muscle tone and active ankle range of motion. The authors concluded that biofeedback was an effective intervention for dynamic equinus in children with CP.

Table 3: Biofeedback Reference Articles

Author, year	Topic	Level of Evidence[1]
Bolek, 2003 [52]	Surface EMG (SEMG) for dorsiflexion	4
Colborne et al, 1994 [53]	PT or SEMG for plantarflexors	3
Dursun et al, 2004 [51]	SEMG for equinus	2
Flodmark, 1986 [54]	SEMG for gait training	4
Kassover et al, 1986 [55]	Auditory EMG for heelstrike	4
Seeger et al, 1981 [56] & 1983 [57]	Auditory EMG for gait symmetry	4
Toner et al, 1998 [58]	SEMG, strengthening & ROM for ankle function	4

[1]Oxford Centre Criteria, 2001

Casting

In 2007, AACPDM published an evidence report which included a systematic review of casting, either alone or in combination with botulinum toxin type A on equinus in children with CP [59]. Of the 22 articles reviewed, 12 reported on casting as a sole

intervention with only one being a level two RCT and the others providing level four evidence. Table 4 lists the articles. The small level two RCT by Bertoti [60] examined 10 weeks of intervention in 16 children with CP where were randomly assigned to a NDT only or NDT plus casting group. A significant percent increase in stride length was found, but other gait variables were non-significant between groups post intervention. In reviewing all casting literature in the study review, Blackmore and colleagues [60] concluded that there was little evidence to support casting versus no casting, but that a no-intervention control group has not been used in any RCT on the intervention. Many of the studies demonstrate a return to baseline values at follow up and it is unclear whether the improvement seen in range of motion is improved stretch tolerance or actual increased range due to the outcome measurement methodology that is typically used.

Table 4: Casting

Author, year	Topic	Level of Evidence[1]
Bertoti 1986 [60]	Inhibitive casts & PT	2
Brouwer et al, 1998 [61] & 2000 [62]	Serial casting	4
Cameron and Drummond, 1998 [63]	Serial casting and PT	4
Cottalorda et al, 2000 [64]	Serial casting	4
Dvir et al, 1991 [65]	Serial casts & NDT	4
Hinderer et al, 1988 [66]	Bivalved casts & NDT	4
Otis et al, 1985 [67]	Bivalved casts	4
Sussman and Cusick, 1979 [68]	Inhibitive casting & PT	4
Tardieu et al, 1982 [69]	Serial casting	4
Watt et al, 1986 [70]	Inhibitive casts & PT	4

[1]Oxford Centre Criteria, 2001

Stretching

Two systematic reviews [71, 72] have recently been published examining the effect of passive stretching to improve range of motion or to affect spasticity in children with CP. Pin and colleagues [71] identified 10 studies, of which, four were RCTs on the topic. Overall the authors reported limited evidence for passive stretching to improve range of motion. The authors reported some evidence for increased range of movement following stretching and decreased spasticity, but no lasting effect. Sustained stretching was preferred to manual stretching. Wiart [72] reported on seven studies with three RCTs and the paper overlapped the findings of Pin and colleagues. The RCTs from both reviews are listed in Table 5. The findings of Wiart also reported limited evidence for passive stretching, active stretching or for positioning to improve range of motion in children with CP.

Table 5: Stretching References

Author, year	Topic	Level of Evidence[1]
Miedaner & Renander, 1987 [73]	Passive stretching	2
O'Dwyer et al, 1994 [24]	Passive stretching	2
Richards et al, 1991 [74]	Single prolonged stretch	2
Tremblay et al, 1990 [75]	Single prolonged stretch	2

[1]Oxford Centre Criteria, 2001

RECOMMENDATIONS

Based on the information reviewed in this paper the following recommendations are made:

- While there is some evidence, it is not consistent evidence (Grade A) for PT as an intervention for children with CP.
- Moderate evidence (Grade B) is present on effectiveness of upper extremity treatment, constraint-induced therapy, use of a functional approach to improving gross motor skills and use of NMES to improve gait function.
- Limited evidence (Grade C) for NDT as a treatment approach, the use of balance training to improve sitting or standing balance, the use of biofeedback to improve gait and the use of casting to improve equinus.
- Inconsistent evidence (Grade D) or moderate evidence for the *ineffectiveness* of strength training for walking velocity or stride length and for passive stretching to increase range of motion and for the use of TES to improve gait.

It is reported widely in the literature that the lack of evidence-based support for an intervention does not negate the clinical effectiveness or efficacy of that intervention if limited research on the subject has been completed and that there is clinical consensus that the intervention is effective. Physical therapy and many of the interventions used by therapists are in this situation with limited high quality research currently available to guide practitioners toward sound evidence-based decisions. The practitioner is left with the need to discriminate where beginning evidence of a lack of support for an intervention, for example passive stretching to improve range of motion, is seen in the literature pending the completion of more definitive research. This will be a long term process as certainly part of the issue lies in the heterogeneity of children with CP and difficulty in gaining the information, although significant strides have been made in this area over the past decade as reported by Kunz et al [2] and Siebes et al [3].

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PHYSICAL THERAPY MANAGEMENT: POINTS FOR DISCUSSION WHAT DO WE KNOW, WHERE SHOULD WE GO?

Dianne J. Russell PhD
***CanChild* Centre for Childhood Disability Research**
McMaster University, Canada

I want to start by acknowledging Dr. Harvey and Dr. Stuberg for undertaking the synthesis and review of this literature. I realize what a big task it was. It is interesting how they both took a very different approach to their reviews. In addition to focusing on different interventions, Dr. Stuberg focused on collating evidence from systematic reviews and RCT studies where possible as the highest level of evidence, whereas Dr. Harvey relied more on individual studies (which included RCTs) and rated the systematic reviews as low level evidence. This brings to light one of the challenges with using and interpreting the Oxford CEBM guidelines for the review of the evidence and recognizing the inconsistencies which may occur in the application of the guidelines. This is in addition to the challenges with interpreting the strengths and limitations of individual articles. It was recognized throughout this meeting that the Oxford guidelines were challenging to apply in our area, where randomized controlled trials are the exception rather than the rule. Dr. Harvey did a nice job describing individual studies documenting specific information about child characteristics and outcomes, making it easy to tease out the generalizability of the findings from the tables. However, when summarizing the evidence for a particular intervention across studies, Dr. Harvey averaged the levels of evidence rather than weighting those studies with higher methodological rigour as providing stronger evidence. I have used their two reviews along with a recent systematic review of randomized trials by Anttila et al.[1] to guide this discussion and identify important questions to consider.

As with many areas in rehabilitation medicine, the evidence for the effectiveness of physical therapy interventions for children with cerebral palsy is sadly lacking. While there has been a lot of work done in the area, we still don't know what interventions work, for whom, and under what conditions. The methodological challenges we face are great. We have no high level (Grade A) evidence for any of our interventions from high quality randomized controlled trials. We have limited Grade B evidence from Marjolijn Ketelaar's well done RCT [2] for the value of a functional approach to therapy (defined as physical therapy with an emphasis on practicing functional activities) to improve capability and performance of functional skills in daily life as measured by the Pediatric Evaluation of Disability Inventory (PEDI) in children with CP primarily in GMFCS levels I and II. While these results are promising, further studies replicating this work are needed. Law et al.[3] are currently conducting a RCT of a functional approach to therapy or "task/context approach" compared with a "child-focused approach" and measuring outcomes at all levels of the International Classification of Functioning, Disability and Health (ICF) [4] which should help with our understanding of the generalizability of the results for functional therapy. Dr. Stuberg also identifies Grade B evidence from several RCTs evaluating Neuromuscular Electrical Stimulation (NMES) to improve gait; however without going back to the original articles and describing the details of the individual study participants in terms of ages and GMFCS levels it is difficult to know to whom these results are generalizable. There is evidence from systematic reviews that strength training programs can increase muscle strength in children and young adults with CP without increasing spasticity, however the evidence from four lower quality RCTs indicate that strength training is not effective in changing gait parameters such as

walking speed and stride length. There is inconclusive evidence regarding its effect on gross motor function as measured with the GMFM. All other areas of physiotherapy that were reviewed have limited or conflicting evidence.

This review of the evidence for physical therapy interventions raises many questions and my job is to highlight some issues that we could consider for discussion. I think the first one is what do we have to do in order to get the quality evidence we need? Assuming that the RCT is the gold standard for determining evidence of effectiveness, is it necessary and is it feasible to conduct a high quality RCT in this area? Unlike work in the area of drug trials, where RCTs are relatively straightforward in terms of randomizing and controlling for important variables, we face incredible challenges related to the difficulty in "blinding" therapists and children to their treatment interventions, compliance, fidelity to the prescribed treatment(s), defining and describing our treatments specifically enough so they can be administered consistently and replicated by others. Families may have difficulties consenting to randomization (especially if it means changing therapists) and may choose not to participate. Is our field in a place where we have enough good lower level evidence with good outcomes to justify the cost and resources needed for an RCT? Whether or not we decide that RCTs are necessary, how will we overcome the methodological challenges associated with doing research in the area of cerebral palsy? I'm going to use the PICO framework (Patient, Interventions, Comparison, and Outcomes) to highlight a few of these challenges.

1) Patient: Patient heterogeneity makes it particularly challenging to get large enough samples in research studies to have the power to detect statistical and clinically important differences between interventions. In studies of general therapy approaches, we may include children from a spectrum of GMFCS levels in order to get large enough samples, perhaps diluting the effects of the intervention if it is only effective for a subgroup of children. Do we need to narrow our focus to determine what works best for children in specific GMFCS levels or at least stratify by GMFCS levels so we can start to tease this out? On the other hand, when samples only include children in limited GMFCS levels, it is important when summarizing the evidence that we are explicit about to whom these results apply.

2) Interventions: Physical therapy "management" or intervention is dependent on the goals of the child and family in consultation with evidence-based, family-centred service providers. Children with CP are extremely variable in their functional capabilities and goals, making the target of intervention challenging. PT interventions as with many rehabilitation interventions are diverse and often a mix of several different approaches making the "active" ingredient difficult to tease out. Thus while it may be artificial to try to "standardize" a treatment approach if we can't clearly describe what it is we do, then how can we evaluate and replicate the results? And if we do find an effective approach, will we have described it in enough detail in order to be able to teach it to others? Have we done a good enough job defining the content and the process of our treatments that will facilitate research, replication and dissemination of treatments into practice?

3) Comparisons: Another interesting dilemma that arises when reviewing the evidence concerns what should be the appropriate control or comparison group? Does it make sense to compare two different intensities of a treatment that has no evidence of effectiveness? How do we interpret the results? Several authors criticize studies for

not having a “no treatment” control group. Is it necessary or ethical to have a no-treatment control?

4) Outcomes: Are we measuring the right outcomes? If our goals are to improve activity and participation of children, we should not only be measuring impairments and assume that improvements will carry over into functional activities. We need to ensure that our outcomes match our goals. Do we have good outcome measures for what we want to evaluate at all levels of the ICF or do we need to focus more effort on developing and validating clinically appropriate, responsive, psychometrically sound outcome measures? Should we be trying to come to consensus on common outcomes? Do we need more multi-centre collaboration with common methods and measures to allow us to combine data sets for meta-analyses? If so, how do we move forward on this? And finally, with limited health care dollars and limited evidence of effectiveness for most of what we do, what is the take home message for evidence-based clinicians, parents and families, and funders of health care?

It is encouraging to note that the methodological quality of studies of motor interventions for children with CP has greatly improved over the years [5,6] however the evidence is still not there and we need to decide what are the important next steps in getting the evidence we need to ensure we are providing children and families with the most cost effective, family-centred, evidence based care.

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OCCUPATIONAL THERAPY MANAGEMENT OF CHILDREN WITH CEREBRAL PALSY

Annette Majnemer PhD, OT
Laurie Snider PhD, OT
School of Physical & Occupational Therapy, McGill University, Canada
&
Ann Christin Eliasson PhD, OT
Department of Woman and Child Health, Karolinska Institute, Sweden

The primary role of occupational therapists is to enable individuals to participate in meaningful occupations of their everyday life. Occupational therapy practice focuses on enhancing occupational performance in self-care, productivity and leisure activities and overcoming the obstacles that limit the ability to participate in these occupations [1]. Occupational therapy is holistic by definition, and includes minimizing impairments, maximizing functioning in all facets of life, and facilitating participation in all life roles. This may be achieved through remediation and training (to acquire or improve abilities and skills), and compensation and adaptation (by modifying the task or the environment to improve functioning). Many new outcome measures are enhancing the therapist's ability to objectively assess children with disabilities and monitor changes over time.

Recent advances in pediatric occupational therapy practice have been prompted by a number of theoretical frameworks and concepts [2] [3] [4]. These include:

* **Client-centred practice** (personal autonomy and choice, life satisfaction)
  - What are the child's preferences and goals that are intrinsically motivating?
* **Family-centred care** (collaboration on goals and priorities)
  - What are the family's priorities and concerns at this time? What resources and supports are available to them?
* **Biopsychosocial framework of the ICF (**International Classification of Functioning, Disability and Health; a holistic view of functioning and health)
  - What areas should be considered in the evaluation and the intervention process to ensure that therapeutic strategies are holistic and encompass all aspects of an individual's functioning? Which contextual factors are either enhancing or inhibiting functional capacities?
* **Person-Environment-Occupation Model** (transactive model)
  - In what way are the impairments and activity limitations of the individual influenced by characteristics of everyday tasks, and by the environment in which these activities are performed? At what level(s) should the OT intervene (person, environment and/or occupation) to promote function?
* **Lifespan approach** (key transition points in development)
  - What are the current challenges for the child at this point in development? When should the occupational therapist re-evaluate the child's needs and concerns in the future?
* **Evidence-based practice** (graduate level education)
  - What new research evidence needs to be considered, in the context of clinical expertise and clients values, to ensure best practice?
* **Life-long learning** (knowledge transfer and exchange)
  - Which new scientific discoveries need to be transferred and used in clinical practice? Which areas of occupational therapy practice need to be investigated through scientific inquiry?

This paper provides a brief overview of the evidence on the effectiveness of selected OT interventions for children with cerebral palsy (CP). Interventions used by OTs are diverse and may focus either on elements of body structure and function, activity, participation and/or the environment. The areas of intervention selected were those in which there is a body of evidence to review, and are discipline-specific (typically performed by occupational therapists). We chose to organize the literature according to the classification proposed by the ICF. For *body structure and function*, we reviewed the evidence on: 1) **casting and splinting** of the upper extremity, and 2) **seating and positioning**. These are devices or adaptations used as tools to prevent or minimize deformity and to facilitate abilities and capacities. For *activity and participation,* we reviewed the evidence for three occupational performance domains; 1) productivity (**hand use**), 2) self-care (**feeding**) and 3) play (**virtual reality**). Facilitation of the use of the upper extremities through constraint therapies was specifically reviewed. Promoting bimanual hand use in children with CP is critical for the successful execution of everyday tasks and activities (ref). With respect to independence in self-care, feeding challenges are a common area of concern for this population and is associated with respiratory and gastrointestinal health risks. Safe and efficient feeding is essential for nutrition and growth, positive parent-child interactions and personal autonomy. Therefore the effectiveness of feeding interventions was reviewed [5]. Play is a primary occupation of children and is an important source of skill development (motor, social, cognitive) and social participation. Specifically, we focused on virtual reality as a new modality for intervention to enhance motivation, mastery and self-efficacy (Rizzo et al, 1998). For each of the areas reviewed, we formulated a primary **PICO** research question (**P**opulation, **I**ntervention, **C**omparison, **O**utcome) according to the domains of *body structure*, *body function*, *activity and participation*. Although, we provide a systematic review of discrete OT intervention approaches (as outlined above), each of these approaches should not be performed in isolation, but rather, should be integrated within the concepts and frameworks highlighted above.

SPLINTING AND CASTING

Search strategy:

The electronic databases that were searched included CINAHL, CIRRIE, the Cochrane Library, EMBASE, ERIC, HealthSTAR, Health and Psychosocial Instruments, MEDLINE, OTSeeker, PEDro, PsycINFO and RehabData, using the key words: tetraplegi* (ie. tetraplegia or tetraplegic), spastic*, quadriplegi*, quadrapare*, pes equinus*, monoplegi*, little* disease, hypotonic*, hemiplegi*, hemipare*, dystoni*, diplegi*, dyskine*, choreoathe*, cerebral palsy, atheto*, ataxi*, cast*, splint*. Only English or French manuscripts were included, for publications up to and including December 2007. All age groups between newborn and 18 years of age were included. Articles on stroke, traumatic brain injuries, adults, and very poorly designed studies were excluded (e.g. anecdotal reports, single subject observations). The search was completed by a research assistant trained in the appropriate search strategies.

Definition of terms:

Orthotic management would include the use of splints or casts to improve function. They may be used alone or in conjunction with other rehabilitation interventions (Kurtz, 2002). ***Splints*** are devices designed to maintain parts of the limb in a particular position, stabilize one or more joints, and/or facilitate specific movement patterns [6]. Splints may be either static (rigid) or dynamic (movable parts), and are

made of moldable plastic. ***Casts*** are used to maintain a body part in a fixed position, and are typically made of plaster or fiberglass. They may or may not be removable. Serial casts involve a series of casts that are changed periodically to progressively increase the stretch on a muscle, whereas inhibitive casts are designed to decrease the effect of reflexes [6].

SPLINTING

Purpose

Static resting splints are used primarily to maintain muscle length and prevent deformities for children with spasticity. They can be used while sleeping or periodically during the day, when the child is less active. A biomechanical (to support body part) and /or neurophysiological (to decrease spasticity) framework is applied, with the primary purpose of maintaining or increasing muscle length so as to improve position, flexibility and quality of movement, and preventing contractures. In addition, prolongation of muscles may decrease spasticity. Other possible benefits are to reduce pain, support weak muscles, minimize involuntary movements or reflexes, and immobilize part of the body. In some cases splints serve as a base for attachments of other devices such as self care aids or toys [6]. The overarching goal is to improve hand function. For example, the wrist may be stabilized in a neutral (slightly extended) position to facilitate a more functional grasp pattern.

Primary PICO research question: Does daily use of a hand splint in children with cerebral palsy result in enhanced hand movement patterns and more functional use of the hand?
P: Infants, children and/or youth with cerebral palsy
I: Hand splint, typically for several hours per day, over several weeks
C: Compared to baseline (prior to use of splint) or compared to a control group
O: Measures of *body structure* (somatosensory evoked potentials), *body function* (spasticity, range of motion, grip strength, arm posture, visual-motor control, visual-perception, motor skills such as grasp) and *activity and participation* (hand use, functional activities); body function measures predominate.

Primary findings:
Nine studies described the effectiveness of upper extremity splints for children with cerebral palsy. Descriptive information regarding these studies appears in Table A and B [7-15] . Children were of varying ages (infants to adolescents) and most had either quadriplegia or hemiplegia. Sample size for 8/9 studies was small (1-12 subjects). Interventions typically involved pre- and post-testing after a period of weeks to months of daily use of a hand splint, with no RCTs. A variety of hand splints were used in these studies, with the primary goal of maintaining the hand and wrist in a well-aligned neutral position and/or to inhibit spasticity. Outcome measures were predominantly observational, with one study using a three-dimensional tracking system. Results suggest improvements in hand and arm use, including grasp, better range of motion and hand positioning, and decreased spasticity. Muscle strength did not improve. Only one study found no positive effects. Some studies did not apply formal statistical analyses, and these studies are indicated in the Table. Improvements in functional activities were not supported by these studies.

Review articles:
Autti-Ramo et al [16] conducted a systematic review of all review articles on the effectiveness of upper and lower limb casting and orthoses in children with cerebral

palsy. This includes the review article by Teplicky et al [6] in which 3 articles on splinting are included. The conclusion of these reviews is that hand splints appear to improve grasp but the ability to use the hand more functionally in everyday tasks remains unclear. Existing studies suggest that splints may be more helpful in children with more severe limitations, however this requires empirical support. Furthermore, the choice of the type of splint needs to be task-specific.

Conclusions and future directions:
The literature in this area is sparse. Studies indicate that following a period of use of upper extremity splints, spasticity may be inhibited and there are improvements noted in range of motion, grasp pattern and general hand use. The long-term effects are unknown. Objective, quantitative measures of hand function have not been used, therefore improvements in grasp and hand use require empirical validation in the future. Use of splinting in conjunction with more functional, task-specific approaches may be warranted to enhance functional gains and should therefore be examined in future studies. **Grade of Recommendation: C**

CASTING

Purpose:
Casting may be used on the upper or lower extremities for short periods of time to stretch a shortened muscle, and thus, to increase range of movement. Therefore, casting may either prevent or correct deformities, decrease spasticity, and possibly also decrease pain and discomfort. Supporting normal alignment and greater range of movement may help promote greater function [6, 16, 17].

Primary PICO research question: Does daily use of an upper extremity cast in children with spastic cerebral palsy result in enhanced upper extremity movement patterns and more functional use of the hand?
P: Infants, children and/or youth with spastic cerebral palsy
I: Upper extremity inhibitive casting, typically for several hours per day, over weeks to months
C: Compared to baseline or compared to randomized control group
O: Measures of *body function* (spasticity, range of motion, grip strength, quality of upper extremity movements, fine motor skills such as grasp) and *activity and participation* (hand use, functional activities); body function measures predominate.

Primary findings:
Eight studies described the effectiveness of upper extremity casts for children with cerebral palsy. Descriptive information regarding these studies appear in Table C and D [18-25]. Children were of varying ages although most were preschoolers or early school-age, and subjects had one of three spastic subtypes (diplegia, quadriplegia, hemiplegia). Sample size for 6/8 studies was small (1-12 subjects), typically involving pre- and post-testing after an extended period of daily use of casts. Two studies by the same group [21, 26] involved larger samples (50+) and implemented a randomized controlled trial. Casts were predominantly inhibitive, with the primary goal of inhibiting spasticity and maximizing range of motion. Casts were worn several hours per day, for weeks to months. Casting was viewed as an adjunct to other rehabilitation treatment approaches. Outcome measures included observational data, measurement of range of motion, and in some cases, application of standardized measures of quality of upper extremity movement and motor skills. Results suggest improvements in passive and active range of motion, and decreased spasticity. There is some

suggestion of improvements in upper extremity movement quality, but this is unclear. Improvements are not sustained on follow-up assessment. The positive effects on quality of movement noted in the first randomized trial [21] were not replicated in the second trial[26]. Authors suggest that this may have been because the first study included older children (4-8 years) who may exhibit greater benefits than younger children (<4 years). The influence of age remains to be determined in future studies.

Review articles:
In addition to the systematic review of all review articles on the effectiveness of upper and lower limb casting and orthoses in children with cerebral palsy conducted by Autti-Ramo et al [16], there are two review articles that focus predominantly on casting. This includes the review article by Teplicky et al [6] in which 3 articles on casting are included, as well as the review by Lannin et al [17] which included articles in children and adults with brain injury. The conclusion of these reviews is that upper limb casting improves range of motion with some possible effect on decreasing muscle tone. The effect on quality of arm movement and hand function remains unclear, although there is some suggestion of improved movement quality in the short-term. It is uncertain if such improvements are merely cosmetic or have a functional impact [16, 17].

Conclusions and future directions:
Casting appears to transiently improve range of motion, with some possible effects on decreasing spasticity and improving quality of movements. The lack of high level evidence limits the ability to strongly support or refute the short and long-term effectiveness of upper limb casting. Clinical indicators for casting appear to be presence of soft tissue contracture, spasticity and limited joint range [17]. The type or severity of cerebral palsy or the possible effects of age were not examined as determinants of effectiveness of casting. In future studies, outcome measures need to focus not only on the direct impact on range of motion and spasticity, but the possible concurrent benefits on hand function. Casting in conjunction with particular task-specific training may prove beneficial in optimizing functional gains, and should therefore be examined in future studies. **Grade of Recommendation: C**

SEATING AND POSITIONING

Search strategy:
The electronic databases that were searched included MEDLINE, CINAHL, the Cochrane Library, OTSeeker, PEDro, and PsycINFO, using the key words: cerebral palsy, quadriplegia, children, posture, position, orthotic device, equipment design, wheelchairs interior design and furnishing, infant equipment, rehabilitation, sitting. Only English language manuscripts were included. All age groups between newborn and 18 years of age were included. Case studies, methodological articles and basic science articles were excluded as well as articles earlier than 1980. Thus, all relevant papers between 1980 until July 2008 were included.

Definition of terms:
Seating is an important though complex issue for children with cerebral palsy. It is presumed that good seating: 1) promotes normal development by maintaining of postural alignment and reducing undesirable tone and reflexes; 2) enhances postural stabilisation of the trunk as a prerequisite to functional performance of upper limb; 3) helps to prevent or delay development of deformity or muscle contractures; 4)

optimises the child's position for feeding, respiratory and digestive function; 5) assists exploration of the child's environment; and 6) improves head control which is essential for orientation and socialisation and to help the child to develop cognitive and communication skills. In short, a seating system is usually chosen with the aim to improve posture and enhance function. It is believed that the seating position affects the potential for future development and independence in daily life. Finally, other important but often neglected factors are the utility and comfort for the children and the seating system's compatibility with the needs of families and transportation arrangements.

Primary PICO research questions: The following research questions were addressed:

1. Does changing sitting ability using special chairs and other devices (Intervention) improve upper extremity function (Outcome) in children with severely impaired CP (Population) compared other positions or chairs (Comparison intervention)
2. Does changing sitting ability using special chairs or other device (Intervention) improve posture (Outcome) in children with severely impaired CP (Population) compared other positions or chairs (Comparison intervention)
3. Does changing sitting ability using special chairs or other devices (Intervention) improve pulmonary function (Outcome) in children with severely impaired CP (Population) compared other positions or chairs (Comparison intervention)

Primary findings:
Four review articles and 22 research articles were selected. In general, the studies were small and many of them published before 2000. Hence the level of evidence to support intervention using seating systems is weak.

1. Does sitting ability by special chairs and other device improve upper extremity function?
The review by Stavsness [27] investigated the possibility of finding a seating position for children with CP which improved the control of their upper extremity. Sixteen articles were found, all of them were published before 1997. Two of the articles were based on small RCTs, and the other studies used before and after designs. The review investigated the influence of seating position, mainly backward-neutral-forward orientation. All but one study supported the neutral to slightly forward orientation (0-15 degree). The seating angle did not appear to influence upper limb function. Typically the seat orientation is suggested to be combined with Functional Sitting Position (FSP) including hip abduction, foot rests, hip belt, and a cut-out tray. The FSP are recommended to be determined for each individual. Reach and grasp were investigated by different methods, scoring systems, questionnaire, a hand function test, as well as motion analysis. The results were investigated over a short time period despite the fact that children spend many hours in their seating system.

Other articles discussing improved upper limb function by special chairs and sitting device
Ten articles were found focusing on upper limb function, only one was published later than 2000 [28]. Some of the articles investigating postural improvement also discuss upper limb function, like those written by Myhr and colleagues [29-31]. In these studies, the children were similarly investigated when positioned for short time periods, ranging from only seconds to a few minutes. Demonstrating improved task performance and satisfaction as well as reduced caregiver assistance are discussed in two studies reporting the difference when lap belts were replaced with rigid pelvis

stabilizer [28, 32]. On an individual level, adaptive seating was found to improve upper limb function, feeding and eating behaviour [33-35]. The control of reaching and grasping was not found to be influenced by sitting position, [36] using a saddle seat or flat bench, [37] different seating angles, [15, 38] or hip flexion [39, 40].

2. Does sitting ability by special chairs and other device improve posture?
Articles were selected because posture was evaluated, however these papers often reported other outcomes. One review investigated the effect of tilt-in-space seating [41](16). It included adults and children with different neurological and neuromuscular impairments. Nineteen studies were included, 10 studies from children with cerebral palsy, 5 were overlapped with the Stavnees review [27]. The studies investigated the immediate effect of tilting the seating. The over all result was that posterior tilt can reduce pressure at the interface under pelvis. The implications for children with CP were unclear.

Does seat inclination affect the function of children with cerebral palsy
The review of McNamara and Casey [42] aimed to evaluate if postural control, muscle activity and upper extremity function was influenced of the seat inclination. Ten studies were included published 1990-2006. There were four single case studies, two studies were longitudinal and four studies were between group comparisons. In general the numbers of subjects were small. For postural control, anterior tilt resulted in reduction in sway for hypertonic children and increased sway for hypotonic children. Upper extremity function improved when sitting in the functional sitting position compared to posterior seat inclination. The conclusion is that anteriorly tilting the seat appears to improve function and that seating systems should be adapted to needs of the individual.

Other articles discussing improved posture by special chairs and other sitting device
Nine articles were found that investigated different types of chairs and adaptations. The quality of the articles improved over time. Almost all studies included profoundly impaired children. Investigation of the Seating Buggy wheelchair shows that if it was well adjusted for each child, the sitting balance was improved [43]. Static correction of scoliosis can be improved with an arrangement of lateral pads on a seating system [44] or by thoraco-lumbar-sacral orthosis [45]. An adaptive seating device incorporating a thoracic lumbar sacral orthosis had a positive outcome effect on feeding and posture in a fairly large prospective study [46]; a finding supported by another older study [47]. Individualized fitting seemed to be important. The functional sitting position with or without a cut-out tray tends to reduce pathological movement and improve upper extremity [29, 30]. For some children the ‘saddle seat’ appears to improve postural control but not upper extremity function [48]. Overall posture was not improved by only using knee blocks [49].

3. Does sitting ability by special chairs and other device improve pulmonary function?
The review by Barks [50] analysed how pulmonary function was affected by positioning of persons with CP who lack trunk control. Twenty-eight references published from 1966 -2003 were included. All reflected either a physiologic or therapy-based model involving function in activities of daily living, but no other health outcomes.

Other articles: Three articles describe the effect on pulmonary function of two different sitting positions. A study of six children demonstrated that the anterior-tilted position does not improve respiratory function compared to horizontal sitting [51]. Children increased their vital capacity and forced expiratory volume in one second and expiratory time when seated in an adaptive seating system compared to a sling wheelchair [52]. A soft TLSO, did not have a negative impact on pulmonary mechanisms in persons with severe cerebral palsy [53].

Conclusion and future directions
Seating position is clearly important to optimising child's experience. However, on the basis from current evidence, it is difficult to come to any rigorous conclusions about the effect of particular seating positions on function. In general the research questions need to be more explicitly defined and build on recent theories for treatment and management. Research on this topic would benefit from larger sample sizes, longer follow up, greater precision in describing independent variables of positioning, and where possible the participation of children in assessing key outcomes.
Grade of Recommendation: C

HAND USE: CONSTRAINT-INDUCED MOVEMENT THERAPIES

Search strategy:
The electronic databases MEDLINE, CINAHL, the Cochrane Library, OTSeeker, PEDro, and PsycINFO were searched using the terms: cerebral palsy, hemiplegia, hemiparesis, constraint-induced movement therapy, constraint-induced therapy, CIMT, CIT, CI-therapy, forced use, MCIT, MCIMT, upper extremity. Only English language manuscripts were included. All age groups between newborn and 18 years of age were included. Articles including children with traumatic brain injuries were included only when they were grouped with children with congenital hemiplegia. Case studies and methodological papers were excluded. The search was completed by a research assistant trained in the appropriate search strategies.

Definition of terms:
The main principle of Constraint-induced movement therapy (CI therapy) is restraint of the uninvolved hand and intensive practice with the involved hand, i.e. hemiplegic hand, during a specified time period. The principle of CI therapy was developed through neurophysiological research by Dr Edward Taub during the 1950s.[54] Somatic sensation was unilaterally abolished in monkeys using dorsal rhizotomy. This caused the monkeys to stop using the affected arm unless the intact limb was restrained, in combination with practicing tasks using the deafferented limb, for one to two weeks. The monkey experiments formed the basis for intervention in adult humans who had sustained a stroke. Gradually the intervention was refined and eventually termed 'constraint-induced movement therapy' [55]. Subsequent studies of CI therapy have examined the efficacy of this intervention for improving involved upper extremity use. There has been experimentation with various types of restraint, interventions and outcome measures, and provided for people with chronic, acute and sub-acute stroke. [56] Today, for adults, restraint for 90% of waking hours is typically used, together with a structured training program.

The primary PICO research question:
This review will address if CI therapy, modified CI therapy and Forced Use (Intervention) will improve hand function (Outcome) in children with hemiplegic CP

(Population) compared to contemporary intervention (Comparison intervention). The question is formulated according to the PICO model. In addition the differences in treatment protocol, age and restraint will be discussed.

Primary findings
There have more than 30 articles describing CI therapy in children since late nineties. There are many case reports and small studies but recently, there have been somewhat larger studies as well. Twelve of them meet the inclusion criteria for this review (Table 1). One Cochrane review was published in 2008.

Cochrane Review
In the Cochrane review by Hoare et al. [57], three articles met the inclusion criteria. These were two RCTs and one controlled clinical trial (CCT) comparing CI therapy, modified CI therapy and Forced Use with traditional service or no treatment. The review includes articles published before August 2006. One RCT showed a trend for a positive effect for one subscale of the QUEST , [58] also partly described by [59]. The other assessments used, without reported psychometric properties, were also positive. The second RCT, [60] demonstrated improvement only on the self-care component in WeeFIM using Forced Use protocol. The other assessments used, without reported psychometrics, were also positive. The CCT was the largest study and demonstrate good effect by using Assisting Hand Assessment (AHA). In conclusion, although there was a positive trend from the four RCTs and one CCT, there is still weak evidence to support CIMT. An adequately powered RCT is recommended to provide further evidence.

Other articles
Four studies used a randomized design [59, 61-63]. These studies can be designated Level I [64] although they were small trials. One study was prospective with a control group [65] consistent with Level III. The remaining papers were level IV studies, three with an ABAA design, [65-68] two with pre and post treatment design [69, 70] and one study with repeated treatments [71]. All these studies reported positive results. However, despite the potential benefits of CIMT, the evidence to date, while promising, is still tentative.

Other results and ongoing studies
In adults, there have been studies reporting reorganization of hand motor pattern from functional magnetic resonance brain imaging. The first article in children with cerebral palsy was published 2007 [69], reporting increased cortical activation in some cases. One study, combining CI therapy with botulinum A toxin, showed no additional effect (Eliasson et al, submitted). There is at least one ongoing study which is further exploring this topic. Likewise there is one study in press combining CI therapy with intensive bimanual training demonstrating similar results (Hoare et al, personal communication). CI therapy has so far been compared to conventional therapy but the issue of intensity (frequency and duration or ‘dosage’) is yet to be resolved.

Conclusion and future directions
Large well controlled studies are lacking. Multi-centre studies are recommended since the prevalence of hemiplegic CP is fairly low. The evidence is still tentative but there are no studies reporting negative results. The training methods need further investigation, especially in relation to other types of intensive training programmes and with a longer follow up periods. No studies have so far been using orthoses in

combination with CI therapy and this might be an interesting avenue for further research.

SELF-CARE: FEEDING INTERVENTIONS

Search strategy:
The electronic databases that were searched included CINAHL, CIRRIE, the Cochrane Library, EMBASE, ERIC, HealthSTAR, Health and Psychosocial Instruments, MEDLINE, OTSeeker, PEDro, PsycINFO and RehabData, using the key words: tetraplegi*, spastic*, quadriplegi*, quadrapare*, pes equinus*, monoplegi*, little* disease, hypotonic*, hemiplegia*, hemipare*, dystoni*, diplegi*, dyskine*, choreoathe*, cerebral palsy, atheto*, ataxi*, feed*. Only English or French manuscripts were included, for publications up to and including December 2007. All age groups between 0 and 18 years of age and all study designs (e.g. RCTs, quasi-experimental, case series, case studies) were included. Medical interventions such as medications, nasogastric feeding, fundoplication or use of a GE tube were not included. Interventions to decrease drooling were also specifically excluded. Articles that were excluded focused predominantly on individuals with stroke, traumatic brain injuries, physical disabilities and adults. Studies with a heterogeneous sample of children with a variety of developmental disabilities to include cerebral palsy (but less than 50%) were excluded.

Definition of terms:
Feeding interventions (i.e. treatments) that are part of this review included individualized sensori-motor approaches, food consistency alterations, positioning techniques, oral orthodontic appliances and adapted equipment. *Sensori-motor* approaches include sensory stimulation (e.g. thermal application-typically cold, vibration around the mouth, stretch pressure to the oricularis oris, olfactory input) and oral-motor facilitation (e.g. massage gums, apply pressure to the teeth and tongue, maintain jaw control, side-placement of textured bolus) [72, 73]. Alteration in the *consistency* of food textures has been used as an intervention to optimize feeding performance [74]. *Positioning* techniques incorporate neurodevelopmental treatment principles and adapted equipment to optimally position the child for safe and effective task training and/or feeding [75]. *Oral appliances* are orthodontic removable plates that may be prescribed to improve oral-motor performance. They can include a tongue stimulator (a button-like protrusion or beads), a tongue shield in the midline or at the side, a lip stimulator with mobile beads over the front teeth, and/or vestibular pads [76, 77]. The occupational therapist carefully evaluates oral-motor abilities prior to and with daily use of the appliance, and monitors the potential benefits on feeding efficiency[78]. In terms of *adapted equipment*, the effectiveness of the Handy 1 Robotic Aid to Eating has been tested. Using a single switch device, food is presented in a controlled or consistent method using this assistive device [79, 80].

Purpose:
Feeding disorders (eating, swallowing) are a common associated condition in children with CP. These difficulties are often associated with poor oral motor control. As a result, these children are at high risk of aspiration, which may cause chest infections or can be potentially life threatening [81, 82]. Growth failure and inadequate caloric intake are important clinical concerns as well [83]. Feeding may take up to 6 hours or more each day for caregivers, and therefore contributes to burden of care [84]. A wide variety of intervention strategies have been evaluated to improve oral motor skills, so

as to be able to feed more safely and efficiently. These include impairment-based treatments (sensori-motor, positioning) as well as the application of modifications and adaptations (oral appliance, food consistency, electric feeder). The purpose of *sensori-motor* techniques is to decrease or increase tone and inhibit abnormal reflexes that are interfering with safe feeding, thus enabling training and strengthening of the oral musculature, and providing greater opportunities to learn and practice feeding skills [85]. Sensori-motor approaches are used to facilitate lip closure, tongue lateralization and rotary chewing, to inhibit tongue thrust and decrease tactile hypersensitivity and encourage swallowing [85, 86]. Solid soft versus mashed food *consistency* can influence chewing and swallowing abilities in children with CP.[87] *Positioning* techniques such as stabilization of the neck and trunk, forward neck flexion or supported recline positions may be used to promote more effective feeding behaviors. Optimal positioning techniques have been guided by videofluoroscopy images of eating and swallowing while in different positions [82, 88]. *Oral appliances* such as the Innsbruck Sensori-motor Activator and Regulator (ISMAR) [89] are used to improve orofacial sensori-motor functions such as sucking, coordination of chewing and swallowing. This appliance is used for jaw stabilization in the initial phase, followed by activation and mobilization of the oral and pharyngeal structures in the second phase. These removable plates may also improve facial expression. Use of an *electronic feeding device* (Handy 1 Robotic Aid) may be helpful in promoting independent self-feeding. This aid provides a predictable presentation of the food, but demands active participation of the child in removal of the food from the spoon.

Primary PICO research question: Does regular application of feeding interventions {sensori-motor, consistency of textures, positioning, oral appliance, adapted device} improve feeding efficiency (mastication, swallowing) and safety (decrease aspirations) and result in enhanced weight gain in children with cerebral palsy?
P: Children (most school age and/or adolescents) with cerebral palsy
I: Impairment-based interventions or modifications/appliances/equipment, regular/daily, either immediate effects or over several months
C: Compared to baseline or to a control group
O: Highly variable: measures of *body structure* (videofluoroscopy), *body function* (anthropometric measurements, pathologic reflexes, oral-motor observations), *activity and participation* (feeding performance, speech skills).

Primary findings:
Seventeen studies that described the effectiveness of particular feeding interventions in children with cerebral palsy were reviewed. Descriptive information regarding these studies appear in Table E and F. Children were typically of school age or adolescence and had moderate to severe motor impairments, although type and severity of CP were often not described. Sample size for 13/17 studies was small (up to 20 subjects). Statistical approaches were primarily descriptive in most studies. Designs were predominantly quasi-experimental (with comparison or control groups) or case series with pre-post testing, with only one randomized controlled trial (although randomization method was not specified). Sensori-motor interventions and oral appliances were typically applied daily for several months, whereas interventions that focused on positioning, consistency changes or adapted equipment involved observations over single or multiple sessions only. Only one study incorporated a multi-modal approach to include oral-motor interventions, behavioral techniques, positioning and caregiver education [90]. Outcome measures were highly variable but often included anthropometric measures, videofluoroscopy, feeding observations with video, and standardized feeding measures. Results to date provide preliminary

evidence supporting the effectiveness of feeding interventions. Specifically, sensori-motor approaches improved oral-motor skills and reduce aspirations, especially in more impaired children (i.e. those who aspirated) [85, 86, 91-93] A single study demonstrated that mashed foods were ingested more rapidly and more safely than semi-solid foods [87]. Individualized positioning programs guided by videofluoroscopy improved eating and swallowing skills [82]. Furthermore, positioning helped to promote successful self-feeding behaviors when accompanied with task training (practice, physical assistance) of self-feeding components such as reach, grasp, and hand to mouth movements [94]. However, for some children, positioning does not facilitate safe feeding and therefore tube feeding or gastrostomy may be necessary [82]. Use of the ISMAR oral appliance for children with moderate dysphagia enhanced mastication and swallowing skills in most children, improved sensibility of the oral cavity, facilitated tongue mobility and lip and jaw control, improved resting posture of the tongue and lips. In addition, there were improvements noted in generalized motor control and facial expression was enhanced as well ([76, 84, 95, 96]. Use of an electronic feeder (assistive device) enabled children to feed independently, although feeding rate was slower for children with some speech skills, when compared to being fed by a caregiver. It was suggested that in higher functioning children, the device may be interfering with conversation and social interaction. While the investigators noted that oral-motor skills improved with this consistent feeding presentation approach, however the device cannot handle all food types therefore limiting the feasibility of use for all meals [97, 98]. For all these feeding interventions, no significant changes to weight gain were noted (except for Clawson et al [90]), therefore caloric supplementation for "catch-up" is often still needed to maximize growth. Improvements in most studies were often not maintained on follow-up (post-intervention). These interventions appeared to be well tolerated overall.

Review articles:
To our knowledge, there are no "systematic" reviews in this topic area. Rogers [5] published a review article on feeding interventions in children with CP. Oral sensori-motor interventions are noted as the most common therapeutic strategy used clinically. Evidence suggests that this intervention approach improves oral-motor skills but not airway protection, feeding efficiency or weight gain. In terms of medical interventions, gastrostomy tube feeding remains an important alternative approach for this population, to prevent growth failure. Gisel (in press) also provides an overview of feeding interventions for children with dysphagia. She concludes that sensori-motor interventions and oral appliances are both effective in improving oral-motor skills and enhancing the efficiency of food intake.

Conclusions and future directions:
Feeding disorders can impact on nutrition, growth and development, respiratory and gastrointestinal functioning, parent-child interactions and family burden [5]. Sensori-motor interventions provided regularly over weeks to months may improve oral-motor skills such as food retention, biting and chewing. Individualized positioning techniques can also optimize oral-motor control. Preliminary evidence would indicate that adjusting food consistency (e.g. thickening of liquid foods) may also be helpful in improving feeding performance. Use of an oral appliance enhances oral sensory perception, jaw stabilization and tongue mobilization with subsequent improvements in feeding skills. Of importance, use of the oral appliance may be associated with improved facial expressions and mouth closure, which has social benefits [84]. Results of a pilot study on preschoolers with spastic diplegia suggested that biological

(oral-motor, positioning), behavioural (positive reinforcements, extinction of negative behaviours) and environmental (parent education) aspects of feeding disorders are mutually interactive. This recent study [90] is the only intervention that applies a multi-modal approach. Interestingly, improvements noted on descriptive analysis included improved oral-motor skills, enhanced caregiver abilities and most importantly, percentile improvements for weight and height. In contrast, weight gain, a marker of feeding efficiency, did not improve in all other feeding interventions studied to date.

Oral feeding experiences, even if difficult, are important to the child and family. Single-modality approaches such as sensori-motor stimulation or positioning yield modest improvements in feeding ability. Consideration of complementary medical interventions such as caloric supplementation with commercial products or use of gastrostomy tubes may be necessary to ensure adequate nutrition and growth [5, 99]. While there are some indications for placement of feeding G-tubes (e.g. children with malnutrition, moderate to severe aspiration, and/or aversive behaviour during oral feeding), a recent Cochrane systematic review (2004) [99] concludes that considerable uncertainty remains regarding the effectiveness of gastrostomy for children with cerebral palsy. Future studies should evaluate the effectiveness of multi-modal (medical, oral-motor, behavioural and environmental interventions and adaptations) approaches to improve feeding performance, growth and development. Clearly, well designed randomised controlled trials with long-term outcomes should be undertaken to evaluate the effectiveness of these multi-modal approaches. **Grade of Recommendation: B-C** (note: evidence was not consistent enough to warrant a grade of B for any one category, although somewhat stronger evidence than for other interventions)

PLAY: VIRTUAL REALITY

Search strategy:

The electronic databases that were searched included CINAHL, CIRRIE, the Cochrane Library, EMBASE, ERIC, HealthSTAR, Health and Psychosocial Instruments, MEDLINE, OTSeeker, PEDro, PsycINFO and RehabData, using the key words: tetraplegi*, spastic*, quadriplegi*, quadrapare*, pes equinus*, monoplegi*, little* disease, hypotonic*, hemiplegia*, hemipare*, dystoni*, deplegi*, dyskine*, choreoathe*, cerebral palsy, atheto*, ataxi*, virtual reality. Only English or French manuscripts were included, for publications up to and including December 2007. All age groups between and 18 years of age and all study designs (e.g. RCTs, quasi-experimental, case series, case studies) were included. Articles that were excluded focused predominantly on individuals with stroke, traumatic brain injuries, physical disabilities and adults.

Definition of terms:

Virtual reality (VR) is a virtual environment system that uses a range of computer technologies that present virtual or artificially generated sensory information in a format that enables the user to perceive experiences that are similar to real-life events and activities [100]. A three-dimensional simulated environment of objects and activities is created and presented either on a computer monitor, or projected on a large screen or within a helmet-mounted display. Specific input devices (e.g. dataglove) allow the user to interact actively with the virtual environment. VR provides the user with the opportunity not only to sense the virtual environment, but

also to interact and explore the environment, relatively free of limitations imposed by their disability. Multi-user formats, which are becoming increasingly commercially available, can provide new social and recreational opportunities to enhance participation for individuals with functional limitations [100]. With respect to rehabilitation, VR is used to create an interactive play environment to achieve particular occupational therapy treatment goals. Note that the outcomes may include improved play skills or play attitudes, but for several studies, also include improvements at other levels (see below). This modality can be used either to evaluate level of performance within a simulated task, or more commonly as a treatment approach. For the latter, VR can incorporate motor learning principles such as repeated practice and positive feedback, so that play-based training of everyday functional activities can occur. Precise laboratory measurements may be made to quantify performance over time. Furthermore, the task demands (e.g. speed, accuracy, complexity of the task) and the type and frequency of feedback can be manipulated and graded, so as to grade performance requirements [101-103]. In addition, benefits of this novel, animated treatment approach include enhancement of creativity and pleasure, a greater degree of control and increased mastery motivation (i.e. persistence with tasks) [102].

Purpose:
VR has been used primarily in research as a new treatment modality for children and youth with cerebral palsy. Motor learning concepts to include the use of feedback and practice are applied as facilitators of motor performance. Through virtual games, children engage in multiple trials of a specific task with ongoing feedback on performance success. The children see themselves on a screen within a virtual environment that responds to their movements [104]. The primary purpose is to improve competence and confidence in motor-based activities [105] and also to engage in play-based activities that are otherwise inaccessible in the real world [106]. Research studies demonstrate that this modality is feasible, highly enjoyable and non-threatening [107]. VR can be used for task training (e.g. electric wheelchair mobility), by initially simplifying the task and subsequently grading the complexity of the task demands. This modality is also advantageous for children with cerebral palsy as it enables the subject to spatially explore complex environments that would be otherwise impossible due to mobility restrictions. VR provides the opportunity to overcome existing obstacles to navigating and exploring the environment safely, thus promoting visual-spatial and perceptual-motor concept formation [101]. Furthermore, spatial and motor learning through virtual environments have been shown to transfer into real environments in adults and in children with other disabilities [100, 101].

Primary PICO research question: Does weekly repetitive training using a play-based VR intervention improve **{**brain reorganization/ motor skills/ spatial skills/ motivation and playfulness/ functional everyday self-care and leisure activities**}** in children with cerebral palsy?
P: Children (4-14 years; most 8-12 years) with cerebral palsy
I: Virtual games, weekly sessions, for 4-8 weeks
C: Compared to baseline or to a control group
O: Highly variable: measures of *body structure* (fMRI), *body function* (motor skills, quality of movements, visual-spatial skills), *activity and participation* (self care and leisure activity performance, playfulness), *personal factors* (motivation, self-perception, self-efficacy).

Primary findings:
Eight studies described the effectiveness of VR for children with cerebral palsy. Descriptive information regarding these studies appear in Table G and H. Children were typically of school age (4-14 years) and had cerebral palsy, although type and severity were often not described. When specified, studies included children who were non-ambulatory, but not exclusively. Sample size for 6/8 studies was small (1-16 subjects). Statistical approaches were primarily descriptive in most studies. Designs varied from observational studies (2), case studies with pre-post testing (4), an experimental study with a control group (1) and one randomized controlled trial. Interventions typically involved weekly VR sessions for 1-2 months. A variety of virtual games were used in these studies, with different goals. As a result, outcome measures were variable and included measures of brain structure and function, motor and cognitive ability, activity limitations in self care and leisure, enjoyment of play and self-perception. Preliminary results indicate that intense use and practice provided by VR may contribute to improved neuroplasticity by facilitating neural motor pathways that are under-utilized. This reorganization seems to be closely associated with the enhancement of age-appropriate skills in the affected limb [108]. The primary objective of the VR training used by Akhutina et al [101] was to enhance spatial skills in children with limited navigation experiences. Children were assigned to an experimental (with VR training) or control group and a wide range of computer-based and standardized outcome measures were used. The first experiment yielded no significant effects, possibly due to differences in level of motor impairment between experimental and control groups, as well as the complexity of the training task. In experiment two, simpler and more varied spatial cognition tasks to include training of spatial terminology was used on larger and more functionally comparable children assigned to experimental and control groups. The experimental group showed significant improvements in visual-spatial abilities following VR training. With repeated practice (e.g. about 100 reaches per 3-minute session), Chen et al [102] noted improvements in accuracy and quality of reaching and in fine motor skills. On follow-up, some maintained changes or continued to improve but this was not consistent in all subjects. VR environments stimulated playfulness (motivation, control, suspension from reality, framing) except for games that were too challenging [106], was highly motivating for children [104, 109] and improved self-perceptions and self-efficacy [104, 107].

Review articles:
No review articles on VR were found that focused specifically on the target population (cerebral palsy).

Conclusions and future directions:
The literature in this area is now emerging (since 2002) and remains fairly preliminary, however evidence to date suggests that VR may prove to be a useful therapeutic modality for children with cerebral palsy. A wide range of potential uses of VR are being proposed and tested, therefore validation of findings is needed. Practice effect and lack of use of blinded evaluators may in part explain improvements on outcome measures. Studies suggest that repetitive training offered by VR play-based activities can facilitate motor and visual-spatial skills, and may influence brain plasticity and reorganization. Furthermore, this modality is highly motivating and engaging, therefore if widely available, compliance and potential benefits are likely to be high. The long-term benefits are unknown. Randomized controlled trials on large samples are needed to ascertain whether this approach is better than traditional rehabilitation interventions [101, 102]. In addition to improving

function, this modality may prove useful in promoting social participation through virtual environments. Greater clarity is needed with respect to which children (type and severity of CP, level of cognitive function required) and which treatment outcomes should be targeted. **Grade of Recommendation: C**

LINKING OCCUPATIONAL THERAPY AND ORTHOTICS:

Orthoses are devices that are applied externally to the child in order to modify structural and functional attributes of the neuromuscular and musculoskeletal systems . Occupational therapists may also use a variety of devices and adaptations, such as upper extremity splinting and casting, seating devices and oral appliances. The use of orthoses may be linked to lessons learnt from Occupational Therapy intervention studies in a number of ways:

- Clinically, these devices (used by orthotists and/or OTs) are helpful in providing stability around joints, facilitating movement, and preventing deformities. As such, they provide biomechanical improvements that may help to overcome limitations in activity and restrictions in participation. However, promotion of activity and participation must be explicitly facilitated (concurrent with use of these devices) with prescribed intervention strategies, and these outcomes need to be measured, in determining the effectiveness of orthotic use. This has been largely lacking in the OT literature.

- Child (client-centred practice) and family (family-centred care) preferences should be considered in the selection and use of devices and adaptations. In order to maximize compliance, the child must be willing to apply the device. Furthermore, family members need to understand the underlying mechanisms and potential benefits of the device, so as to ensure that they incorporate its use as part of daily life activities. Any negative consequences of use of the devices (e.g. self-image, sleep disturbances, discomfort) need to be "solved" by the treatment team, in partnership with the child and family.

- Most OT intervention studies we reviewed focused on a single treatment modality (e.g. a specific device or a particular intervention approach). In reality, clinicians apply multi-modal (remediation, compensation, adaptation) strategies, intervening at the body function, activity/participation and/or environmental levels, with direct treatments (e.g. to increase hand reach, learn to eat with a spoon, recognize socially acceptable feeding behaviors) and with application of particular adaptations or devices (e.g. seating device, oral appliance, less distracting environment). For example, a recent feeding study or current randomized trials using Botox that incorporate function-based interventions using activities preferred by the child, can serve as models.

In summary, disability is the product of characteristics of the individual, such as impairments and activity limitations and personal factors, as well as attributes of the individual's physical, social and attitudinal environment. Therefore, future trials should adopt an integrated "multi-modal" approach with emphasis on the child and family's needs, and selection of interventions with greater focus on maximizing community integration and participation, either by enhancing abilities (e.g. at body function and activity level) and/or by modifying the task or environment. Orthotic

devices are one piece of that puzzle. Many possible approaches may be considered to address functional challenges, and the home, school and community environments may each require different strategies to ensure optimal functioning. Goals of treatment need to be individualized rather than prescriptive, that is, developed with consideration of the family's priorities and the child's preferences.

TABLE A: Upper Extremity Splinting

Authors, year	Title	Oxford score	Splint type
McPherson, 1991[15]	Objective evaluation of a splint designed to reduce hypertonicity	4	Snook's spasticity reduction splint
Exner & Bonder, 1983 [7]	Comparative effects of three hand splints on bilateral hand use, grasp, and arm-hand posture in hemiplegic children: A pilot study	2b	3 types: orthokinetic cuff, short opponens thumb splint, MacKinnon splint
Currie & Mendiola, 1987 [8]	Cortical thumb orthosis for children with spastic hemiplegic cerebral palsy	4	Cortical thumb orthosis
Flegle & Leibowitz, 1988 [9]	Improvement in grasp skill in children with hemiplegia with the MacKinnon splint	4	MacKinnon splint
Goodman & Bazyk, 1991l [10]	The effects of a short thumb opponens splint on hand function in cerebral palsy: A single-subject study	4	Short thumb opponens
Kerem et al, 2001 [11]	Effects of Johnstone pressure splints combine with neurodevelopmental therapy on spasticity and cutaneous sensory inputs in spastic cerebral palsy	2b	Johnstone pressure splint
Reid, 1992 [12]	An instrumentation approach for assessing the effects of a hand positioning device on reaching motion of children with cerebral palsy	4	Hand positioning device (aligns hand with forearm, resting position of wrist and fingers)
Reid & Sochaniwskyj, 1992 [13]	Influences of a hand positioning device on upper-extremity control of children with cerebral palsy	4	Hand positioning device (wrist slightly extended, thumb abducted and opposed, and MCP joints and fingers in slight flexion)
Kinghorn & Roberts, 1996 [14]	The effect of an inhibitive weight-bearing splint on tone and function: A single-case study	4	Inhibitive weight-bearing splint

Oxford Score:
1b: Individual RCT study
2b : Experimental cohort study with well-defined control group, *or* low quality RCT
3b: ABA design
4: Quasi-experimental = to include either a pre-post design or descriptive cohort study

TABLE B: Upper Extremity Splinting

Authors, year	Subjects; sample size; age range	Methodology	Outcome measures / Variables *	Main findings
McPherson, 1981[15]	Increased wrist flexor tone; N=5 (7 hands); 10-18 years	Quasi-experimental; Incremental exposure (0-60 min., 2x/day), 5 weeks (compared to pretest)	Hypertonicity	Reduction in hypertonicity with increased exposure to splint. No permanent changes noted. *Results were tested statistically.*
Exner & Bonder, 1983 [7]	Hemiplegia; N=12; 3-16 years	Randomized control trial; 8 hrs/day, 6 weeks per splint, 2 week rest periods	Bilateral hand use, grasp skill, arm-hand posture	Improvements over time in orthokinetic cuff (41.6% bilateral skills) and MacKinnon (58.3% improved bilateral hand use and/or grasp; no differences between splints used. More impaired children had greater changes noted. *Results were tested statistically.*
Currie & Mendiola, 1987 [8]	Hemiplegia (moderate); N=5; 20-26 months	Quasi-experimental, compared to baseline; 1 week (pre-post)	Observation of the hand position and function	Improved position of the thumb, improved prehension and function. *Conclusions were based on descriptive data.*
Flegle & Leibowitz, 1988 [9]	Hemiplegia; N=3; 2.5-7.5 years	Multiple baseline; 8 hrs/day	(Grasp skill test)	Improvements in grasp skills, especially for those with poor grasp initially. *Conclusions were based on descriptive data.*
Goodman & Bazyk, 1991 [10]	Quadriplegia (moderate); N=1; 4 year old	A-B single subject; 3 hrs, 2x/day and overnite, 4 weeks baseline, 4 weeks splinting	Active range of motion, grip and pinch strength, (Box and Block tests) stacking cubes, lateral pinch	Improvements in areas assessed except palmar and tip pinch strength. *Conclusions were based on descriptive data.*
Kerem et al, 2001 [11]	Diplegia; N=34, 36-82 months	Experimental; 20min/day, 3 months (compared to NDT)	(Goniometry, Modified Ashworth Scale, somatosensory evoked potentials)	Improvements in range of motion, spasticity and SEP. *Results were tested statistically.*
Reid, 1992 [12]	Spastic quadriplegia, athetoid, dyskinetic; N=5; 5.8-11.1 years	Quasi-experimental; 1 hr/day, 6 weeks	(3D tracking system for upper extremity reaching)	Differing improvements in upper extremity reaching in individual children, but no significant differences for group data. *Results were tested statistically.*

TABLE B Upper Extremity Splinting (continued):

Authors, year	Subjects; sample size; age range	Methodology	Outcome measures / Variables *	Main findings
Reid & Sochaniwskyj, 1992 [13]	Spastic and athetoid cerebral palsy; N=10; 5-12 years old	Before-after trial; 3 sessions	(EMG, 3D motion analysis, "home-made" visual-motor performance test [based on Beery Development Test of VMI, Frostig Developmental Test of Visual Perception,] Miller Assessment for Pre-schoolers and BOTMP)	While wearing the splint, differences noted in the average velocity of movement, movement time, movement units in 8/10 individuals; muscle activation patterns resembled normal patterns in several children. Scores on visual-motor tests were not statistically significant (group). *Results were tested statistically.*
Kinghorn & Roberts, 1996 [14]	Quadriplegia; N=1; 20 months	Single-subject; 3 x 8 weeks	Hand tracing, arm-hand posture, functional activities (block play for active grasp, voluntary release, ball play)	No effects. *Conclusions were based on descriptive data.*

Hr(s): hour(s); min.: minutes; EMG: electromyography;
VMI: Visual Motor Integration; BOTMP: Bruininks-Oseretsky Test of Motor Proficiency

- Outcome measures/Variables: when studies specified actual measures used, these are indicated in the column, otherwise the variables observed and documented are specified.

TABLE C: Upper Extremity Casting

Authors, year	Title	Oxford score	Cast type
Smith & Harris, 1985 [18]	Upper extremity inhibitive casting for a child with cerebral palsy	4	Upper extremity inhibitive casting (elbow)
Smelt, 1989 [19]	Effect of Inhibitive weight-bearing mitt on tone reduction and functional performance in a child with cerebral palsy	4	Inhibitive weight bearing mitt (removable plaster cast of hand)
Yasukawa, 1990 [20]	Upper extremity casting: Adjunct treatment for a child With cerebral palsy hemiplegia	4	Long arm cast on impaired extremity (elbow, wrist, hand) and short arm cast on unimpaired (wrist, hand)
Law, et al. 1991 [21]	Neurodevelopmental therapy and upper-extremity inhibitive casting for children with cerebral palsy	2b	Upper extremity inhibitive bi-valve cast (wrist)
Tona & Schneck, 1993 [22]	The efficacy of upper extremity inhibitive casting: a single-subject pilot study	4	Upper extremity inhibitive casting
Copley, et al. 1996 [23]	Upper limb casting for clients with cerebral palsy: a clinical report	4	Upper limb serial casting (wrist and/or elbow)
Law, et al. 1997 [26]	A comparison of intensive neurodevelopmental therapy plus casting and a regular occupational therapy program for children with cerebral palsy	2b	Upper extremity inhibitive casting (wrist)
Yasukawa, Malas, & Gaebler-Spira, 2003 [25]	Efficacy for maintenance of elbow range of motion of two types of orthotic devices: A case series	4	Serial casting of both elbows (bi-valve vs. Ultraflex casts)

TABLE D: Upper Extremity Casting

Authors, year	Subjects; sample size; age range	Methodology	Outcome measures / Variables *	Main findings
Smith & Harris, 1985 [18]	Severe spastic quadriplegia; N= 1; 5.5 years old	Case study; 3 hrs/day, 13 months	Range of motion (goniometry)	Helped maintain range of motion in both elbows and prevented the increase of contractures at the elbow joint. *Conclusions were based on descriptive data, observed over time.*
Smelt, 1989 [19]	Spastic hemiplegia; N=1; 17 months	Single subject ABA design; twice/week for 15 minutes,13 weeks	Passive range of motion of hand, tone of hand, Erhardt Developmental Prehension Assessment	Increase in grip circumference (passive range of motion), hand weight-bearing surface area (tone in hand) and hand use. However, the two latter gains were lost at follow-up. *Time series: Descriptive analysis; C statistic*
Yasukawa, 1990 [20]	Spastic diplegia; N=1; 2 years, 9 months	Single subject case study; casted for 4 weeks	Observation	Elbow extension and forearm supination improved, strength of right arm also increased. *Statistical analysis for single subject designs.*
Law, et al. 1991 [21]	Quadriplegia and hemiplegia; N=73; 1.5-8 years	Randomized control trial (casting with or without NDT); minimum 4 hrs/day, 6 months	Peabody Developmental Motor Scales–Fine Motor, Quality of Upper Extremity Skills Test (QUEST)	Casting in combination with neurodevelopmental therapy program resulted in improved upper extremity movement quality and wrist extension *Descriptive and statistical analyses.*
Tona & Schneck, 1993 [22]	Cerebral palsy; N= 1; 8.5 years	Single subject (AB withdrawal); 11 days	Observation, Modified Ashworth Scale, Biodex system	Reduced tone and gains in active and passive range of motion. However, these gains were not maintained at follow-up. *Statistical analysis for single subject designs.*
Copley, et al. 1996 [23]	Spastic quadriplegia and hemiplegia; N= 11; 5.3-17.8 years	Quasi-experimental; 4-6 weeks	Range of motion (goniometry), Spasticity (patterns of tome reduction observed), Achievement of functional goals	Spasticity was reduced in all and ROM improved after treatment. Improvements were maintained on 6-month follow-up. Improved function associated with less spasticity. *Percentage change noted.*
Law, et al. 1997 [26]	Diplegia, hemiplegia, quadriplegia; N=50; 1.5-4 years	Randomized controlled trial (crossover); at least 4 hrs/day, 4 months	Peabody Developmental Motor Scales –Fine Motor, Quality of Upper Extremity Skills Test, Canadian Occupational Performance Measure, parent's perception of improvement	No improvement was noted in hand function, quality of upper extremity movement or parents' perception of hand function after treatment. *Descriptive and statistical analyses of results.*

TABLE D Upper Extremity Casting (continued):

Authors, year	Subjects; sample size; age range	Methodology	Outcome measures / Variables *	Main findings
Yasukawa, Malas, & Gaebler-Spira, 2003 [25]	Quadriplegia (spasticity at elbows); N= 3; 7 years	Case series; 4 hrs/day and at night	Modified Ashworth Scale, passive range of motion, caregiver questionnaire	The Ultraflex cast proved to better maintain the range of motion at the elbows and facilitated length-tension adjustments. *Descriptive statistics, changes over time.*

hr(s): hour(s); min.: minutes

* Outcome measures/Variables: when studies specified actual measures used, these are indicated in the column, otherwise the variables observed and documented are specified.

Table E Seating & positioning

Authors, year	Title	Oxford	Type
2007, Vekerdy [46]	Management of seating posture of children with cerebral palsy by using thoracic-lumbar-sacral orthosis with non-rigid SIDO frame.	2b	Thoracic-lumbar sacral orthosis with rigid SIDO frame
2007, McDonald and Surtees [49]	Longitudinal study evaluating a seating systems using sacral pads and kneeblocks for children with cerebral palsy	3b	Kneeblock,
2007, Hatta et al [43]	Evaluating the relationships between the postural adaptation of patients with profound cerebral palsy and the configuration of the Seating Buggy's seating support surface.	2b	wheelchair fitting in the Seating Buggy
2007, Michael et al [41]	Tilted seat position for non-ambulant individuals with neurological and neuromuscular impairment: a systematic review.	2b	Systematic review
2007, McNamara & Casey [42]	Seat inclinations affect the function of children with cerebral palsy: A review of the effect of different seat inclines	2b	Systematic review
2006, Stavness [27]	The effect of positioning for children with cerebral palsy on upper-extremity function: a review of the evidence	2b	Systematic review
2004, Barks [50]	Therapeutic positioning, wheelchair seating, and pulmonary function of children with cerebral palsy: a research synthesis	2b	Systematic review
2004, Shoham et al [45]	The influence of seat adjustment and a thoraco-lumbar-sacral orthosis on the distribution of body-seat pressure in children with scoliosis and pelvic obliquity.	3b	orthosis
2003, Holmes et al [44]	Management of scoliosis with special seating for the non-ambulant spastic cerebral palsy population--a biomechanical study	3b	Special seating
2001, Rigby et al [110]	Effects of a wheelchair-mounted rigid pelvic stabilizer on caregiver assistance for children with cerebral palsy	3b	wheelchair-mounted rigid pelvic stabilizer
1999, Leopando et al [53]	Effect of a Soft Boston Orthosis on pulmonary mechanics in severe cerebral palsy.	4	Soft Boston
1999, Reid et al [32]	Functional impact of a rigid pelvic stabilizer on children with cerebral palsy who use wheelchairs: users' and caregivers' perceptions.	3b	chair mounted rigid pelvic stabilizer compared with a traditional wheelchair lap belt
1996, Reid [48]	The effects of the saddle seat on seated postural control and upper-extremity movement in children with cerebral palsy.	2b	saddle seat
1995, Myhr et al [31]	Five-year follow-up of functional sitting position in children with cerebral palsy.	4	Abduction orthosis and Functional Sitting Adaptation
1993, Myhr and von Wendt [30]	Influence of different sitting positions and abduction orthoses on leg muscle activity in children with cerebral palsy.	4	Abduction orthosis and Functional Sitting Adaptation
1993, Miedaner and Fiore [35]	Effects of adaptive positioning on psychological test scores for preschool children with cerebral palsy	1b	Fitted wheelchair
1992, McClenaghan [38]	Effects of seat-surface inclination on postural stability and function of the upper extremities of children with cerebral palsy.	2b	seat-surface
1991, McPherson [15]	Analysis of upper extremity movement in four sitting positions: a comparison of persons with and without cerebral palsy	2b	Seat angle
1991, Myhr and von Wendt [30]	Influence of different sitting positions and abduction orthoses on leg muscle activity in children with cerebral palsy.	4	Abduction orthosis and Functional Sitting Adaptation

Table E Seating & positioning (continued)

Authors, year	Title	Oxford	Type
1991, Reid and Sochaniwskyj [51]	Effects of anterior-tipped seating on respiratory function of normal children and children with cerebral palsy	2b	Seat angle
1989, Noronha et al. [36]	The effect of positioning on the hand function of boys with cerebral palsy.	3b	Standing sitting and prone
1987, Hulme et al [34]	Effects of adaptive seating devices on the eating and drinking of children with multiple handicaps.	4	Adaptive sitting device
1987, Nwaobi [40]	Seating orientations and upper extremity function in children with cerebral palsy.	4	Seat angle
1986, Colbert et al [33]	DESEMO seats for young children with cerebral palsy.	4	DESMO seat
1986, Nwaobi and Smith [52]	Effect of adaptive seating on pulmonary function of children with cerebral palsy.	4	Adaptive seating system
1984, Seeger BR. [39]	Will increasing hip flexion above 90 degrees would improve hand function in seated cerebral-palsied children	4	Seat angle

Table F Seating & positioning

	Aim	Subject, age	Outcome	Main finding	Methodology
Reviews					
2006, Stavsnees [27]	Investigating the effect of positioning on upper-extremity function: A review of the evidence	16 articles after 1980 up to?		The evidence supports the positive effect of a neural to slightly forward orientation	Small studies , few RTC
2007, Michael et al [41]	Does tilted seat position have an effect for non-ambulant individuals	19 articles, 10 for CP		Posterior tilting can reduce pressures at the interface under the pelvis	Small studies , few RTC
2007, McNamara and Casey [42]	A review of how Seat inclinations affect the function	10 articles 1990-2006		Neutral and anterior seat do positively affect function and individual adaptation are preferable	Small studies , few RTC
2004, Barks [50]	Does the seating effect pulmonary function	28 articles		Not possible to sum up the result	Small studies , few RTC
Improved posture by adapted seating and special device					
2007, Vekerdy [46]	To investigate the effects of a special seating device by using thoracic-lumbar-sacral orthosis with non-rigid SIDO frame.	47 children mean age of 53.2 months +/- 30.1 SD	X-ray questionnaires about daily activities	Regular application of the TLSO-SIDO had a beneficial effect on feeding problems and on posture.	Experimental prospective study, pre- and post-intervention
2007, McDonald and Surtees [49]	Evaluation of Adaptive seating systems using sacral pads and kneeblocks for improving the posture and stability in sitting.	23 children, 7 - 14 years	Measure of force	The seating systems using a sacral pad and kneeblock may not improve overall posture but may improve hip position in with cerebral palsy.	Single-subject design Repeated measure (6 times)
2007, Hatta et al [43]	Evaluating of the relationships between postural adaptation and the configuration of the Seating Buggy's seating support surface.	21 children	Active Balanced Seating scale and by a seating expert.	Properly adjusted depth of thoracic support and distance of the lumbar support were related to satisfactory head-neck alignment and sitting balance of the patients with profound cerebral palsy.	Experimental Comparative study. Postural adaptation were compared for 11= appropriate or nearly appropriate fitting, 10= ill fitting
2004, Shoham et al [45]	To determine the effect of a thoraco-lumbar-sacral orthosis (TLSO) on the distribution of body-seat interface pressure in children with concomitant scoliosis and pelvic obliquity	15 children with CP and scoliosis and pelvic obliquity when seated	Measure maximum pressure, mean pressure and contact area X-ray	Application of a TLSO significantly reduced the spinal curvature and interface sitting pressure. Manipulation of sitting by use of wedges under the pelvis had no significant effect on pressure	Descriptive study Data collection at baseline and at 10 degrees 'push up' and 10 degrees wedge insertion, with and without the TLSO.
2003, Holmes et al [44]	To investigate the effects of special seating on lateral spinal curvature in the non-ambulant spastic cerebral palsy population with scoliosis.	??	The shape of the spine was measured	Significant static correction of the scoliotic spine can be achieved. By the careful configuration of the lateral pads, significant correction of a scoliosis can be obtained.	Descriptive

Table F Seating & positioning (continued)

	Aim	Subject, age	Outcome	Main finding	Methodology
1999, Reid et al [32]	Evaluation of the functional impact of a Rigid Pelvic Stabilizer (RPS) compared with a wheelchair lap belt	6 children mean age 10,4	COPM, a structured weekly interview	Clinically significant changes in task performance and satisfaction when the RPS was worn as compared to the lap belt for all subjects.	ABA design
1995, Myhr et al [31]	A five year follow up of functional sitting position	10 children	Scoring of video-filmed sitting position	8 of the 10 who had used the functional sitting approach had slightly improvement	Comparative Study
1993, Myhr and von Wendt [30]	Influence of different sitting positions and abduction orthosis on leg muscle activity	8 children with CP and controls	EMG	Abduction orthosis and horizontal and forward-leaning seats might decrease lower-extremity muscle activity, and improve upper-extremity function.	Comparative Study.
1991, Myhr and von Wendt [29]	Investigation of best sitting position	23 children	Scoring of video-filmed sitting position	Pathological movements were minimised, postural control and arm and hand function improved with a forward-tipped seat, with a firm backrest supporting the pelvis, arms supported against a table and feet permitted to move backward.	Descriptive AB
Seating and pulmonary function					
1999 Leopando et al [53]	Effect of a Soft Boston Orthosis on pulmonary mechanics in severe cerebral palsy.	12 children 5-23 yr	Pulmonary resistance, compliance, tidal volume, minute ventilation, work of breathing, oxygen saturation, and end-tidal CO2 tension were measured	Soft Boston Orthosis does not impact negatively on pulmonary mechanics and gas exchange in young people with severe cerebral palsy.	Comparative study, AB with and without the orthosis and in the supine position without the orthosis.
1991, Reid and Sochaniwsk [51]	Effect on anteriorly-tipped seating on respiratory function	12 children, 6 control 6 CP, mean age 6 years	respiratory parameters of tidal volume, respiration rate, and minute ventilation	No significant differences in respiratory parameters could be attributed to seat base position	Experimental Prospective control
1986, Nwaobi and Smith [52]	Effect of adaptive seating on pulmonary function of children with cerebral palsy.	8 children. 5-12 yr	The vital capacity (VC), forced expiratory volume in one second (FEV1) and expiratory time (ET)	Increase in VC, FEV1 and ET in the adaptive seating system compared with the standard wheelchair.	Comparative study, AB
Does sitting ability by special chairs and other device improve upper extremity function					
2001, Rigby et al [110]	Investigation of the impact of a Rigid Pelvic Stabilizer (RPS) compared with a traditional lap belt.	6 children	Five bimanual or reaching tasks was evaluated using a six-point scale by parents.	The RPS appeared to impact directly on reducing caregiver assistance for 30% of the tasks,	ABA design

Table F Seating & positioning (continued)

	Aim	Subject, age	Outcome	Main finding	Methodology
1996, Reid [48]	Evaluate upper extremity movement using the saddle seat versus flat bench		Time and path length to reach	No significant difference	Experimental cross over design
1993, Miedaner and Finuaf [35]	Evaluation of the relation between unsupported sitting versus fitted	N= 12	Cognitive test with reach and grasp item	Improved upper limb function for fitted seated in tilted wheelchair	RTC
1992, McClenaghan et al [38]	Relation between seat angel and functional use of upper extremity	N=10 CP N=10 control	Time and accuracy	Seat angle do not affect upper extremity motor performance	Prospective Case Control study
1991, McPherson [15]	Relation between seat angel and functional use of upper extremity	N=10 adolescents	Number of movement segments to complete reaching task	No significant difference in quality of arm movements	Experimental cross over design
1989, Noronha et al. [36]	The effect of positioning on the hand function of boys with cerebral palsy.	10 boys (mean age = 12.5 years, SD = 1.2 years)	Jebsen-Taylor Hand Function Test, A scale modified from Hohlstein (1982) was used to measure quality of grasp on each subtest of the Jebsen-Taylor test.	No significant improvement for between sitting and standing, Improved ability for feeding and reaching/ handle small objects	Randomized Controlled Trial.
1987, Hulme et al [34]	To explore the effects of adaptive seating on oral-motor functioning	11 children 1-4 yr	An assessment instrument with a behavioural base was used for the seven direct observations	Sitting posture and head alignment during eating and drinking improved, children progressed from bottle to cup drinking and from eating blended to chopped or cut-up food.	Repeated measurements
1987, Nwaobi [40]	To explore the how sitting orientation affect upper extremity	N=13	Time in second to activate a switch	Significant difference between neutral position and all other position	Repeated measure cross sectional
1986, Colbert [33]	Investigate the effect of the custom-molded DESEMO seat for posture and function.	12 children 18m-8 yr		Feeding improved dramatically for 2 children. 3 children showed no improvement or progression of their curves. The positioning device assisted head control for 10 children but failed in 2.	Comparative, AB
1984, Seeger [39]	Testing if increasing hip flexion above 90 degrees would improve hand function in those seated	12 children	Time and accuracy	Increasing hip-flexion angle in seating and extensor spasticity appears to have no effect on hand function.	Prospective Case control study

TABLE G Constraint-Induced Movement Therapy (CIMT)

Authors, Year	Title	Participants	Oxford
2002 Willis et al.[62]	Forced use treatment of childhood hemiparesis	Hemiplegia (stroke, CM, trauma, unknown etiology)	1b
2004, Taub et al.*[59] 2002, DeLuca* [58]	Efficacy of constraint induced movement therapy for children with cerebral palsy with asymmetric motor impairment Intensive therapy with casting for children with hemiplegic cerebral palsy: A randomized controlled trial (Dissertation)	Hemiplegia (CP)	1b
2005, Eliasson et al. [111]	Effects of constraint-induced movement therapy in young children with hemiplegic cerebral palsy: an adapted model	Hemiplegia (CP)	2b
2005, Naylor & Bower [66]	Modified constraint-induced movement therapy for young children with hemiplegic cerebral palsy: a pilot study	Hemiplegia (CP)	4
2005, Sung et al [61]	Efficacy of forced use therapy in hemiplegic cerebral palsy	Hemiplegia (CP)	2b
2006, Bonnier et al ** [67] 2003, Eliasson et al ** [65]	Effects of constraint-induced movement therapy in adolescents with hemiplegic cerebral palsy: a day camp model Clinical experience of constraint induced movement therapy in adolescents with hemiplegic cerebral palsy - a day camp model	Hemiplegia (CP)	4
2006, Charles et al. [63]	Efficacy of a child-friendly form of constraint-induced movement therapy in hemiplegic cerebral palsy: a randomized control trial	Hemiplegia (CP)	1b
2006, Gordon et al. [68]	Efficacy of constraint-induced movement therapy on involved upper-extremity use in children with hemiplegic cerebral palsy is not age dependent	Hemiplegia (CP)	3b
2007, Charles et al [71]	A repeated course of constraint-induced movement therapy results in further improvement	hemiplegia	3
2007, Juenger et al [69]	Cortical neuromodulation by constraint-induced movement therapy in congenital hemiparesis	Hemiplegia due to MCA and preserved contra lateral motor projection	4
2008, Wallen et al [70]	Modified Constraint-induced therapy for children with hemiplegic cerebral palsy: A feasibility study	Hemiplegia (CP)	4
2008, Hoare et al [57]	Constraint-induced movement therapy in the treatment of the upper limb in children with hemiplegic cerebral palsy: a Cochrane systematic c review	Congenital hemiplegia	1a
* and ** the same data are reported in both articles			

TABLE H Constraint-Induced Movement Therapy (CIMT)

Authors, Year	N	Ages	Intervention	Research Design		Outcome measures	Main finding
2002, Willis et al.[62]	25 Tx=12 Cont.= 13	1–8 y	1 month, no training	RTC Pre–and 1 mo post–intervention & 6-mo follow–up	Short arm cast	Modified PDMS and parent interview	Improvement on both modified PDMS and parent report, positive result of half of the group who returned for follow up
2004, Taub et al. [59] 2002, DeLuca [58]	18 Tx=9 Cont=9	7–96 m	3 weeks, restraint during waking hours, including shaping, 6 h/d	RTC with cross over design Pre- & post–intervention & 3 mo follow-up	Bi-valved full arm cast	EBS, PMAL, TAUT	Significant improvement on all but non validated tests
2005, Eliasson et al.[111]	41 Tx. =21 Cont.=20	18 mo-4y	8 weeks, Training and restraint 2 h/d	Prospective + control Pre- & post- intervention, 6 mo follow-up	Stiff glove	AHA	Significant improved bimanual performance (AHA), sustained at follow up
2005, Naylor & Bower [66]	9	21-61 mo	4 weeks, training 2x1 h/wk and 1h/d in home program.	Case-series design ABAA Pre- & post-intervention 4-wk follow-up	Gentle physical restraint + verbal instruction	QUEST	Significant improvement, sustained at follow up
2005, Sung et al [61]	31 Tx =18 Cont =13	>8y	6 weeks, training 2x30min/w for both groups	RTC Pre- & post-intervention	short arm cast,	Box and blocks WFIM EDPA	Improvement of all assessment, although questionable, since it occurred in both hands
2006, Bonnier et al [67] 2003, Eliasson et al [65]	9	13–18 y	2 weeks, training and restraint 7 h/d, group intervention	Case-serie design Pre–and post–intervention 5-mo follow–up tests	Stiff glove	B–OS, J–T Test, AMPS, grip strength, in–hand manipulation, Novel MotorTask Performance	Significant result of B-OS, J-T test, in hand manipulation, novel Motor Task, sustained at 6-month

TABLE H Constraint-Induced Movement Therapy (CIMT) (continued)

Authors, Year	N	Ages	Intervention	Research Design		Outcome measures	Main finding
2006, Charles et al. [63]	22 Tx.=11 Cont.= 11	4-8 y	2 weeks, restraint and training 6 h/d	RTC Pre- & post-intervention. 1& 6-mo follow-up	Sling	J-T Test B-Os CFUS, Strength, tactile sensation, muscle tone	Significant improvement for J-T test, B-Os and CFUS, sustained at 6-month
2006, Gordon et al. [68]	19 Younger =11 Older=8	4-8 y 9-14 y	2 weeks, restraint and training 6 h/d	Prospective single group design	Sling	J-T Test B-Os CFUS Strength, tactile sensation, muscle tone	Significant improvement in both group for J-T test, B-Os and CFUS, no difference between groups, sustained at 6-month
2007, Charles et al [71]	8	5-11 y	2 weeks, restraint and training 6 h/d	ABABA design	sling	J-T Test B-Os CFUS	Significant improvement on all test both after first and second period of treatment, sustained at 6-month
2007, Juenger et al [69]	10	10-30 y	12 days restraint and training 10 h /day	Before and after		fMRI WMFT	All participants improved at WMFT 4 showed increased activation in primary sensori-motor cortex 3 unclear, 2 no changes
2008, Wallen et al [70]	10	6m-8y	8 weeks, restraint and training 2h /day,	Prospective pre-post feasibility study	Mit	COPM, GAS, PMAL, AHA, MUL	COPM, PMAL and GAS improvement

Abbreviations: CP=cerebral palsy, CM= cerebral malformation, Tx.= treatment, J-T Test= Jebsen-Taylor Test of Hand Function, PDMS= Peabody Developmental Motor Scales, AMPS= Assessment of Motor processing Skills, B-Os= Bruininks-Oseretsky Test of Motor Proficiency, AAUT= Actual Amount of Use Test, MAL= Motor Activity Log, PMAL= Pediatric Motor Activity Log, TAUT= Toddler Arm Use Test, EBS= Emerging Behavior Scale, AHA= Assisting Hand Assessment, QUEST= Quality of Upper Extremity Skills Test, CFUS= Caregiver Functional Use Survey. EDPA=Erhardt Developmental Prehension Assessment, fMRI = functional Magnetic Resonance Imaging, WMFT = Wolf Motor Function Test, COPM = Canadian Occupational Performance Measure, GAS= Goal Attainment Scale, MUL= Melbourne Upper Limb Test

TABLE I Feeding Interventions

Authors, year	Title	Oxford score	Feeding treatment approach
Haberfellner & Rossiwall, 1977 [89]	Appliances for treatment of oral sensori-motor disorders	4	ORAL APPLIANCE: Innsbruck Sensori-motor Activator and Regulator (ISMAR)
Banerdt & Bricker, 1978 [94]	A training program for selected self-feeding skills for the motorically impaired	4	POSITIONING/TASK-TRAINING: Proximal and trunk support, midline orientation, coupled with task training (self-feeding) and adapted equipment
Helfrich-Miller et al, 1986 [81]	Dysphagia: Its treatment in the profoundly retarded patient with cerebral palsy	4	ORAL SENSORI-MOTOR FACILITATION: Thermal stimulation, oral motor treatment and dietary modification
Ganz, 1987 [73]	Decreasing tongue thrusting and tonic bite reflex through neuromotor and sensory facilitation techniques	3b	ORAL SENSORI-MOTOR FACILITATION: desensitize oral region, inhibition of abnormal oral and postural reflexes, facilitation of muscle tone
Fischer-Brandies et al, 1987 [95]	Therapy of orofacial dysfunctions in cerebral palsy according to Castillo-Morales: First results of a new treatment concept	4	ORAL APPLIANCE: Orthodontic palatal plates (stimulates tongue and upper lip), with oral-facial physiotherapy (e.g. Vojta, Bobath) in one third
Croft, 1992 [74]	What consistency of food is best for children with cerebral palsy who cannot chew?	2b	FOOD CONSISTENCY: Soft solid versus mashed food
Morton et al, 1993 [82]	Videofluoroscopy in the assessment of feeding disorders of children with neurological problems	4	POSITIONING/CONSISTENCY AND OTHER TECHNIQUES: Recommendations for changes in trunk position (forward flexion or reclined position), as well as utensils, consistency (based on videofluoroscopy)
Larnert & Ekberg, 1995 [91]	Positioning improves the oral and pharyngeal swallowing function in children with cerebral palsy	4	POSITIONING: Extended versus flexed neck with 30 degrees reclined sitting
Gisel et al, 1995 [72]	Effect of oral sensori-motor treatment on measures of growth, eating efficiency and aspiration in the dysphagic child with cerebral palsy	2b	ORAL SENSORI-MOTOR FACILITATION: Stimuli used for tongue lateralization, lip control, chewing vigor; use of textures
Gisel et al, 1996 [83]	Oral-motor skills following sensori-motor therapy in two groups of moderately dysphagic children with cerebral palsy: Aspiration vs nonaspiration	2b	ORAL SENSORI-MOTOR FACILITATION: Stimuli used for tongue lateralization, lip control and vigor of chewing; use of textures
Pinnington & Hegarty, 1999 [80]	Effects of consistent food presentation on efficiency of eating and nutritive value of food consumed by children with severe neurological impairment	4	ADAPTED EQUIPMENT: Handy-1 Robotic Aid to Eating
Gisel et al, 1999 [77]	The Innsbruck Sensori-motor Activator and Regulator (ISMAR): Construction of an intraoral appliance to facilitate ingestive functions	4	ORAL APPLIANCE: Innsbruck Sensori-motor Activator and Regulator (ISMAR)
Gisel et al, 2000 [76]	"Whole body" mobility after one year of intraoral appliance therapy in children with cerebral palsy and moderate eating impairment	2b	ORAL APPLIANCE: Innsbruck Sensori-motor Activator and Regulator (ISMAR)
Gisel et al, 2001 [78]	Impact of oral appliance therapy: Are oral skills and growth maintained one year after termination of therapy?	2b	ORAL APPLIANCE: Innsbruck Sensori-motor Activator and Regulator (ISMAR)
Pinnington & Hegarty, 2000 [79]	Effects of consistent food presentation on oral-motor skill acquisition in children with severe neurological impairment	3b	ADAPTED EQUIPMENT: Handy-1Robotic Aid to Eating
Gerek & Ciyiltepe, 2005 [112]	Dysphagia management of pediatric patients with cerebral palsy	4	ORAL APPLIANCE: Castillo-Morales Device (CMD) coupled with speech language therapy and oral-motor range of motion exercises
Clawson et al, 2007 [90]	Use of behavioral interventions and parent education to address feeding difficulties in young children with spastic diplegic cerebral palsy	4	ORAL SENSORI-MOTOR FACILITATION: oral-motor exercises (range, strength) followed by feeding; coupled with positioning and behavioral techniques (positive reinforcement, extinction of negative behaviors); parental involvement

TABLE J Feeding Interventions

Authors, year	Subjects; sample size; age range	Methodology	Outcome measures / Variables*	Main findings
I. ORAL SENSORI-MOTOR FACILITATION				
Helfich-Miller et al, 1986 [81]	Severe CP (spastic quadriplegia, cerebral encephalopathy, microcephaly); N=6; 10-31 years	Case series, intermittent repeated measures; 3x/day but thermal increased to 6x/day, everyday for 12 months	Modified barium swallow, ENT exam, in depth oral-motor examination, videotaping of feeding	Improvements made in efficiency and speed (especially pharyngeal transit times) of swallowing. Important reductions in aspirations. Decreased incidence of upper respiratory tract infections for those who remained on thermal stimulation program. *Percentage change over time.*
Ganz, 1987 [73]	Severe cerebral palsy; N=1; 8 years old	Single-subject ABA design; daily/14 weeks at meal time followed by 4 weeks without treatment	Morris Pre-Speech Assessment Scale (oral-motor function), number of tongue thrusts and tonic bites between 6th -15th presentation of food during a meal	Decreased tongue thrust with both solid and semi-solid foods. Other positive changes included: merging tongue lateralization, stable jaw movements with the cup, decreased tongue protrusion with liquids, better suck-swallow-breathing coordination, decreased associated movements such as head extension, decreased stereotyped phasic bite and release pattern in chewing, beginning to chew with lips closed. However, poorer elevation of tongue with jaw separation for liquids and solids. *Results were tested statistically.*
Gisel et al, 1995 [72]	Moderate to severe cerebral palsy with moderate eating impairment n=27 ; mean age= 5.1 (2-10 years)	Experimental; Group 1: 20 weeks treatment, Group 2 & 3: 10 weeks control then 10 weeks treatment; (group 1/2: non-aspirators, group 3: history of aspiration); 5-7 minutes/day,5x week before lunch/snack	Modified Functional Feeding Assessment-subtest of the Multidisciplinary Feeding Profile, Gisel Video Assessment, Videofluoroscopy	Eating efficiency in non-aspirators did not change markedly in response to oral sensori-motor therapy. In the aspirating group, eating efficiency improved with puree, but decreased with solids. Children maintained their percentile rank in weight- and skinfold-for-age measurements. Findings suggest that eating efficiency is not a good estimator of treatment outcome but rather related to severity of eating impairment. *Results were tested statistically.*
Gisel et al, 1996 [83]	Cerebral palsy (moderate to severe motor impairment) n=27; 2.5-10 years	Experimental; 10 control followed by 10 weeks treatment; 5-7 minutes/day, 5 days/week, prior to lunch or snack; (group 1 : 20 non-aspirators, group 2 : 7 aspirators)	Weight and skinfold measures, Modified version of the Functional Feeding Assessment subtest of the Multidisciplinary Feeding Profile, Gisel Video Assessment, Videofluoroscopy	There were significant improvements in spoon feeding (lip control and swallow pattern), chewing (lateralization of the tongue), and swallowing. Children that aspirated performed significantly worse (baseline) than those that did not aspirate. There were no significant changes in rotary chewing or drinking skills. Children maintained their pretreatment weight-age percentile but did not show any catch-up growth. *Results were tested statistically.*
Clawson et al, 2007 [90]	Spastic diplegia, half required nutritional supplementation; N=8; 18 months-4.7 years	Quasi-experimental with pre/post testing; Primarily descriptive analyses; 4x/day; Average duration: 5.8 weeks, with follow-up for 1 year	Beckman Oral-motor Assessment, Anthropometric measures	More efficient (shorter time) chewing and swallowing, faster transit time from intake to swallow. Greater caloric consumption, less need for supplementation. Greater height and weight gain (percentile ranks increased). Caregivers were more effective in administering feeding, (after behavior program: instruction, prompts, consequences). *Descriptive analysis.*
II. CONSISTENCY				
Croft, 1992 [74]	Cerebral palsy (all types); N=67 CP, 64 controls; 3-18 years	Experimental; 2 sessions over 6 week period; Group 1:normal to near-normal speech, Group 2: poor speech; Group 3: no speech	Video observation, time to complete feeding portion	Children who had no speech took significantly longer to eat non-mashed than mashed potato; Children with CP were more likely to cough or choke while eating more solid foods, especially those in Group 3. Study suggests that mashed foods are safer and eaten more quickly than soft solid foods in children with poor oral motor skills.
III. POSITIONING				
Banerdt & Bricker, 1978 [94]	Cerebral palsy' N=1; 2.5 years	Case study; Training sessions 3 days/week, 4 months, at morning and afternoon snack time	Observation, Recording of independence, cueing, prompting, mandate (assistance)	Number of independent responses from the child increased throughout treatment. New self-feeding behaviors were maintained with minimal assistance 5 months after termination of treatment. *Results were tested statistically.*

TABLE J Feeding Interventions (continued)

Authors, year	Subjects; sample size; age range	Methodology	Outcome measures / Variables*	Main findings
Morton et al, 1993 [82]	Most had cerebral palsy, all were malnourished; N=14; 4-16 years	Case series; Observations on videofluoroscopy	Videofluoroscopy with multidisciplinary review	Children with difficulties mainly in the oral phase fed best in the reclined position, those with difficulties mainly in the pharyngeal phase fed best in the erect position, particularly if they had upper-esophageal sphincter spasm with tonic labyrinthine reflex. Seating and positioning recommendations were made for feeding which parents found to be helpful, however they reported no changes in terms of weight gain. *Descriptive analysis.*
Larnert & Ekberg, 1995 [91]	Tetraplegia with dystonia (aspiration, recurrent pneumonia); N=5; 3-10 years	Quasi-experimental; Videofluoroscopy observations when sitting upright compared to 30 degrees reclined and neck supported and flexed; Single session	Videofluoroscopy	In the reclined position with neck flexed, aspiration decreased in all subjects, oral leak diminished in 2 children, retention of puree improved in one child. *Descriptive analysis.*
IV. ORAL APPLIANCE				
Haberfellner & Rossiwall, 1977 [89]	Mixed forms of spasticity, athetosis and/or ataxia N=9; 6-12 years	Quasi experimental, pre-post testing with descriptive analyses only; Progressively increased use of appliance; 18 months	Clinical observations: oral sensibility to touch, oral stereognosis, phonation and articulation, position of tongue tip during swallows, mastication, deglutation, speech, lip seal, drooling, nasal breathing	Normalization of sensibility, lip seal, transport of saliva, nasal breathing. Improvement of speech articulation. *Descriptive analysis.*
Fischer-Brandies et al, 1987 [95]	Cerebral palsy; N=71; l 4-14 years	Quasi experimental, pre-post testing with descriptive analyses only; Several hours/day except when eating or sleeping; average duration:15 months (0.5-3.0 years)	Oral sensory-motor functions via formal neuropediatric examination	Improvements noted in spontaneous tongue position and coordination of tongue movement, food intake, speech development and drooling (in at least half of the children). In 5 children, treatment was discontinued due to no apparent improvements. Spontaneous position of lips and the constant open mouth remained unchanged in most cases. The positive effects of physiotherapy versus the appliance could not be differentiated in this study. *Descriptive analysis.*
Gisel et al, 1999 [77]	Moderate spastic quadriplegia; N=1; 12 years, 10 months	Case study; 6 months control, 12 months with appliance (stabilization, then mobilization phases)	Anthropometric measures (weight, skinfolds), Video observation during feed, Functional Feeding Assessment	Change in facial expression (jaw more retruded in resting position; more relaxed facial musculature). Improved functional feeding skills (spoon feeding, biting, chewing, cup drinking, straw drinking) and visible aspects of the swallow. Improved eating efficiency of 3 standard textures, with catch-up of weight during treatment phase. Able to self feed. . Concurrent improvements in ambulation and upper extremity functions (possibly related to improved nutritional state and/or better postural control) *Descriptive analysis.*
Gisel et al, 2000 [76]	Tetraparesis with moderate motor impairment; N=20; 4-13 years	Randomized controlled trial, Group A: 12 months ISMAR, Group B: 6 months standard rehab-control, followed by ISMAR for 12 months; worn daily; Randomization method not specified.	Formal video observation: postural and ambulatory skill measurements, Sitting Assessment Scale, Pathologic reflexes and movements, Video facial expression analysis	Significant improvement in sitting postural control (axial skeleton) and upper extremity control after one year of ISMAR therapy. Ambulatory status in children who relied on wheelchair mobility also significantly improved, above the level of maturation. These changes were not observed in the control period. These changes were associated with improvements in jaw stabilization and oral-motor control. Half of the children showed marked improvement in oral posture (e.g. lip closure, closed mouth) but not tongue position. *Results were tested statistically.*

TABLE J Feeding Interventions (continued)

Authors, year	Subjects; sample size; age range	Methodology	Outcome measures / Variables*	Main findings
Gisel et al, 2001 [78]	Tetraparesis with moderate motor impairment; N=17; 6-15 years	Experimental, Group A: 2 years with ISMAR (n=9), Group B: 1 year with ISMAR, 1 year without (n=8). (note: sample from Gisel 2000 study, however some children were non-randomly reassigned to different groups	Anthropometric measurements, Functional Feeding Assessment	Prolonged use of ISMAR was tolerated without complications. No significant differences were found in the 7 domains of functional feeding or in weight gain on follow-up (18-24 months). Maturation was equally effective as ISMAR therapy after the first year of use. *Results were tested statistically.*
Gerek & Ciyiltepe, 2005 [112]	Cerebral palsy; N=7, 8-17 years	Case series (descriptive), 6-8 hours/day with rest periods to all day by week 4; for 12 weeks; With Vojta and oral-motor range of motion exercises	Deglutition skills, Lip and tongue position, Goniometric measures of labial movements, Phonation time	Improvements noted in bilapial lip control and lip closure. Most obvious changes noted in degluition skills (higher consistency in food intake, decreased risk of aspiration, better saliva control). Parents expressed satisfaction with the results of therapy. *Descriptive analysis.*
V. ADAPTED EQUIPMENT				
Pinnington & Hegarty, 1999 [80]	Most (17) had cerebral palsy, 3 traumatic brain injury, Lesch-Nyhan syndrome or encephalitis; all dependent feeders, wheelchair users; N=20; 7-17 years; but 4 withdrew - could not use device	Quasi-experimental (AB within-subject design); Use of electric feeder (consistent method), when feasible for selected meals compared to delivered by hand (standard method); 9 months	Dietary analysis program (for energy and protein content), Diary recordings over 5 days for intake and eating efficiency, Duration of meals and rate compared to quantity of food eaten, Anthropometric measurements	Food intake and weight gain can be maintained using assistive device compared to feeding by a caregiver. No change in the amount of energy or protein consumed. Efficiency of eating is reduced (slower) with use of the assistive device, particularly for children with some speech who performed more poorly with the device when compared to children with no speech. *Results were tested statistically.*
Pinnington & Hegarty, 2000 [79]	Most cerebral palsy, 3 other diagnoses (above); N=16; 7-17 years	Quasi-experimental (ABA within-subject design); baseline, intervention and postintervention phases- 3 months each; selected meals; 9 months	Video recording/ analysis, Schedule for Oral-motor Asessment (oral-motor behavior and postural control during eating)	Statistically significant improvements in components of oral-motor behaviour (e.g. mastication, swallowing) were found when a consistent method of food presentation was employed. Improvements were not always retained on follow-up in the post-intervention phase. Children with more limited speech skills appeared to benefit more. *Results were tested statistically.*

* Outcome measures/Variables: when studies specified actual measures used, these are indicated in the column, otherwise the variables observed and documented are specified.

TABLE K Virtual Reality

Authors, year	Title	Oxford score	VR type
Reid, 2002 [107]	Benefits of a virtual play rehabilitation environment for children with cerebral palsy on perceptions of self-efficacy: A pilot study	4	Virtual reality (Mandala Gesture Xtreme VX technology, 1996) - Games* to promote trunk midline and lateral movement and control
Reid, 2002 [105]	The use of virtual reality to improve upper-extremity efficiency skills in children with cerebral palsy: a pilot study	4	Virtual reality (Mandala Gesture Xtreme VX technology, 1996) - Games* to encourage arm reaching
Akhutina et al, 2003 [101]	Improving spatial functioning in children with cerebral palsy using computerized and traditional game tasks *Experiment 1*	2b	Virtual environments (Super Scape VRT 3-D construction package) - Desk-top activities and training tasks (e.g. moving through a maze)
	Improving spatial functioning in children with cerebral palsy using computerized and traditional game tasks *Experiment 2*		- Games (e.g. sorting, matching, naming cards; figure construction)
Reid, 2004 [106]	The influence of virtual reality on playfulness in children with cerebral palsy: a pilot study	4	Virtual reality (Mandala Gesture Xtreme VX technology, 1996) - Games*
Harris & Reid, 2005 [109]	The influence of virtual reality play on children's motivation	4	Virtual reality (Mandala Gesture Xtreme VX technology, 1996) - Games* to encourage motivation and engagement in play activities
You et al, 2005 [108]	Cortical reorganization induced by virtual reality therapy in a child with hemiparetic cerebral palsy	4	Virtual reality (IREX VR therapy system) - Games involving manipulation of objects, to encourage movement
Reid & Campbell, 2006 [104]	The use of virtual reality with children with cerebral palsy: A pilot randomized trial	2b	Virtual reality (Mandala Gesture Xtreme VX technology, 1996) - Games* requiring arm reaching
Chen et al, 2007 [102]	Use of virtual reality to improve upper-extremity control in children with cerebral palsy: A single-subject design	4	2 VR systems: VR-based hand rehabilitation training system and EyeToy-Play system - Target and tracking games (e.g.butterfly, pegboard, bubbles, boxing, kungfoo) to improve goal-directed and anticipatory reaching

*examples of games: volleyball, drumming, soccer, snowboarding, juggling, painting, bubbles

TABLE L Virtual Reality

Authors, year	Subjects; sample size; age range	Methodology	Outcome measures / Variables*	Main findings
Reid, 2002a [107]	Spastic quadriplegia or diplegia; N=3; 8-12 years	Case study, pre/post test design two 90 minute sessions per week, 4 weeks	Canadian Occupational Performance Measure (COPM) -mainly self-care and leisure activities were identified	Performance and satisfaction scores were rated higher at post-test for all participants (clinically significant). Suggests beneficial effects with respect to self-efficacy. *Descriptive analysis.*
Reid, 2002b [105]	Spastic quadriplegia or diplegia; N=4; 8-12 years	Case study, pre/post test design; 1.5 hrs/week, 8 weeks	Quality of Upper Extremity Skills Test (QUEST), BOTMP (item 6 of subtest 5- motor accuracy)	Scores improved on the BOTMP accuracy score. Change in quality of upper extremity movements was variable (no consistent effect). *Descriptive analysis.*
Akhutina et al, 2003 [101]	Cerebral palsy; N=21; 7-14 years; 12 experimental, 9 control	Experimental design; 30-60 min/session, 6-8x over a one month period	Computer-based: Koos Block Design Test, Clown Assembly Test Other tests: Decentration of Viewpoint Test, Directional Pointing to Hidden Object Test, Raven Progressive Matrices, Benton Judgment of Line Orientation Test, Arrows subtest of the Nepsy-Neuropsychological Test Battery for Children, Roads Test	No significant changes were observed (Most children in experimental group were non-ambulatory whereas most controls were ambulatory) *Results were tested statistically.*
	Cerebral palsy (most diplegia); N=45; age not specified; 23 experimental, 22 control	Experimental design; 30-60 min/session, 6-8x over a one month period		Visual-spatial abilities of the children in the treatment group improved more compared to the control group *Results were tested statistically.*
Reid, 2004 [106]	Cerebral palsy N=13; 8-12 years	Observational study (videotape analysis) 8 1-hour sessions	Test of Playfulness	Children exhibited all elements of playfulness (intrinsic motivation, internal control, suspension of reality, and framing) while playing the VR games. Playfulness varied with the type of virtual environment, some being more engaging than others. *Descriptive and qualitative analyses.*
Harris & Reid, 2005 [109]	Cerebral palsy; N=16; 8-12 years	Observational study (videotape analysis) 1 hr/week, 8 weeks	Pediatric Volitional Questionnaire (measures motivation, level of engagement)	VR play is a motivating activity, therefore of interest as an intervention tool. Level of volition was influenced by the type of virtual environment (e.g. degree of variation in the game, degree of challenge, competition.
You et al, 2005 [108]	Hemiplegia; N=1; 8 years	Case study; 60 min/day, 5x/week for 4 weeks	fMRI, BOTMP, modified Pediatric Motor Activity Log Questionnaire, Upper Limb subtest of the Fugl-Meyer assessment	Improved functional motor skills (use) and better quality of movement (control, coordination) in affected upper limb. Post intervention, spontaneous reaching, self-feeding and dressing were noted (not possible prior to VR). Concurrently, there was greater activation of the contralateral sensori-motor cortex (as opposed to ipsilateral or bilateral). *Descriptive analysis.*
Reid & Campbell, 2006 [104]	Cerebral palsy; N=31; 8-12 years 19 experimental, 12 control (standard care)	Randomized controlled trial; 1.5 hours/week, 8 weeks	Canadian Occupational Performance Measure (COPM), Quality of Upper Extremity Skills Test (QUEST), Harter Self-Perception Profile for Children (SPPC)	Significantly increased scores for the social acceptance subscale favoring the experimental group. VR appears to increase motivation in experimental group. Many children in the control group were lost to follow-up. *Descriptive and statistical analyses.*
Chen et al, 2007 [102]	Spastic cerebral palsy; N=4; 4-8 years	Single-subject design, AB with 2-4 week follow-up; 2 hrs/week, 4 weeks	Reaching kinematics, Fine Motor Domain of PDMS-2 (subtests: grasping, visual-motor integration)	3/4 children showed improvement in some aspects of quality of reaching which was partially maintained 4 weeks after treatment. Scores on the PDMS-2 increased (1-11 points) for all children; scores on the visual-motor integration subtest improved for 3/4 subjects. *Statistical analyses (single subject design).*

* Outcome measures/Variables: when studies specified actual measures used, these are indicated in the column, otherwise the variables observed and documented are specified.

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OCCUPATIONAL THERAPY MANAGEMENT: POINTS OF DISCUSSION

Marjolijn Ketelaar, PhD
Senior researcher
Rehabilitation Center De Hoogstraat, the Netherlands

The reviewers did a marvellous job in reviewing the whole range of occupational therapy papers in a very systematic way, using the PICO system to formulate the research questions. An important aspect to focus on in the discussion is on the outcomes. All research questions have been formulated in terms of different outcomes, using ICF terminology. For example, for Virtual reality the PICO question was formulated (p17): Does weekly repetitive training using a play-based VR intervention improve: (1) brain reorganization, (2) motor skills, (3) spatial skills, (4) motivation and playfulness, (5) functional everyday self-care and (6) leisure activities in children with CP? This means that for virtual reality, which is just an example, but illustrative for the other topics, in fact there are 6 research questions, all focusing on another outcome.

I think it is important to distinguish these outcomes explicitly in the conclusions and recommendations. For example looking at the different papers and the different outcomes, it was found that in one study with an Oxford level of 4 there were significant improvements on the performance and satisfaction levels of the COPM. There was another study, also with an Oxford level of 4, which found no change in quality of movement, and you found a third study, Oxford level 2b, in which it was found that social acceptance improved of the children training with virtual reality games compared to a control group.

I think it makes sense to differentiate between outcomes when concluding on the levels of evidence. From this respect I think it is fair to conclude that there is no evidence that virtual reality may improve quality of movement (grade of recommendation C), and that there is some evidence that it may improve social acceptance (grade of recommendation B).

The second topic I think is important to discuss relates to the steps in leading to the grades of recommendation. When looking at these tables I felt the Oxford level was not very easy to use. And in the PT chapter I recognized the same struggle. When do you give a paper a level of I, IIa, IIb, or IV? IN the PT chapter, two reviewers described a paper on an RCT I am very familiar with, and the first reviewer gave it an Oxford level of IIb, and the other one gave it an Oxford level of I. When I looked at the tables fr the OT studies I recognized the same struggle; for example looking at the papers on splinting there was a paper with a quasi-experimental design of McPherson and colleagues, described as Oxford level 4. Another paper with a quasi-experimental design (Kerem and colleagues) was assigned a level of 2b, and an RCT of Exner & Bonder received a level of 4.

Why do I think this is important? Well, at the end of the conference we want to summarize the recommendations, and base them on the Grades of Recommendation A-D, which in turn are based on the Oxford levels. But are we really sure about the levels of evidence? So, to put it another way, from a research perspective I would be interested to know how another reviewer would rate the same papers and what is the reliability of this classification system.

The third topic I would like to emphasize is the topic of subgroups. This came up yesterday as well. The GMFCS and the MACS are very helpful classification systems to describe the children in a study. If a study for example included children with MACS levels I and II, it should be explicitly described in the Recommendations that follow in the guidelines. For splinting and casting some first indications were presented that it might be useful, especially for specific subgroups, based on severity and age. From the review it became clear that severity was rarely mentioned. I see it as a major challenge for the coming years to distinguish subgroups for different interventions.

This brings me to my last point which has to do with working mechanisms. When I look at the papers that have been reviewed I see two different ways of thinking. First, interventions focusing on improving body function or body structure with the implicit assumption that improvements in these areas will lead to improvements of activities and participation. And second, interventions that focus directly at optimising activities and participation. Right now there is some evidence that changes at the level of body function do not relate very strongly to changes at the level of activities.

This means it becomes very important to become explicit in our thinking in terms of working mechanisms and to think about our major outcomes (again). Yesterday we discussed the statement if social inclusion/participation should be the overarching goal for health care. If we agree with that it has important consequences for the outcomes we should study, and even more importantly to make clear what is the theoretical perspective, what's the theoretical pathway or mechanism to reach that goal?

MEDICAL MANAGEMENT

Jules Becher MD PhD
Professor of Paediatric Rehabilitation Medicine
VU University Medical Center
&
Deb Gaebler-Spira MD
Professor, Departments of Physical Medicine and Rehabilitation & Pediatrics
Rehabilitation Institute of Chicago & Northwestern Feinberg School of Medicine

The definition of cerebral palsy has been expanded to incorporate aspects of the condition that affect activities or functional abilities (1). This definition is in keeping with the World Health Organization's concepts of participation, abilities, and body structure and function (2). Optimizing the medical condition of the child with cerebral palsy aims to improve body structures and function and increase participation. Children with cerebral palsy represent a heterogeneous group of children with varying medical co-morbidities and secondary musculoskeletal impairments. Medical co-morbidities represent impairments of body structure and function. The use of the Gross Motor Functional Classification System (3) allows a reliable way of classifying and predicting gross motor function and provides a useful stratification to base realistic habilitation and rehabilitation goals on. The GMFCS represents a continuum of the severity of motor disorder. Current literature shows that medical conditions have a significant relationship with the child's GMFCS level.This paper will review interventions based on the use of the GMFCS classification.

The children in GMFCS I, II, and III are anticipated to walk with different levels of competencies. The goals for this group include i) the development of basic and higher gross motor skills such as standing, walking, running and jumping, ii) improvement of the efficiency of gait and iii) maintenance and prevention of deterioration in gait. More concrete treatment goals are decreasing gait deviation by improving alignment, improving range of motion, improving strength, and decreasing the related issues that may interfere with walking.

The children in GMFCS classes IV and V will have limited abilities to move around at home, at school and in the community. Mobility and self-care goals are for sitting and comfortable positioning, transfers, dressing, changing diapers and – in general - maximizing the interaction with the environment. Improving general health status and decreasing medical complications have a high priority in this group. For all children, doing their best depends on the child having a base of rest, nutrition, and medical well-being. As about 85% of the children has a spastic motor disorder, this paper is focused on the treatment of spastic Cerebral Palsy (4).

The first part of this paper will provide focused systematic reviews highlighting areas that affect medical conditions that affect life expectancy, nutrition, and bone health will be reviewed for children in GMFCS classes IV and V, as well spasmolytic therapy as oral medications and Intra-Thecal Baclofen therapy (ITB) to improve care and comfort. Although most interventions lack randomized control studies of adequate size, sufficient cross-sectional information exists to define problems that are in need of trials.

THE NON-AMBULANT CHILD (GMFCS level IV and V)

This chapter considers those children who are non-ambulatory (Level IV, ability function in supported sitting; Level V, severe limitations in head and trunk control with need for extensive assistance for care and mobility). Rehabilitation goals are directed toward sitting and comfortable positioning, transfers, and maximizing interaction with the environment. In addition to functional impairments, children with GMFCS Levels IV and V are also likely to have comorbid medical problems. Preventing and/or treating medical complications and improving overall health status are a high priority in this group. For all children, optimal function depends on a base of rest, nutrition, and medical well-being.
Of the many medical issues that affect children and families, we consider three that are of major importance to families and also have implications for orthotic practice: life expectancy, nutrition, and bone health. We also review two commonly used treatments for improvement of care and comfort: intrathecal baclofen (ITB) and oral spasmoytic medication. Treatments on single muscle level are not reviewed (e.g. adductor spasticity with botulinumtoxin A). Although most interventions lack large randomized control studies, sufficient cross-sectional information exists to define problems that are in need of trials.

METHODS

The following databases were searched for randomized clinical trials (RCTs) and systematic reviews: Medline, the Cochrane Library, the Cumulative Index to Nursing and Allied Health Literature (CINAHL), the Physiotherapy Evidence Database (PEDro), the Center for International Rehabilitation Research and Information and Exchange (CIRRIE), and the National Rehabilitation Information Center (NARIC). If RCTs and systematic reviews were not available, additional references were reviewed. The Oxford Center level of evidence-based medicine was used to grade the evidence of each study.
The literature was analyzed focusing on a PICO question (Patient population, Intervention, Comparison, Outcome). The authors used the information to discuss the implications for orthotics.

What factors are important for limited life expectancy in children with cerebral palsy?

Life expectancy is the average survival time of a population; it is convenient measure of the health of a population but it cannot predict the lifespan of an individual. It is a way of looking at trends in general health, and it is important in planning health services and systems of care such as school, rehabilitative services, and social support services.
Population-based life expectancy is calculated using statistical tools that incorporate complex statisticsl using complex statistical relationships. Large longitudinal databases, such as those in the U.K. and Australia, have demonstrated consistent findings relating the number and severity of functional disabilities to survival.[2,3] Survival curves were calculated and are available (Figure 1).[2,3]

Life expectancy is negatively related with severe motor involvement, cognitive impairment and visual impairment (Figure 1).

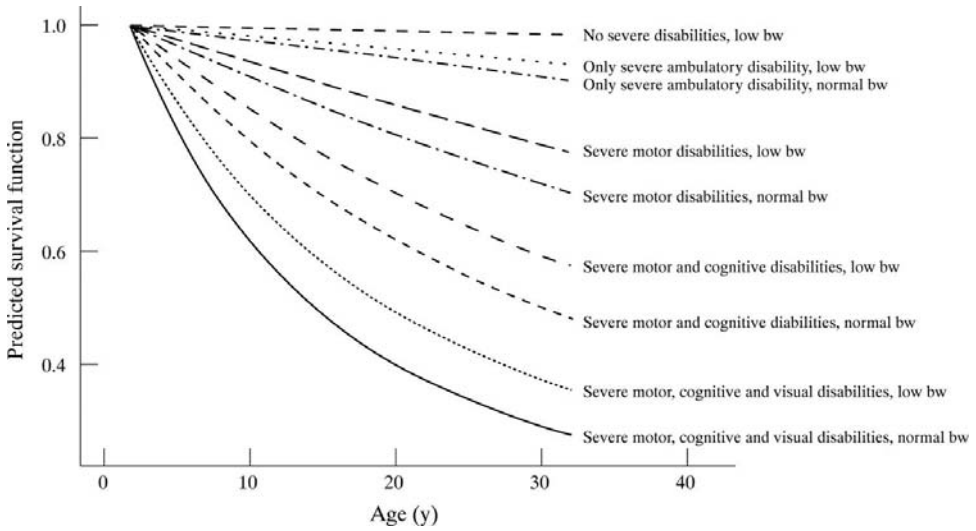

Figure 1 Predicted survival from the age of 2 years
Normal bw, >2500 grams, low bw, <2500 grams.
(from: Hutton JL, Pharoah PO. Effects of cognitive, motor, and sensory disabilities on survival in cerebral palsy. Arch Dis Child 2002;86:84-9.)

In a study in the U. S., pooled repeated observations were analyzed with logistic regression for 28,513 children (4 to 14 years) and 30,185 adults, yielding 407,895 person-years of follow-up. Life expectancy is limited for persons without head control (between 10-20 yrs), and better for persons fed by others than tube fed. However, they found a steady decline in mortality in severely affected children and in those fed with gastrostomy tubes over the previous 20 years. [4]
Strauss also reviewed the databases from the U.K. and Australia and concluded that the findings were quite consistent with each other and with the findings in the U.S. study.[5] The authors of these studies of cerebral palsy and life expectancy stress the need for consistency in definitions of disabilities and in methods for calculating life expectancy. They also emphasize the importance of stratifying data to calculate life expectancy for those least severely affected by cerebral palsy, the need to take secular trends into account when comparing life expectancy between countries
Countries that have instituted registries are beginning to provide longitudinal data that informs our ability to estimate survival.
Severity of impairment (motor, cognitive and visual) is associated with decreased life expectancy. Studies have consistently shown that the association of motor disability is greater than the effect of cognitive disability in this regard.
Recommendation: Orthotist should use the GMFCS classification to classify a child's ability to move around and being aware of a limited life expectancy. Orthotists can also assist in improving health status by monitoring weight and height .
Implications for orthotics: The overall life expectancy of children with CP is – in general – very good, but is at risk in those children with severe impairments (motor, cognition and visual). More than 90% of all children with CP become adults with CP. Interventions of the orthotist should always be put into this life-span perspective.

Does gastrostomy improve body structure and function in children with cerebral palsy?

A major concern for children with severe cerebral palsy is adequate nutrition and growth.[6] The ability to sustain weight and growth is essential to further development.[7] Factors leading to lengthy feeding and inadequate caloric intake include poor coordination of suck and swallow, poor GI motility, gastroesophageal reflux, and difficulty positioning for feeding.[8] Inadequate caloric intake then leads to poor growth and poor nutritional status.

A search was performed for systematic reviews of gastrostomy feeding in children with Level IV and V cerebral palsy performed since 2004. There have been no randomized controlled trials; but there have been eight studies showing that children fed by gastrostomy tubes show greater gains in weight, height, and skinfold thickness than children without gastrostomy tubes. The levels of evidence in these studies range from 2c to 3b. Other studies, however, with evidence levels of 2b and 2c, provide multicenter cross-sectional and prospective longitudinal cohort data. These studies consistently demonstrated improved growth and increased fat mass index (see tables). In addition, one study with 2c evidence with small numbers showed that best growth was associated with fewest days of health care use and more social participation.[8] Although it now appears that gastrostomy tubes can lead to increased growth, it is not clear how much weight is appropriate. Low energy expenditure may result in a high fat content, raising the possibility of relative overfeeding and excessive weight gain, causing difficulty for parents in transferring and positioning their child.[9,10,11] The level of evidence remains poor quality for impact of growth and size on ability for caregivers to continue to care for the child; however, the ethical issues in performing a large-scale well-designed study with such a vulnerable population will require expert input and human subjects review scrutiny.
Recommendation: Optimal weight is unclear but children can grow with attention to adequate nutritional intake.
Implications for orthotics: Orthotic fit and function are affected by weight; research for tolerance of orthoses related to change in growth is necessary. Awareness of swallowing difficulties and knowing the method of feeding may help the orthotist anticipate the frequency of need for modification and refabrication.

Does bone health affect body structure and function?

Bone health—structure, composition, density, and strength is a source of concern in children with cerebral palsy. There is good evidence that children with cerebral palsy are at risk of fractures, particularly children who are older and non-weight bearing. Several factors that may contribute to risk have been suggested, including severity of disease, bone mineral density, nutritional status, and use of anticonvulsants.
A systematic search of electronic databases was performed. No systematic reviews were found. Most relevant studies had evidence levels ranging from 2c to 3b.
Eight studies of fractures in children with cerebral palsy were identified. These studies were generally case control studies and demonstrated consistent findings: the risk of fracture increases in older children, those with greater disease severity, and non–weight bearing status. There was, however, one cross-sectional multicenter longitudinal study by Stevenson, who concluded that disease severity was not an independent risk factor for fracture.[12]

Bone mineral density (BMD) has been studied in nine recent articles. Study results showed some inconsistencies, but the most persuasive information was provided by Henderson in a prospective longitudinal observational study, showing that BMD in

the distal femur decreased with greater severity of disease, and increasing age.[13] There have not been consistent findings about the role of anticonvulsants or about the use of serum markers, such as parathyroid hormone, alkaline phosphatase, and inorganic phosphorus as predictors of decreased BMD or increased risk of fracture.
The scientific evidence at present supports the conclusion that bone health is affected by weight bearing status, poor nutrition, and severity of disease. Evidence also supports the notion that fractures are partially a failure of bone health. There is less evidence regarding nutritional status and anticonvulsant therapy and little evidence in support of serum factors as predictors of risk. The relationship of BMD and fractures remains to be determined. Many studies are limited by their retrospective nature, small sample size, and use of bone mineral density (BMD) instead of fracture rate as outcome. It is difficult to predict which patients with cerebral palsy will benefit from treatment (specifically bisphosphonates, and Vitamine D), increased weight bearing, and improved nutrition.
Recommendations for treatment are not yet available because of the absence of firm evidence; larger, appropriately designed studies are needed. Weight bearing seems to be an important factor and could be encouraged in non-ambulatory children with CP.
Implications for orthotics: Since weight bearing stimulates BMD in healthy children, orthoses may enhance weight bearing in non-ambulatory children with CP and – by this – may be beneficial to their bone quality.
Research is needed in collaboration with orthotists regarding proper alignment and the ability of equipment and orthoses to provide sufficient weight bearing (standing or supported walking) for bone health.

Do oral medications improve body structure and function for children with cerebral palsy?

Oral medications are frequently used in children with cerebral palsy, although there have been few large randomized studies to evaluate this practice. A systematic review of oral antispastic drugs published in Neurology in 2004 found twelve studies—six on stroke, three on spinal cord injury, and three on cerebral palsy.[14] Tizanidine was assessed in four studies, Dantrium in four studies, baclofen in three, diazepam in two, and gabapentin in one. Most studies were small, of short duration, and had no consistent efficacy variables.

Review of published articles since 2004 on diazepam, Dantrium, and baclofen show few RCTs. One RCT involved administration of a single bedtime dose of diazepam to 114 children with cerebral palsy. This study demonstrated subjective improvement in parent report of well-being and burden of care for children receiving treatment.[15] Another study involved use of two different dose regimens of diazepam in 174 children; the two-dose regimen had poorly defined outcomes but showed dose-related improvements in sleep and decreased resistance to movement, not defined as spasticity.[16] Another RCT was performed using oral baclofen with 15 children; medication was more effective than placebo in tasks such as transfers and positioning, although there were inconsistent results in other assessment tools.[17] As with other areas of study discussed in this paper, consensus is difficult to establish because of the variety of methodologies, outcomes, and assessment tools used.

Studies support the use of diazepam for reducing tone, improving nighttime discomfort, and improving motor activity. Other medications have conflicting outcomes for function, but muscle tone is decreased when using baclofen, Dantrium,

and tizanidine. Subjective improvement in comfort and well-being may be at least as important as other outcomes.
Recommendation: The need to decrease spasticity and improve comfort and well-being is an appropriate indication for oral medications. Oral anti-spasticity medications lend themselves to use in RCTs. Such studies are warranted and should involve larger numbers and better outcome tools than those in previous studies. Outcome studies should include orthotic tolerance.
Implications for orthotics: The use of oral medications is relevant to orthotists, since reducing tone can affect a defined positioning and wearing goal. Collaborative research efforts with orthotists are warranted to evaluate the role of orthotic tolerance in the management of spasticity.

Does intrathecal baclofen improve body structure and function in children with cerebral palsy?

The use of intrathecal baclofen (ITB) for reducing spasticity associated with cerebral palsy has grown over the past decade. Baclofen is most frequently administered as a continuous infusion through a programmable delivery system. In a recent article, 29 studies published between 2000 and 2007 were systematically reviewed.[18] Almost all studies were case series, case control studies, cohort studies, or case studies; evidence levels were low, and assessment tools were inconsistent. Nevertheless, considerable evidence supports the use of ITB for reducing hypertonia. This affects orthoses and positioning for children with GMCFS Levels IV and V. Some studies suggest subjective improvement in functional mobility and caregiver burden, but these results are not based on statistically significant data. Few studies have attempted to evaluate activities, and almost none have looked at participation.
The complications of CITB are most often related to surgical procedure for implantation of the drug delivery system or the function of the drug delivery system. Complications are frequent and demand expert and immediate care.[19]
Considerable evidence supports the use of ITB in reducing spasticity, although its effect on activities , participation, and quality of life has not been established.
Recommendation: Spasticity management should include ITB if hypertonia is severe and warrants aggressive management. Children using ITB should be followed closely for complications by an expert team. Functional gains and changes in quality of life and caregiver burden should be measured in controlled studies with consistent definitions and assessment tools.
Implications for orthotics: Modifying severe spasticity will affect orthotic tolerance and application and may potentially affect patterns of deformity. Orthotic management will be altered by ITB, requiring close observation and follow-up.

CONCLUSIONS FOR THE NON-AMBULANT CHILD

Developing a treatment plan for a child with GMCFS Level IV or V is complicated, since this group of children presents complex and variable medical comorbidities. Using evidence-based practice is desirable but not always possible, given the lack of randomized controlled trials and the variety of methodologies, outcomes, and assessment tools used in published clinical studies. However, using available evidence-based practice in combination with a defined habilitation plan that considers the child, family, and community defines state of the art medical management.

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THE AMBULANT CHILD (GMFCS level I, II & III)

This part of the paper will also review interventions based on the use of the GMFCS classification (5). The children in GMFCS I, II and III are anticipated to walk with different levels of competencies. The goals for this group include decreasing gait deviation by improving alignment, improving range of motion, improving strength, and decreasing the related issues that may interfere with walking.

Since the last consensus report of the ISPO (6) about the treatment of children with Cerebral Palsy, new medical treatment options to reduce the effect of spasticity has become available. According to the International Classification of Functioning (7), effect of treatment can be described on the level of body function and structure, and on the level of activities and participation. This paper will provide focused systematic reviews highlighting areas that affect activity level for children in GMFCS level I, II and III, by reviewing spasmolytic therapies (Botulinumtoxin A and selective dorsal rhizotomy) for improvement of the level of activities. The efficacy of these treatments on the level of body function and structure (decrease of spasticity / muscle tone) has been demonstrated (8-10).
Botulinum toxin A is a drug, which can be used for intramuscular injections. It blocks the acetylcholine dependent synaptic transmission from the nerve to the motor unit temporary (3 to 6 months). Selective dorsal rhizotomy is a surgical procedure in which sensory roots at the intrathecal space at lumbar level (from S1-S2 to L2) partially are cut bilaterally (11).

Methodology

Keywords for the search in digital medical literature databases were “cerebral palsy and (clinical trial or review) and (botulinum toxin or rhizotomy)“, from November 2003- November 2008, in PubMed, Embase and the Cochrane Library. The data from an Evidence based guideline for the diagnosis and treatment of Cerebral Palsy in the Netherlands (2007) are also used (www.cbo.nl/product/richtlijnen/folder20021023121843/rl_cerebrale_parese_07.pdf/view). Only the results of systematic reviews and randomized clinical trials are considered. The literature was analyzed focusing on a PICO question (Patient population, Intervention, Comparison, Outcome).

Does treatment with Botulinum toxin A improve walking ability in children with CP?

Results of the analysis of the literature are summarized in Table 8. Although many case reports and pre-post studies have reported statistically significant improvement of gross motor function in spastic Cerebral Palsy by means of the Gross Motor Function Measure (GMFM), the evidence in randomized clinical trials is still inconclusive. The selection of patients for treatment is a key issue in the treatment with BTX-A: should all muscles with spasticity be treated or should only limiting impairments for mobility (e.g. pain in an Ankle Foot Orthosis by high muscle tension, crouch gait with fatigue) be treated with a goal oriented rehabilitation program? In the study of Moore, all muscles with spasticity has been treated repeatedly after recurrence of spasticity. In the study of Bjornson, only young bilaterally involved children have been treated in the Gastrocnemius Muscle (GM). At this age, hyperactivity of the GM is a more common problem, while the weakness can

cause limitations at an older age (12). Children with a flexion gait pattern with functional problems, treated with a comprehensive rehabilitation program (physical therapy, orthoses) improved significantly, but criteria for selection are still unclear (13). In this study, the specific contribution of Botulinumtoxin A treatment in the improvement in gross motor function is not clear. The study of Reddihough addressed a heterogeneous group of children and was not based on treatment of functional problems.
Recommendation: Treatment with Botulinumtoxin A can potentially improve walking ability in a selected population of children with spastic CP. Selection criteria for treatment are still unclear.
Implication for orthotics: Botulinumtoxin A is effective in reducing spasticity, and should be considered in case of limitation in the use or in the potential effect of an Ankle Foot Orthosis to improve gait by spasticity related problems. Botulinumtoxin A treatment should be seen as one component of a comprehensive rehabilitation program (physical therapy, exercises, orthoses).

Does treatment with Botulinumtoxin A improve bilateral hand function?

In children with upper limb involvement (hemiparesis, GMFCS level I, II or III), only improvement of bilateral hand function is important to improve the level of activities, as the least involved upper limb will be used as the dominant upper limb. Therefore, the the level of activities is the relevant outcome to assess the improvement of abilities (as dressing, drawing, scribing) by treatment of upper limb function.
In Table 9, the results of systematic reviews and recent randomized clinical trials are summarized. Combined treatment of occupational training and botulinum toxin A treatment showed significant improvement only on subjective goals for treatment, not on capacity and performance assessment tools(14). The secondary trial analysis had not been published for the effect of each therapy modality at the time of this review. Looking to the effect sizes, it is not very likely that each therapy alone could achieve a significant improvement. The study of Russo (15) found comparable results.
Recommendation: There is no scientific evidence for improvement of bilateral hand function by Botulinumtoxin A treatment of the upper limb in children with spastic CP and unilateral involvement.
Implications for orthotists: Botulinumtoxin A is effective in reducing spasticity, and should be considered for the upper limb, only if there are complaints in the use of orthoses on the level of body functions and body structures (pain, contracture, thumb in palm deformity).

Does Selective Dorsal Rhizotomy improve walking ability?

Selective Dorsal Rhizotomy, a neurosurgical procedure to reduce spasticity, is the only well studied medical treatment for children with spastic CP (16). A small but significant improvement in mobility (GMFM-66) has been demonstrated in a meta-analysis , based on 3 Randomized Clinical Trials (Table 10). Moreover, in a long term follow up study the gait improvement after SDR in children with CP turned out to be stable. The criteria for selection of the patients has been described (17;18).
Recommendation: Selective Dorsal Rhizotomy is a treatment option for a selected group of children with bilateral spastic CP to improve mobility.
Implications for orthotics: Selective Dorsal Rhizotomy is effective in reducing spasticity, and in improving mobility. Limitation of the use or effect of an Ankle Foot

Orthosis to improve gait by spasticity related problems can be improved by Selective Dorsal Rhizotomy in a selected group of patients.

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Table 1. What factors are important for life expectancy in children with CP?

Author year /LoE	Research design	Subjects	Intervention	Outcome measures	Main findings
Strauss 2008 /1b	Def of life expectancy and how calculated; revised estimates based on improved analytical methods; issues regarding quality of care; comparison of life expectancies among countries	People with CP receiving services from state of CA between 1983-2002 (as below in Strauss, 2007)	None	Life expectancy using adjusted statistical methods	Consistent findings in life expectancy in large databases in US, UK, and Australia; need to stratify data for least severely affected pts; need to adjust for secular trends in survival.
Strauss 2007 /1b	Statistical analysis using pooled repeated observations method, with logistic regression analysis. Based on data collected by CA (US), including survival, severity of CP, mode of feeding, mobility	People with CP receiving services from state of CA between 1983-2002: 28,513 children (4-14 y) and 30,185 adults (≥15 y)→407,895 person-years.	None	Death rates by age, mobility, feeding status	Declining mortality in those fed by gastrostomy over 20 years (decline of 3.4% per year); increased longevity in most severely disabled.
Hutton 2006 /1b	Analysis of Mersey Cerebral Palsy database (UK); discussion of statistical methods needed for valid assessment of life expectancy.	1647 people with CP registered	None	Survival curves by functional disability	More severe disability associated with poorer survival; severity of motor disability has greater predictive power than severity of cognitive disability. But valid estimation will depend on cohort with accurate birth and death data and clear definitions of functional disability.
Hutton 2002 /1b	Analysis of Mersey Cerebral Palsy database (UK); statistical analysis	1942 individuals registered	None	Survival curves associated with cognitive, motor, and sensory deficits, and birthweight	Survival varies by number and severity of disabilities and birthweight. Poorest prognosis for those with severe motor, cognitive, and visual disability, and normal birthweight.

Table 2. Does gastrostomy improve body structure and functional outcomes in children with CP?

Author year/LoE	Research design	Subjects	Intervention	Outcome measures	Main findings
Henderson 2007 /2c	Multicenter, cross-sectional single observational assessment	75 children <19 y with CP in residential care, 205 with CP living at home	None	Anthropometric measures	Use of feeding tube → ↑weight, height, skinfold thickness. Pts in residential care older; significantly higher percentage with feeding tube
Campanozzi 2007 /3b	Prospective case series	21 consecutive children (mean age 5.8 y) with CP and severe MR	Caloric increase (20%), treated w/ omeprazole if + for GERD	Measurement of growth and motor function before and after 6 mo nutritional tx.	15 (71%) had ↓ FBM; of these, 73% + for GERD and 60% + for CC. 14/15 completed 6-mo tx. BMI ↑ (P=0.08); GMFM signif ↑ in 9/14, unchanged in 5
Day 2006 /2c	Cross-sectional study, multiple measurements	24,920 with CP (ages 2-20 y) receiving svcs from the state of CA from 1987- 2002; 11,496 had Grade V CP	None	141,961 ht/wt measurements→developed growth curves stratified by severity of disability. Compared BMI of pts with grade V CP with and without G-tube	Those with feeding tubes had ↑ height and weight compared to those without (2-5 kg heavier) Increase in weight attributable to use of feeding tubes; most pronounced at 10 y (M and F) and 15 y (M)
Stevenson 2006 /2c	Multicenter, region-based cross-sectional study; cluster analysis methodology	273 children (2-18 y) with CP GMFCS III-V	None	Anthropometric measures; health care use; and social participation over previous 4 weeks	Best growth associated with fewest days of health care use and fewest days of social participation missed. Smaller children more likely to have more severe GMFCS grade. Children with Grade V without feeding tube more likely to be in smallest group than those with feeding tube (4% vs 20%)
Sullivan 2006 /2b	Prospective, longitudinal, observational.	40 children with CP (median age 8.5 y): 39 GMFCS IV-V; 22 with G-tube feeds, 18 fed orally. Compared patients with each other and with 1990 British ref. centiles (without disabilities)	None	Measures of body fat and energy expenditure: resting metabolic rate, total energy expenditure, lean body mass, fat mass; fat mass index, fat-free mass index	Both CP groups had higher body fat and less energy expenditure than ref pop. RMR and TEE lower in G-tube group than orally fed group, but G-tube group had significantly (P<0.02) larger fat mass index than orally fed group. G-tube group 2.41 kg/m^2 >ref; orally fed group 1.15 kg/m^2 >ref standard. Orally fed group had 3.74 kg/m^2 less FFM than ref; G-tube group had 4.58 FFM than ref. (trend, NS)
Craig 2006 /2c	Prospective case series	76 children <18 y (median age 40 mo); 32 with CP, 25 with genetic abn 11 with degenerative disease, 8 w/o dx	Gastrostomy± anti-reflux procedure	Growth; surgical complications; change in symptoms. Growth and sx assessed preop and 3 or 6 mo postop.	Two-thirds had catch-up growth (P=0.001). Major postop complications in 17%. Significant improvements in drooling, vomiting, and constipation

Table 2. Does gastrostomy improve body structure and functional outcomes in children with CP? (continued)

Author year/LoE	Research design	Subjects	Intervention	Outcome measures	Main findings
Kong 2005 /2c	Cross-sectional	110 children-CP (nonambulatory); 48 tube-fed, 62 orally fed, 119 normal controls	None	Measurement of weight and height (height est. from ulnar length in CP), skinfold thickness	Weight-for-height z scores nl in tube-fed group; signif ↓ in orally fed pts w/ CP compared to tube-fed grp and normal controls. Skinfold thickness in CP most reliable at triceps; unreliable in LE (muscle atrophy)
Sullivan 2005/ 2c	Longitudinal, prospective, cohort study; no controls (ref data)	57 children ≤18 y (median age 52 mo) with poor weight gain; 77% Grade V.	Gastroscopy ± fundoplication	Growth/ complications/ health at baseline, 6 mo, 12 mo	Weight gain at 12 mo ($z=+1.24$, $P<0.0001$; mid-upper arm $z=+1.02$, $P<0.0001$.) One serious complication (peritonitis). No change in frequency of chest infxn, but ↓ hospitalizations.

BMI, body mass index; CP, cerebral palsy; FBM, fat body mass; FFM, fat-free mass; FMI, fat mass index; FFMI, fat-free mass index; G-tube, gastrostomy tube; GERD, gastroesophageal reflux disease; GMFCS, Gross Motor Function Classification System; RMR, resting metabolic rate; TEE, total energy expenditure

Table 3. Systematic studies of gastrostomy

Author year	Search strategy	Intervention	Comparison	Methodology of individual studies	Main findings
Vernon-Roberts 2007	Cochrane, Medline, EMBASE, CINAHL ISI Web of Science, Child Health Library; include only RCT	Gastrostomy + anti-reflux tx	Effectiveness of anti-reflux surgery and anti-reflux meds in pts w/neurologic impairments and GER	No studies met criteria	No RCT
Sleigh 2004 (Arch Dis Child)	Cochrane, Medline, EMBASE, CINAHL, LILACS, databases of theses: include CT, systematic reviews, published observational studies; exclude nonsystematic reviews, qualitative studies	Gastrostomy / jejunostomy	Death rate, growth, GER, complications / caregiver wellbeing	2 cohort studies, 15 case series, 8 case reports	Inconsistent criteria for same outcomes; little robust evidence re: gastrostomy
Sleigh 2004 (Cochrane Database)	Cochrane, Medline, EMBASE, CINAHL, LILACS, databases of theses; include only RCT	Gastrostomy / jejunostomy	Nutritional status for children with CP ≤16 y	No studies met criterial	No RCT
Samson-Fang 2003	Medline, CINAHL, HealthSTAR, ClinPSYC, Cochrane Database: include clinical studies comparing gastrostomy with oral feeding	Gastrostomy	Effectiveness in children <18 with CP	10 studies, all with levels of evidence IV and V	Most show improvement after gastrostomy. Low levels of evidence; few controls; little statistical data, little info about caloric intake or body composition

Table 4. Bone Health: Risk factors for fracture in children with CP

Author year /LoE	Research design	Subjects	Intervention	Outcome measures	Main findings
Khoury 2007/3b	Case-control	Non-ambulatory, (2-20 y/o, mean 11 y)	None	Fracture	Risk of fx ↑ in those with BMI >17; risk factors: combination of BMD of femur + BMI
Presedo 2007 /3b	Case-control	156 with fx,150 without; 55% with spastic quadriplegia	None	Fracture	82% fx lower extremity; trauma was cause of 32% of fx; 45% fx with unknown cause. Significant risk factors: severity of CP, ↓ ambulatory status, anticonvulsants
Ko 2006 /3b	Case-control	Children living at a disabilities unit in Hong Kong, 4-19 y/o (mean 10 y); 19 with fx, 90 without	None	Fracture	Risk factors: Lower weight for age, longer duration of post-surgical immobilization
Leet 2006 /3b	Case-control	50 children with fx, 368 without fx (1.3-18 y/o, mean 10.6 y), 58% quadriplegia	None	Fracture	Increased risk for fx in older pts, those with mixed tone, feeding tube, sz, valproate, use of standing equipment
Stevenson 2006 /2c	Longitudinal, cross-sectional, multicenter	297 children with CP, GMFCS III-V (2-19 y/o, mean 9.3 y). Baseline: 46 w/ fx, 251 without fx	None	Fracture	Follow-up: 24 fx among 260 pts followed for 605 person-years. Estimated fracture rate 4.0% per year. Disease severity not significant.
King 2003 /3b	Case-control	Non-ambulatory pts, 5-48 y/o (mean 15 y) with spastic quadriplegia	None	Fracture	Lower BMD z-score (–2.81 vs. –2.11)
Bischof 2002 /3b	Case-control	Pts living in care facility in South Africa, 6-29 y/o (mean 17.5 y), 20 with fx, 20 without fx	None	Fracture	Anticonvulsant use, higher ALP, PTH, lower Pi
Henderson 1997 /2b	Retrospective cohort	43 children (3.3-17.2 y/o, mean 7.9 y) with spastic CP. Baseline BMD measurements taken; followed for 162 person-years	None	Fracture	9 fx; most involved femur; most occurred with minimal trauma. Previous fx and hip spica casting significant for predicting future fx. BMD z-score not significant in predicting fx.

ALP, alkaline phosphatase; BMD, bone mineral density; BMI, body mass index; CP, cerebral palsy; fx, fracture; GMFCS Gross Motor Function; Classification System; Pi, inorganic phosphorus; PTH, parathyroid hormone; sz, seizure

Table 5. Bone Health: Effect of bone mineral density in children with CP

Author, year	Research design	Subjects	Intervention	Outcome measures	Main findings
Henderson 2005 /2c	Prospective longitudinal, observational	69 patients (55 observed >2 years, 40 observed >3 years); GMCFS III-V; 2-17.7 y/o (mean 9 y)	None	BMD	Total body BMD increased with age but distal femur BMD decreased. Risk factors for low BMD: greater severity of disease, poorer growth/nutrition, advancing age. No variable at baseline was a predictor for change in BMD
Henderson 2004 /3b	Cross-sectional	107 with CP, GMCFS III-V, 2-21 y/o (mean 12 y)	None	BMD	Additive risk factors. Weight z-score best predictor of BMD z-score. Risk factors: lower weight z-score, increasing age, greater disease severity, prior fractures, anticonvulsant use, feeding difficulties.
King 2003 /3b	Case-control	48 pts (42 w/ CP), non-ambulatory with spastic quadriplegia, 5-48 y/o (mean 15 y); number of controls not reported	None	BMD	Lower BMD in patients. Age, weight, or height, serum analysis, anticonvulsant use not significant
Nakano 2003 /2c	Cross-sectional	123 institutionalized adults with CP, 21–46 y/o	None	BMD	Risk factors: use of anticonvulsant in both male and female, higher ALP in men, decreased ambulation in women
Unay2003 /3b	Case-control	67% tetraplegic, 55% non-ambulatory, 2.7-14.2 y/o (mean 5.1); 40 with CP, 40 without	None	BMD	Tetraplegic patients had lower BMD than hemiplegic; ambulatory status and physical therapy not significant
Henderson 2002 /3b	Case-control	117 with CP, >250 without; GMCFS III-V, age 2-19 y/o (mean 9.7 y)	None	BMD	Osteopenia in most non-ambulatory children by 10 y. Risk factors: neurological impairment, feeding difficultly, anticonvulsant use, lower triceps skinfold
Tasdemir 2001 /3b	Case-control	63% non-ambulatory, 50% malnourished, 10 mo to 12 y/o, (mean 4.1 y); 24 with CP, 19 without	None	BMD	BMD less in all patients, even less in non-ambulatory patients. Risk factors: higher Ca and P levels. Malnutrition, anticonvulsant use not significant
Chad 2000 /3b	Case-control	17 with spastic CP, 894 without, good nutritional status, 76% non-independent ambulators, 7.6-13.8 y/o (mean 10.9 y)	None	BMD	Risk factor: non-independent ambulation
Henderson 1995 /3b	Case-control	139 children (3-15 y/o, mean 9 y) with spastic CP; 95 controls	None	BMD	BMD nearly one SD greater in controls than pts. Risk factors: ambulatory status, nutritional status, pattern of involvement, duration of immobilization, low Ca intake. Previous fx, anticonvulsant use, vit D levels not significant

ALP, alkaline phosphatase; BMD, bone mineral density; BMI, body mass index; CP, cerebral palsy; fx, fracture; GMFCS Gross Motor Function; Classification System; Pi, inorganic phosphorus; PTH, parathyroid hormone; sz, seizure

Table 6. Do oral medications improve body structure and function for children with cerebral palsy?

Author year/LoE	Research design	Subjects	Intervention	Outcome measures	Main findings
Murphy 2008 /2	Randomized, double-blind, crossover trial	8 children (mean age 11.5 y) GMFCS Level III-V	Modafinil vs. placebo	Modified Ashworth; Caregiver Priorities and Child Health Index of Life with Disabilities (CPCHILD),	No improvement in spasticity or quality of life
Lubsch 2006 /2b	Retrospective chart review	87 children (<18 y/o) with spasticity due to CP (80%) or TBI (20%)	Oral baclofen and/or clonidine	To calculate mean dosages of baclofen and clonidine based on the duration of spasticity postinjury. Secondary objectives: determining correlations between dosage and age, injury type, location of spasticity, comorbid seizures, or concomitant antispasticity meds.	86 on baclofen, 31 on clonidine; 33% of those on baclofen also on another antispasticity med. Strongest predictor of both baclofen and clonidine dosage in children with CP or TBI is time since injury. Patterns: baclofen dosages for age, 2 y: 10-20 mg/day (max 40 mg/day), 2-7 y: 20-30 mg/day (max 60 mg/ day), 7 y: 30-40 mg/day (max 200 mg/day) and clonidine maintenance dosage of 0.4 mg/day (max 1.8 mg/day).
Scheinberg 2006/ 2	Double-blind, randomized crossover study	15 children (7.4 y ± 2.7 y) with CP, GMFC IV-V	baclofen	Modified Tardieu Scale Goal Attainment Scale, Pediatric Evaluation of Disability Inventory	Improvement in GAS; No change in Tardieu, PEDI, inconsistent parent responses.
Vásquez-Briceño, 2006/2	6-month double-blind RCT	10 children with CP, 30 controls	Tizanidine vs. placebo	spasticity, Ashworth scale, posture tone scale, reflex scale and liver function test	Significant ↓ Ashworth scale and reflex scale vs placebo ($p = 0.0001$); no abnormalities in liver function tests
Mathew 2005/ 2	RCT	114 children with CP	Single bedtime dose of diazepam vs placebo	Well being of child (reported) Burden of care (reported) Behavior profile of child (observed)	59 children diazepam, 55 placebo Statistically significant changes with diazepam: Improved well being Lessened burden of care Improved behavior
Mathew 2005/ 2	RCT	173 children (<12 y/o) with CP	2 different doses of diazepam (half-dose vs full-dose) vs placebo	MAS, goniometry	Statistically significant improvements in movement and spasticity. Full dose much more effective than half-dose
Vargus-Adams, 2004/ 4	Prospective, uncontrolled case series	11 children (51 mo ± 11 mo) Grade II-IV CP	baclofen	GMFM, MAS	No significant change n MAS, GMFM; improved parent report
Montane 2004/	Systematic review of double-blind RCT PubMed, Medline, Cochrane	469 patients; 67 (3 studies) with CP	Dantrium-37 Baclofen-20 Placebo-57	Crossover Crossover Parallel groups	Improvement in Ashworth scale No functional change

CITB, continuous intrathecal baclofen; CP, cerebral palsy; DDS, drug delivery system; GMFCS, Gross Motor Function Classification System; ITB, intrathecal baclofen; LE, lower extremity; MAS, modified Ashworth Scale; QALY, quality-adjusted life-years; ROM, range of motion

Table 7. Does intrathecal baclofen improve body structure and function in children with cerebral palsy?

Author year	Search strategy	Subjects (description, number, etc.)	Intervention	Comparison	Methodology of individual studies (RCT, case study etc)	Main findings
Kolaski 2008	Electronic databases 2000-2007; people with CP and ITB	29 studies (including 4 studies with ≤50% subjects with CP who were not analyzed separately)	ITB (TD or CITB)	Spasticity, neurophysiologic assessment, dystonia, ROM, orthopedic deformity, ADL, Care/comfort, caregiver burden	29 studies: one level II, all others level IV or V; no consistent assessment tools.	Spasticity: decreased in UEs/ LEs Neurophysiologic: ↓ tibial nerve H reflex and LE nerve flexor reflexes Dystonia: ↓in mean BADS in 63/69; in smaller study no significant differences Comfort/pain/sleep: Improved pain and comfort in multiple anecdotal studies Orthopedic deformities ↓hip migration; accelerated scoliosis in some studies—possible ↑ asymmetrical paraspinal muscle relaxation Motor function: Mixed findings on GMFCS, functional mobility Caregiver burden: anecdotal improvement, little statistical data.
Kolaski 2007	Electronic databases 2000-2007; people with CP and ITB	36 studies documenting complications, non-controlled case series or single case reports, plus 21 other reports with relevant information	ITB	Adverse effects of medication; surgical complications; complications related to presence and function of drug delivery system; complication trends	Non-controlled case series, single case reports	Variable adverse reports (CNS, GI). Surgical complications in 25% to 40%. DDS problems in ≤15%. Most common problems = infections, catheter problems, CSF leaks. Problems seen short-term and long-term after placement. Probs more common in pts ≤8 y/o than older patients.
de Lissevoy 2007	15 studies of ITB in children with CP—health utility index developed by physicians; cost calculated based on insurance claims; bootstrap modelling applied	484 with severe CP	ITB	Cost-effectiveness analysis with effectiveness measured in quality-adjusted life-years (QALY) Incremental cost (total cost of ITB minus total cost of usual care) divided by quality adjusted life-year (QALY ITB minus QALY usual care)	Variable	Benefit of ITB in children = $42,000 per QALY (cochlear implant = $12,774; Palivizumab prophylaxis of RSV in children with congenital heart disease =$114,337)

BADS, Barry-Albright Dystonia Scale; CITB, continuous intrathecal baclofen; CP, cerebral palsy; DDS, drug delivery system; GMFCS, Gross Motor Function Classification System; ITB, intrathecal baclofen; LE, lower extremity; QALY, quality-adjusted life-years; ROM, range of motion; RSV, respiratory syncytial virus; TD, test dose; UE, upper extremity

Table 8. Does treatment with Botulinum toxin A improve walking ability in children with CP?

Author year /LoE	Research design	Subjects	Intervention	Outcome measures	Main findings
(19) 2008 / 1c	RCT, placebo controlled	64 CP children, mean age 5.3 / 4.8 years intervention and placebo group . Mean GMFM (88?) scores 66.5 / 69.1	At most 3-monthly Bont-A injections at different sites of the lower limb during 2 years	GMFM (88?), PEDI	NS
(20) 2007 / 1b	RCT, placebo controlled	33 diplegic CP children (mean age 5.5 yrs), GMFCS 1-3	12 IE Bont-A in Gastrocnemius M. bilateral	GMFM-88/66, ECI, COPM, GAS	GMFM-88 difference score at 24 weeks (P<.001), GMFM-66 (P<.03); CPOM score at 12 weeks (P<.04), NS at 24 weeks. ECI NS.
(21) 2007 /1b	RCT, control group usual care	46 CP children, GMFCS 1-4	Multilevel Bont-A, SC, AFO, PT	GMFM-66, EC	GMFM-66 at 24 weeks (P<.01). EC NS.
(22) 2006 / 1a	Systematic review Cochrane, Medline, Embase, Cinahl, DARE, PEDro, SCI, Australasian Medical index	CP children	Bont-A injections Lower and Upper Limb in combination with other therapies	Gait	Insufficient evidence to support or refute the use of therapy interventions after Bont-A injections.
(23) 2002/ 1b	RCT cross over, Bont-A with PT or only PT.	49 CP children (mean age 4yrs 1mth) GMFCS 1-4	Bont-A injections Lower Limb, PT	GMFM-88	NS
((24) 2006/2a	Systematic review of all available avidence	CP children	Bont-A injections in Upper and Lower Limb in combination with other therapies	Gait, GMFM, COPM, GAS	Expert opinion based recommendations

RCT = Randomized Clinical Trial; CP = Cerebral Palsy;GMFCS = Gross Motor Function Classification System; Bont-A = Botulinum toxin A; GMFM = Gross Motor Function Measure; COPM = Canadian Occupational Performance Measure; EC(I)= Energy Cost (Index); GAS = Goal Attainment scaling; SC = serial casting; AFO = Ankle Foot Orthosis; PT = Physical Therapy; NS = not significant difference. LoE = Level of Evidence Oxford Centre for Evidence-based Medicine Levels of Evidence (May 2001).

Table 9 Does treatment with Botulinumtoxin A improve bilateral handfunction?

Author year /LoE	Research design	Subjects	Intervention	Outcome measures	Main findings
(25) 2007 / 1c	RCT 4 treatment groups (Bont-A + OT, Bont-A, OT, control)	72 CP children, 33 unilateral, 39 bilateral, mean age 5 yrs 11mth	Bont-A injections upper limb, OT.	COPM,GAS, MA, QUEST of treated limb, PEDI	COPM and GAS significant improvement Bont-A +OT group versus control.# ; MA, QUEST, PEDI NS
(26) 2007 / 1b	RCT Bont-A + OT, OT only	43 unilateral CP children, mean age 8.6 yrs.	Bont-A injections upper limb , OT	AMPS, PEDI, PEDsQL, GAS	At 3 mth GAS (p=.001), at 6 mth NS; others NS
(27) 2006 /1a	Systematic review of PUBMED, CINAHL, PICARTA, EMBASE, PEDRO, and the Cochrane Controlled trial register.	64 CP children	Bont-A injections of the upper limb	Heterogeneous sets of outcome measures were used in 3 RCT's .	NS effects on functional outcome measures.
(28) 2004 /1a	Cochrane systematic review Cochrane, Medline, Embase, Cinahl databases.	14 and 29 CP children	Bont-A injections of the upper limb	PEDI, QUEST	No evidence to support or to refute the efficacy of Bont-A injections of the upper limb.
((29) 2007/1a	RCT, comparing 3 Bont-A treatments with 2, in combination with OT.	42 unilateral CP children (4 yrs, sd 1.61), GMFCS 1.	OT, Bont-A injections upper limb.	QUEST, COPM,PEDI, GAS	NS between the 2 groups

RCT = Randomized Clinical Trial; CP = Cerebral Palsy; Bont-A = Botulinum toxin A; COPM = Canadian Occupational Performance Measure; QUEST = Quality of Upper Extremity Skills Test; PEDI = Pediatric Evaluation of Disability Inventory; GAS = Goal Attainment scaling; MA = Melbourne Assessment of Unilateral Upper Limb Function; AMPS = Assessment of Motor and Process Skills; PEDsQL = Pediatric Quality of Life Inventory; OT = Occupational Therapy; NS = not significant difference. # = no adequate statistical trial analysis. LoE = Level of Evidence Oxford Centre for Evidence-based Medicine Levels of Evidence (May 2001).

Table 10 Does Selective Dorsal Rhizotomy improve walking ability?

Author year /LoE	Research design	Subjects	Intervention	Outcome measures	Main findings
(30)2002 / 1a	Meta analysis of 3 RCT's	90 CP GMFCS 1-4; mean age 5,5 yrs	SDR surgery with PT versus PT only	GMFM-66	GMFM-66 + 2.66 (P=.0002)
(31) 2007 / 1b	Prospective cohort study	11 of 14 ambulatory CP children 10 years after surgery, mean age 27.9 yrs.	SDR surgery and PT 10 years ago	Gait	NS change in gait.

RCT = Randomized Clinical Trial; CP = Cerebral Palsy; GMFCS = Gross Motor Function Classification System; GMFM = Gross Motor Function Measure; SDR = Selective Dorsal Rhizotomy; PT = Physical Therapy; LoE = Level of Evidence Oxford Centre for Evidence-based Medicine Levels of Evidence (May 2001).

MEDICAL MANAGEMENT: POINTS OF DISCUSSION

Jan Willem Gorter, MD PhD
***CanChild* Centre for Childhood Disability Research**
McMaster University, Canada

Since the first publication on 'cerebral palsy' by Little (1862) up to the most recent publication on the revised definition of CP (Rosenbaum, 2007) the picture of children and adults persons with CP has changed from a child with deformities due to asphyxia (a good example of the medical model) to a developmental childhood onset disability requiring a lifespan perspective in the context of the family (a developmental and social model). The framework of the International Classification of Functioning, Disability and Health (ICF, 2001) goes beyond the health condition (CP) and the direct impairments in body function and structure. It also shows the person's functioning in terms of activities / participation and the interaction (bidirectional) between the level of functioning and environmental factors (family) and personal factors. Unfortunately, the framework does not show the paediatric perspective, in which growth and development are the key factors.

In last decades two major shifts in the medical management of CP can be observed:

1. A shift from impairment based medicine (i.e. cure or fix the problem) to a habilitation approach in order to enhance a person's developmental capabilities and performance (activities and participation).

2. The shift from patient-centered to a family-centered approach, i.e. i) the acknowledgement of the unique strengths, resources and needs of all family members, and ii) the emphasis on partnership between the patient, the family and service providers.

The functional classification of a heterogeneous condition like CP by the Gross Motor Function Classification System (GMFCS) helps us to make meaningful distinctions, (functional) prognosis (e.g. prediction of gross motor function, associated impairments) and to set realistic goals given a certain level of functioning. The authors of this section used the GMFCS levels as a framework for planning medical management (GMFCS level I-III, potential walkers versus IV & V, potential wheelchair users). In clinical practice, however, the service provider is often challenged by the variability in children with CP and the complexity in individual cases. The central question in medical management therefore is: what works best for whom and when?

In the beginning of the 21st century we must accept that, although attempts are being made to prevent and to cure CP (the ultimate goal), the reality is that CP is a lifelong condition both for the child and the family. In general the following goals in the medical management in CP can be distinguished:

1. To prevent additional impairments (e.g. life threatening complications, malnutrition/ obesity, secondary musculoskeletal impairments)
2. To Cure impairments (e.g. relieve of current distress)
3. To Improve / optimize activities and participation (optimizing daily functioning)

4. To Manage a life with CP in society (e.g. maintenance, respite care, self-management)

The most recent and high-level evidence papers (systematic reviews) were used if available. The search method of the literature is missing and should be stated in the method section. A good point is that the authors addressed issues of medical management following the PICO framework (patients, intervention, control and outcome). The majority of the studies lack a long term follow-up (except from the survival studies, studies on nutritional status and 1 study on the long term follow=u after SDR) and information on side-effects / complications. Given the complexity and heterogeneity of CP it would be helpful to identify the children who might benefit most of the intervention and who do not. I think that we need a better understanding of the natural course and determinants of functioning in children, youth and adults with CP. Longitudinal studies (e.g. the PERRIN program in the Netherlands, Move & Play study in Canada, Australia) are currently underway.

Points for discussion

Life expectancy
- Overall good life-expectancy leads to an increasing number of adults with CP. There is a need for epidemiological studies.
- Implications for orthotics: long-term use in a developmental /realistic perspective (person, parent & professional).

Nutritional status / Gastrostomy
- Optimal weight is unknown. Recommendation: (at least annual) documentation of body weight and length (BMI)
- Orthotics: adaptations (cost issues?)

What treatment works best for who? When to start and for how long? What information do we have about cost-effectiveness of medical interventions (in terms of direct and indirect costs, think of all the time and effort of the parents!)?

Botulinum Toxin-A – walking ability
- What do we know about timing and duration of BTX-A treatment: in relation to the development of gross motor skills (1-7 years), efficiency of walking (7-10 years) and maintenance (10 – older)
- Effects of BTX-A in combination with AFO's are suggested to prevent secondary musculoskeletal impairments. Are we able to find evidence for this statement?
- Outcome measurement issues:
  - Selecting the most appropriate measure GMFM-88 with and without orthosis, quality of movement (Quality-FM), efficiency of gait?
  - Interpreting the data (what is a clinically relevant difference?)

Selective Dorsal Rhizotomy – walking ability
- SDR is effective in the short term. Think of the lifespan perspective: is it worth the effort? (perspectives of person, parent and professional)

Botulinum Toxin A bilateral handfunction

- Indication is more specified: prevention of deformities, relieve of distress, optimizing function (no evidence), ease of care?
- Outcome measures: with orthosis?
- Combination with intensive therapy?

Spasmolytica / tone reduction

- **Oral**
  - Side-effects, dosage and duration
  - Goal setting
  - Focal versus generalised spasticity/ hypertonia

- **ITB (GMFCS level IV-V)**
  - Indication criteria: ease of care/handling
  - Long-term side effects (safety)
  - Organisation of care (operation, follow-up)

In line with other interventions I can see the knowledge and evidence growing on medical interventions in CP: to prevent impairments, to cure impairments, to optimize functioning and to manage problems in CP. Also in this field there are more questions than answers. Directions for further discussion/ future research are:

Organisation of medical care

- How do we organise our health care in children with complex lives, do children and youth have access to multidisciplinary teams or the services they are looking for? Do we give youth a voice and evaluate their perception of the services they get?
- How do we take care of the medical management transition into the adult world (is there a formal link between paediatric and adult services, but also is there a link between paediatric services and family doctors / community services?)
- Do we address issues as the availability of services / medical expertise required for specific interventions (e.g. dorsal rhizotomy, ITB)

Clinical practice

- Do we really incorporate a lifespan perspective in our medical management?
- Are we ready to reconsider our way of thinking: from impairment based therapy towards an approach focussing on activity / participation?
- How good are we in informed shared clinical decision making in medical management (do we know what information should be shared, and how we do make decisions?) Do we deal with the different perspectives (person (patient), parent, professional) in the short and in the long term?

ORTHOPAEDIC SURGERY IN THE MANAGEMENT OF CEREBRAL PALSY

H. Kerr Graham MD FRCS (Ed) FRACS
Professor of Orthopaedic Surgery, University of Melbourne
Murdoch Childrens Research Institute, Australia

Percutaneous tenotomy of the Achilles tendon was the first orthopaedic operation, introduced in the early part of the 19th Century in Germany by Louis Stromeyer. Although the initial application of percutaneous Achilles tendon tenotomy was for equinus associated with club foot or polio, the English physician, William John Little, who himself had a clubfoot deformity, travelled to Germany to observe Dr Stromeyer's work. Dr Stromeyer performed an Achilles tenotomy on Dr Little who was so impressed with the results that he introduced the technique in England and became such an advocate for research into the causes of cerebral palsy and treatments based on tenotomies, that in England cerebral palsy is still referred to as "Little's disease". Despite the nearly 200 years which have passed since the introduction of tenotomy for equinus deformities in cerebral palsy, controversy still exists as to the best method for the correction of fixed equinus deformity in children with cerebral palsy.

Animal Models in Cerebral Palsy

A significant restraint in basic research in the understanding of deformities in cerebral palsy is the lack of a suitable animal model. Many researchers have attempted and failed to develop a reproducible model for human cerebral palsy. Two studies have used the hereditary spastic mouse. The first confirmed the imbalance between the longitudinal growth of the gastroc-soleus muscle tendon unit in relation to tibial growth as the primary cause for fixed equinus deformity[1]. A later study showed that the equinus deformity could be prevented by the injection of Botulinum toxin A within the first few days of life, implicating spasticity as a causative factor in the development of fixed equinus deformity[2].

Natural History

In the absence of a suitable animal model, the study of the natural history of the development of fixed musculoskeletal deformities assumes even greater importance. The development of cerebral palsy registers, the development of a common language for the classification of gross motor function (GMFCS) and the development of new measurement tools has improved our understanding of the natural history of musculoskeletal deformities in contemporary populations of children with cerebral palsy. These studies suggest that diminished voluntary activity may have a greater impact on the severity of musculoskeletal deformity than spasticity or muscle imbalance.

SINGLE LEVEL SURGERY TO SINGLE EVENT MULTILEVEL SURGERY

A key development in strategic thinking of how to manage fixed musculoskeletal deformity in cerebral palsy has been the move from single level surgery, repeated at frequent intervals throughout childhood, to single event multilevel surgery (SEMLS). The weakness of repeated surgery at single anatomic levels (ankle, knee, hip) was identified by Rang, Gage and others but it was Rang who characterised this approach

as the "birthday syndrome"[3]. As the surgeon starts surgical correction at the ankle level for equinus deformity, followed one to two years later by surgery at the knee level for flexion contracture, followed by surgery at the hip level for hip flexion contracture, the child undergoes repeated hospitalisation and spends each birthday either in hospital, in cast or in rehabilitation. A second major development has been the emphasis moving from soft tissue surgery (lengthening of contracted muscle tendon units and tendon transfers) to an emphasis on bony reconstructive surgery. The concept of lever arm deformities has been articulated by Gage and others and includes torsional deformities of long bones, typically medial femoral torsion and lateral tibial torsion as well as joint instability including hip displacement and mid foot breaching[4]. Lever arm deformities have been linked to lever arm disease. Specifically, if the already weakened muscle tendon units are unable to generate adequate extensor moments, progressive crouch gait will ensue. With the development of multilevel surgery as a practical reality, the need for objective measurement to identify gait deviations and measure the outcomes of multilevel procedures became self evident. The development of practical instrumented gait analysis has been pivotal in the development of multilevel surgery and in assessing outcomes. The majority of publications in the field of surgery focus on the surgical procedure and its outcome and the commonest publication is a retrospective cohort study describing the outcome of a specific surgical procedure. The weakness of this approach in cerebral palsy has become increasingly recognised with publications detailing widely different outcomes for surgery for equinus deformity according to the cerebral palsy subtype (hemiplegia versus diplegia) as well as by GMFCS. The ability to describe cerebral palsy by movement disorder, topographical distribution and gross motor function has resulted in new thinking in the application of surgical techniques, the design of surgical trials and the reporting of outcomes[5].

The reporting of surgical outcomes is also in the process of rapid change. The earliest studies in the literature focus on the morbidity and mortality of surgical procedures and report outcomes in the broadest terms, usually in the impairment domain. Over time, standardised goniometric measurements and observational gait scales became more widely used. The development of instrumented gait analysis has been pivotal in the assessment of gait before and after intervention in a valid and reliable manner. With the introduction of the International Classification of Functioning by the World Health Organisation, a much broader perspective is now being employed in the measurement of outcomes of orthopaedic procedures particularly single event multilevel surgery (SEMLS).[6] In this area the remaining challenges include the development of responsive measures for the measurement of participation, quality of life and cost effectiveness of combined orthopaedic, orthotic and rehabilitation measures.

Orthopaedic surgeons have gradually embraced a multidisciplinary context for the application of orthopaedic surgery in the management of children with cerebral palsy. Such approaches may include spasticity management, optimising nutrition, strengthening programs, and the use of orthoses in the context of orthopaedic surgery for the correction of fixed musculoskeletal deformities.

ORTHOPAEDIC SURGERY: REVIEW OF EFFECTIVENESS

Orthopaedic Surgery for children and adolescents with cerebral palsy may be classified as follows:

- Upper limb surgery

- Lower limb surgery:
    - Single-level surgery
    - Multi-level surgery - unilateral SEMLS
      - bilateral SEMLS
- Surgery for hip displacement:
    - Preventative
    - Reconstructive
    - Salvage

This review paper concentrates provides an overview of upper limb surgery, hip surgery and lower limb surgery; the subsequent paper examines the current evidence base specifically for the surgical management of equinus deformity in cerebral palsy

UPPER LIMB SURGERY

Surgery for the upper limb in cerebral palsy is performed much less frequently than surgery for the lower limb and there are enormous variations in practice around the world. As with lower limb surgery, the multidisciplinary context including the need for movement disorder management, strengthening, and the use of orthoses is increasingly recognised. In addition as with the lower limb there has been a move from the use of single level procedures to a more integrated multilevel approach. This has included the need for more global outcome measures including validated functional scales. However kinematic analysis for upper limb movements lags well behind gait analysis in the lower limb. This is principally because the upper limb has such a wide repertoire of movements and lacks the single stereotypical pattern of gait.

A systematic review of interventions for the upper limb in children with cerebral palsy was conducted by Boyd and colleagues and reported in 2001[7]. The development of the Manual Ability Classification Scale (MACS) as the equivalent of the GMFCS is a major step forward in upper limb assessment. Other scales such as the Melbourne Assessment of Unilateral Limb Function are very useful.

As with lower limb surgery, increasing numbers of surgeons are moving from a single level approach to a “single event multilevel surgery” approach in reconstructive surgery of the hemiplegic upper limb. One of the few recent reports in the literature detailing outcomes of a true multilevel surgery report is that by Johnstone and colleagues in 2003[8]. In this study a true multilevel surgery protocol was applied and a useful analysis of outcomes included cosmesis scores, functional scores, hygiene scores and dressing scores before and after surgery.

ORTHOPAEDIC SURGERY IN THE MANAGEMENT OF HIP DISPLACEMENT: REVIEW OF EFFECTIVENESS

The natural history of hip displacement in children with cerebral palsy has become clearer following large population based studies**.** The study from the south of England by Scrutton and colleagues is unique because the data collection was longitudinal, objective (based on serial hip radiographs) and involved both a large data set as well as high levels of ascertainment[9]. These authors have confirmed the high incidence of hip displacement in children with cerebral palsy, the timing and rate of displacement and have espoused the need for regular clinical and radiological hip surveillance. The concept of hip surveillance has been employed by other centres and recently reviewed[12,13]. Additional studies on the natural history of hip displacement in children with cerebral palsy have focused on population based studies based on cerebral palsy registers and the use of the GMFCS[10,11]. These studies confirm that the incidence of hip displacement in children with cerebral palsy is approximately 33% and is directly related to GMFCS but not related to the type of movement disorder[10,11]. Taken together these studies make a pervasive case for hip surveillance and early preventative surgery[12,13].

A key concept in hip surgery has been the development of a simple classification by Miller and colleagues suggesting that hip surgery be classified as preventative, reconstructive or salvage[14]. Preventative surgery refers to procedures principally adductor releases, which are designed to prevent progressive hip displacement in children who show early signs of hip displacement on hip radiographs. When preventative surgery fails or children present with very displaced or dislocated hips, reconstructive surgery may be employed. This refers to various combinations of adductor releases, femoral osteotomy and pelvic osteotomy. This is being developed into the concept of one stage hip reconstruction with reports from several centres detailing good results[15,16,17].

Some older children and adolescents who present with severe hip displacement combined with severe femoral head and/or acetabular deformity may not be suitable candidates for reconstructive surgery. If severely destroyed hips are symptomatic, some form of salvage surgery may be required all of which involve loss of the hip joint. These procedures include various forms of excision of the femoral head, valgus osteotomy, arthrodesis, and arthroplasty.

Prevention of Hip Displacement

The prevention of hip displacement has been attempted by a variety of methods including orthoses, interventions for spasticity, and surgery or combinations of the above.

Given that the risk factors for progressive hip displacement in children with cerebral palsy are now reasonably well known, early prevention by abduction bracing using either bracing at the hip level or a more extensive bracing system including the trunk and pelvis, would seem to be an attractive option. Isolated bracing or postural control systems have been reported in pilot studies but have not been studied in adequately powered randomised trials. There are good grounds to suggest that orthoses alone might not be effective including poor tolerance and the developing understanding of proximal femoral geometry as a major contributor to hip displacement in children with cerebral palsy.

Spasticity Interventions Combined with Bracing

Because of the reservations outlined above, that bracing children with high levels of adductor tone might not be well tolerated, Graham and colleagues studied the effects of combining injections of Botulinum toxin A to the hip adductors and hamstrings, combined with the SWASH brace (Sit Walk And Standing Hip Orthosis) in a large multicentre randomised clinical trial[18]. The principal findings of their study was that the combined intervention was relatively well tolerated and did result in a measurable decrease in the rate of hip migration. However hip displacement continued in both the treatment and control group but simply at a lower rate. In a follow up study, it was determined that although fewer patients had proceeded to hip surgery during the trial, after exit from the trial, the majority of children in both the treatment and control groups required extensive surgery to keep their hips in joint including both adductor surgery as well as bony reconstructive surgery. They concluded that the combination of injection of Botulinum toxin and the SWASH brace had such a minimal effect on hip displacement that it could not be recommended either in terms of safety or efficacy.

It is important to note that neither selective dorsal rhizotomy (SDR) nor intrathecal Baclofen (ITB) have been effective in preventing hip displacement in children with cerebral palsy and there is some evidence that SDR may increase the risk of hip displacement.

The failure of interventions for spasticity to prevent hip displacement in children with cerebral palsy and the increasing recognition of the high incidence of abnormalities in the proximal femur led Robin and colleagues to conduct a population based study of proximal femoral geometry in children with cerebral palsy. This study found a very high incidence of both increased femoral neck anteversion (FNA) across all GMFCS levels as well as progressive increase in neck shaft angle (NSA) which was directly related to GMFCS level[19]. They concluded that the shape of the proximal femur is largely determined by gross motor function as determined by GMFCS and suggests that bony reconstructive surgery might be required to stabilise the hips in many children with cerebral palsy. The results of this study led Yu and colleagues to investigate the effectiveness of hip adductor surgery for the prevention of hip displacement in children with cerebral palsy.

In 2004 Stott and Piedrahita reported on the effects of surgical adductor releases for hip subluxation in cerebral palsy, an AACPDM evidence report[20]. This was an extensive, systematic, structured review which followed AACPDM guidelines. It provides a detailed over view of the extensive literature in the field with an excellent summary of the findings of the principal studies and a useful commentary on the strengths and weaknesses of the evidence. They concluded that "the body of evidence for adductor surgery comprises very low levels of evidence. There is no level I or II study of hip adductor release for hip subluxation and most studies were coded as level IV. Small sample sizes, heterogenous interventions, poorly defined outcome measures and lack of statistical analysis were common reasons for the judgement of weak internal validity".

Yu and colleagues embarked on a single centre, long term, retrospective analysis of the outcomes of adductor surgery in children with cerebral palsy in which they stratified their analysis by GMFCS level in the form of Kaplan Mier survivorship

analysis. Additional factors investigated were age at index surgery, migration percentage (MP) at initial surgery, type of surgical procedure and experience of the surgeon. They found that the success of adductor releases in the prevention of hip displacement and the need for additional surgery, was determined principally by GMFCS level. Children at GMFCS level II had a very high success rate and children at GMFCS V a very low success rate. They concluded that studies investigating adductor releases must be analysed by GMFCS level and that the abnormal shape of the proximal femur might be the most plausible explanation for the high failure rate at GMFCS levels IV and V[21].

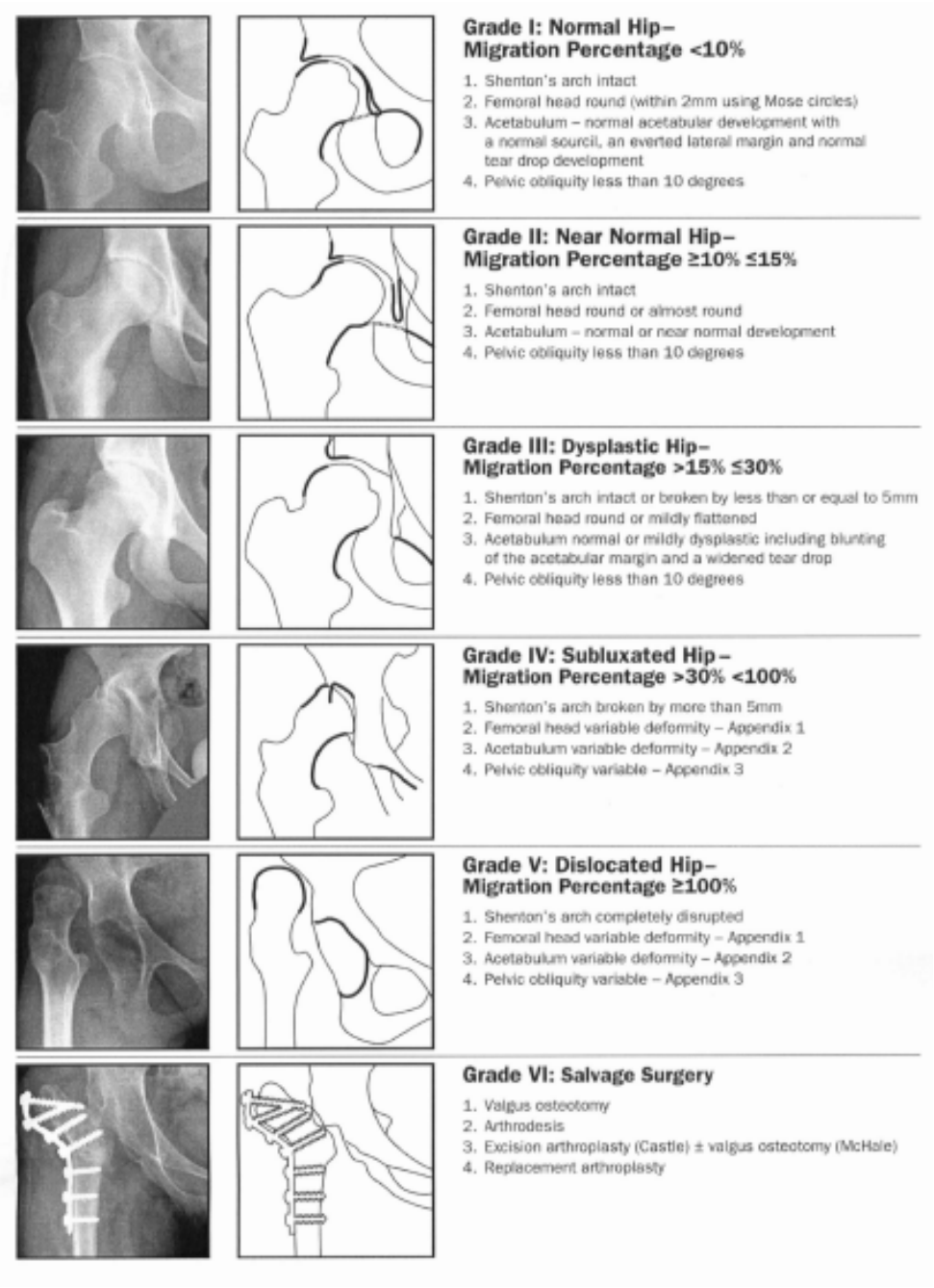
Reconstructive Surgery for Hip Displacement

Reconstructive surgery refers to various combinations of adductor releases when combined with bony reconstructive procedures such as femoral osteotomy (varus derotation osteotomy of the femur) and pelvic osteotomy (Para-Ilial, Dega or San Diego osteotomies). In recent years there has been a move towards "one step hip reconstruction" in which adductor releases, femoral varus derotation osteotomy and pelvic osteotomy are combined in one sitting. Several centres have reported promising early results from such approaches[15,16,17]. However the outcome measures in most of these studies have been in the impairment domain mainly focusing on radiographic measures such as migration percentage before and after surgery.

There has been limited information on range of motion, function, or quality of life. In this regard it should be noted that a validated quality of life tool for children at GMFCS level IV and V has been developed by Nararayan and colleagues[22] and may well be the ideal tool in studies in this complex area.

In addition there has been no agreed system for classifying the morphology of the hip joint in broad categorical terms in adolescents with cerebral palsy. Robin and colleagues have recently published a cerebral palsy hip classification which may be useful in both natural history and outcome studies[23]. (Figure 1)

Figure 1 Classification of hip subluxation/dislocation



GAIT CORRECTION SURGERY

The majority of surgical procedures in the lower limbs in children with cerebral palsy are designed to improve gait and function. Lower limb surgery can be classified as single level, multilevel, unilateral or bilateral. The number of procedures is very extensive and the possible combinations almost infinite. For the purposes of this review it was felt appropriate to concentrate on two key areas:

1. Surgery for equinus deformity
2. Single event multilevel surgery (SEMLS)

Equinus deformity is the most common deformity in children with cerebral palsy and surgery for equinus was not only the first orthopaedic operation recorded but has the longest continuous history in medical history, approximately 200 years. In addition, surgery for equinus deformity is the most common component of single event multilevel surgery.

Single Event Multilevel Surgery (SEMLS)

The concept of multiple operations on both lower limbs in one surgical session seems to have developed simultaneously in both North America and Europe about 20 years ago. One of the clearest descriptions of the concept was by Rang who described single level surgery repeated on multiple occasions as the birthday syndrome and advocated correction of all deformities simultaneously[3]. However one of the first reports was by Norlan and Tkaczuk in 1985[24]. They reported commencing their multilevel surgery program in 1975 and reported the outcome of multilevel surgery in 40 children claiming an 80% success rate. As would be expected from a report from that era, they classified the children as having spastic diplegia, spastic tetraplegia or spastic hemiplegia. In addition, the emphasis in the surgical program was on lengthening contracted flexor muscles including the hip flexors, the knee flexors, and the ankle plantar flexors. Additional procedures included adductor releases, transfer of tibialis posterior, subtalar arthrodeses and plantar fasciotomies. However the study also included children who could not walk, requiring obturator neurectomies and femoral osteotomies. Again as typical of studies from this era, the principal outcome measures were the rate of complications, recurrence rates and the need for supplementary osteotomies with almost no objective data on functional outcomes.

The first report utilising objective outcome measures was that by Nene and colleagues in 1993[25]. They reported on the outcome of simultaneous multiple operations in 18 children with spastic diplegia utilising the Physiological Cost Index (PCI) of walking. They reported that 14 patients had a measurable reduction at one year and that the more severely affected patients took up to two years to reach a new functional plateau. Again the surgical program was essentially muscle tendon lengthenings including intramuscular psoas tenotomy, fractional lengthening of the hamstrings and transfer of the rectus femoris. Significantly fewer gastrocsoleus lengthening procedures were performed and they also employed bony reconstructive surgery including femoral osteotomies, tibial osteotomies and stabilisation of the foot by either subtalar arthrodesis or triple arthrodesis.

In 1984 Gage and colleagues reported on the value of pre and postoperative gait analysis in patients with spastic diplegia who underwent multilevel surgery. Although the emphasis in the paper was on the use of computerised gait analysis, this may be the first report on the outcome of multilevel surgery in children with cerebral palsy, certainly the first with objective outcome measures[26]. In 1987 Brown and McManus reported on the outcome of 57 children with spastic diplegia who had a bilateral combined adductor and proximal hamstring lengthening which in some cases was combined with a lengthening of the Achilles tendon. They reported changes in hip and knee range of motion. In addition they attempted a functional classification (non-ambulant, ambulant plus aids, independent) which showed that a significant improvement in ambulatory status was recorded in many children after surgery[27].

With the increased use of three dimensional gait analysis, there is more objective information on the natural history of gait changes in children with cerebral palsy have

been published form several centres. Three studies which utilised motion analysis all confirmed broadly similar findings. These include deterioration in gait velocity and temporospatial characteristics, increased dorsiflexion at the ankle with concomitant increased flexion at the hip and knee as well as increased stiffness[28,29,30].

The more recent literature in multilevel surgery follows two main trends. Firstly there has been greater emphasis on detailed analysis using instrumented gait analysis as typified by Saraph and colleagues in 2002[31]. They reported on the outcome of 25 ambulatory children with diplegic cerebral palsy who had a mean of 8.2 lower limb procedures. They reported on physical examination findings, kinematic and kinetic data in the sagittal plane before and at least three years after surgery. They emphasised the need to integrate information from clinical, kinematic and kinetic data. This concept was taken further by Davids and colleagues[32] who promoted the idea of a diagnostic matrix for the comprehensive evaluation of children before and after multilevel surgery.

The second trend has been the promotion of a broader assessment of children with cerebral palsy according to the National Centre for Medical Rehabilitation Research NCMRR and World Health Organization International Classification of Functioning Models. Oeffinger and colleagues reported a large multicentre study in which the relationships between Gross Motor Function Classification Systems (GMFCS), Gross Motor Function Measure (GMFM), Pediatric Orthopaedic Data Collection Instrument (PODCI) and instrumented gait analysis were reported. GMFM sections D and E scores correlated with the largest number of the other tools and GMFM section E score can be used to predict GMFCS level with a high degree of accuracy. They concluded that their study established justification for using the GMFCS as a classification system in clinical studies. The relationships between functional outcome measures for assessing children with ambulatory CP were further explored in a large study by Sullivan and colleagues with important conclusions for the design of future studies[33]. In a very large multicentre study, Oeffinger and colleagues[34] and Bagley and colleagues[35] reported on outcome assessments in children with cerebral palsy at GMFCS levels I-III as well as the discriminatory ability of outcome tools. The information from these studies is crucial in the design of balanced outcome studies in the future[33-36].

Despite studies reporting the outcome of multilevel surgery with emphases on various aspects such as the changes in rehabilitation during the first year or the duration of benefit, the majority of studies continue to be retrospective and uncontrolled[37-41]. To date the only control study is by Zwick and colleagues in 2001 but the subjects were not randomised[42]. Preliminary results of the first randomised control trial of SEMLS has recently been reported by Thomason and colleagues[6].

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SURGICAL CORRECTION OF EQUINUS DEFORMITY IN CHILDREN WITH CEREBRAL PALSY: A SYSTEMATIC REVIEW

Nathan White MB BS (Hons), B.Physio (Hons)
Benjamin Shore MD, FRCSC
H. Kerr Graham MD, FRCS (Ed), FRACS
The Royal Children's Hospital, Melbourne, Australia

Equinus is the most common deformity in cerebral palsy and may be defined as the inability to dorsiflex the foot above plantigrade, with the hindfoot in neutral and the knee extended. However there are many other definitions for both equinus deformity and equinus gait which contribute to the difficulty in interpreting the extensive literature on the subject.

Apart from the large number of original papers, there are a number of review articles of which two are noteworthy. In 2001, Goldstein and Harper published an annotation in Developmental Medicine and Child Neurology entitled "Management of Cerebral Palsy: Equinus Gait"[1]. This was the report of a multidisciplinary workshop convened to explore the current state of knowledge, best clinical practice and research needs for the management of equinus gait associated with cerebral palsy (CP). This review provides a scholarly update on many aspects of equinus gait and was greatly strengthened by the multidisciplinary approach as well as an analysis of the evidence base. The analysis compared the evidence base for physical therapy, orthoses, casting, Botulinum toxin A and surgery across the domains of treatment aims which had been suggested by Condie and reported by Stuberg[2]. Condie and colleagues suggested that the aims of management of equinus in cerebral palsy were the prevention of deformity, correction of deformity, the promotion of a base of support, the facilitation of training skills and an improvement in the efficiency of gait. When these aims of treatment were considered in the context of the evidence base, Botulinum toxin A was noted to have level 1or level 2 grading whereas surgery had consistently level 4 and 5 grading.

In a more recent and more traditional review of surgical studies, Koman and colleagues reviewed recurrence of equinus deformity in cerebral palsy patients following surgery, in 2003[3]. This is a very useful review of 31 published articles from 1938 to 2001 and focused mainly on the issue of recurrent equinus deformity. The conclusions of the review were that age at first surgery is the number one predictor of recurrent equinus deformity. For this reason temporising measures such as the use of Botulinum toxin A, casting and orthoses should be considered in younger children to postpone the age at first surgery for as long as possible.

There is a very extensive literature on the management of equinus deformity in cerebral palsy by both operative and non-operative measures. However despite the large volume of published studies, the review by Koman and colleagues[3] and the annotation by Goldstein and Harper[1] both found very poor levels of evidence to support surgical intervention for equinus deformity. The complexity and evolving nature of cerebral palsy in children, coupled with the difficulties of designing and conducting randomised clinical trials, have led to a paucity of robust evidence to guide surgical practice. This study was undertaken to examine the current evidence base for surgical management of equinus deformity in cerebral palsy.

Central to discussion of the surgical management of equinus is the issue of recurrent deformity and the need for additional surgery as well as the opposite outcome, over-correction, calcaneus deformity and crouch gait. However detection of these dichotomous outcomes requires studies of adequate power, appropriate duration and sensitive, appropriate and objective outcome measures. In addition, the child and their cerebral palsy type was considered to be of great importance. Cerebral palsy can be classified in the domains of movement disorder, topographical distribution and gross motor function[4]. Single-event multilevel gait improvement surgery (SEMLS) has become the standard of care in many institutions for the treatment of walking children with CP[5,6,7,12]. More recently, equinus surgery was preformed in the setting of multilevel surgery and these articles were considered in this review. Other factors to be considered in the review, having been suggested in previous studies to be important include the surgical procedure, the postoperative care, the use of orthoses and night splinting.

METHOD

Search strategy

In March 2008, an electronic search was performed of MEDLINE (1966-March 2008), Embase (1980-2006), and the Cochrane database. Keywords used in the search strategy included 'cerebral palsy', 'equinus', 'ankle', 'achilles', 'gastrocnemius', 'gait analysis', 'contracture' and 'calf'. Key terms were matched to Medical Subject Headings (MeSH) index, and exploded or searched as keywords. Surgical procedures identified as being used in the correction of equinus deformity were also searched as keywords. Targeted searching was undertaken from reference lists of the articles included for review, and other key articles concerning the topic.

Inclusion and exclusion criteria

Studies were shortlisted for inclusion if they pertained to the surgical management of equinus in a paediatric population with cerebral palsy, and were available in English. Where doubt existed, the article was shortlisted. Additional inclusion and exclusion criteria were then applied to shortlisted articles. Articles were excluded where they examined a novel surgical intervention or had fewer than ten cases followed up. Articles included for review had a mean follow up period of greater than five years, or used instrumented gait analysis, with a mean follow up of greater than 12 months.

Data extraction

Included articles were examined in depth and key data extracted with the use of a custom data extraction form. This form focused on themes of sample characteristics, surgical intervention, outcome measures, data handling, follow up, and complication rates. Oxford Centre for Evidence Based Medicine levels of evidence[8] were used to grade the selected studies. In surgical research, randomization is not always possible or feasible[9] and in this review all included articles were non-randomized**.** As a result, the methodological index for non-randomized studies (MINORS)[10] was applied to further assess the quality of each article.

RESULTS

Search strategy yield

The initial electronic search identified 6905 articles for consideration, from which a short list was generated. From this, two authors identified 49 possible inclusions for review. These were examined in depth. After applying inclusion and exclusion criteria, 35 studies were included. Table **O**ne lists the articles included and their characteristics.

Descriptive aspects of reviewed studies

The studies which were reviewed ranged in sample size from 9 subjects[30] to 156 subjects[29] with an average of 38. It should be noted that in some papers, the number of "lower limbs" is reported rather than the number of subjects[14]. Given that hemiplegic children are managed by unilateral surgery and diplegic children can be managed by either unilateral or bilateral surgery, this can be very confusing and is another mark of the low quality of many of the reports. The mean age of subjects ranged from 4.8 to 19 years with many studies reporting a wide age range at index surgery. The majority of studies reported the gender of the patients but many did not include this potentially important information.

The type of cerebral palsy was in general poorly described. Very few studies reported the type of movement disorder, not all included such basic information as the topographical distribution and only f**ive** referred to gross motor function[11,19,22,26,27]. Studies which gave a breakdown of either movement disorder or topographical distribution, frequently utilised out of date classifications.

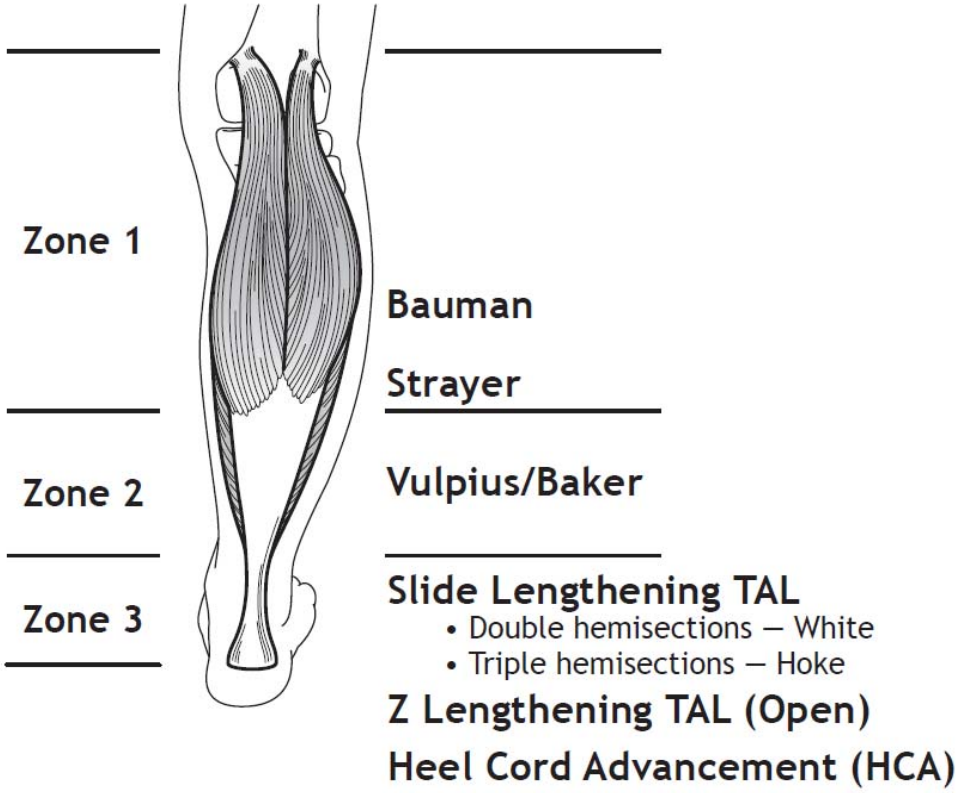
Outcome measures used included goniometry, observational gait analysis or rating scales, instrumented gait analysis, surgery for recurrence, bracing for calcaneal gait pattern, measures of spasticity, physician rating scale and parental satisfaction. Such measures were commonly used to derive an outcome scale rating the overall result from good to poor. The duration of mean follow up varied considerably, from 1 to 14 years. For the purposes of this review, studies with less than five years follow up were only considered if they used instrumented gait analysis. Nineteen studies used instrumented gait analysis, with an average follow up of 2.8 years.

Operative Intervention

The surgical intervention was not always clearly identified by either a generic term or an eponym. Some studies reported on a single clearly described operative procedure such as the White slide tendo Achilles lengthening[23]. Others compared multiple procedures and in these studies there was a tendency to refer to the original operative description[15]. Some articles specified modifications of earlier procedures[29]. Finally, some studies reported on combinations of surgical procedures for equinus for example the Strayer distal gastrocnemius recession combined with the slide lengthening of the Achilles tendon[16]. For clarification and consensus an 'intramuscular lengthening' was interpreted as a Baumann procedure, a 'gastrocnemius recession' was interpreted as a Strayer procedure and a 'gastrosoleus aponeurotic lengthening' was interpreted as a Vulpius procedure (Figure 1).
From proximal to distal, the procedures which were included in the 35 selected articles were the Baumann procedure, the Strayer distal gastrocnemius recession, the

Baker and Vulpius gastrocsoleus lengthening, the White and Hoke slide tendo Achilles lengthenings (percutaneous and open), open Z tendo Achilles lengthening (TAL) and heel cord advancement (HCA). The operative procedures are illustrated in relation to three specific zones of the triceps surae in Figure 1. Zone 1 is from the gastrocnemius origin and ends at the most distal fibres of the medial belly of the gastrocnemius. The operative procedures in Zone 1 are the Baumann procedure and the Strayer distal gastrocnemius recession. Zone 2 is from the distal gastrocnemius belly to the end of the soleus muscle fibres and includes the Baker and Vulpius gastrocsoleus aponeurotic lengthenings. Zone 3 is the Achilles tendon and includes all forms of lengthening of the Achilles tendon as well as heel cord advancement (HCA).

Figure 1 Operative procedures categorised in three zones of the triceps surae



Postoperative Care

The immediate postoperative care was reasonably well described in most studies. The older literature reported above knee casts for at least six weeks with long periods of restricted weight bearing[14]. The more recent literature reports the use of below knee casts and much earlier weight bearing[11,13,18,19,22,25,27,28,37]. The utilisation of ankle-foot-orthoses after cast removal, the type of ankle-foot-orthosis and the utilisation of night splints varied greatly and were poorly described.

The outcome measures reported were in order of frequency, clinical examination, clinical rating scales, gait rating scales, goniometry, three dimensional gait analysis, the Modified Ashworth Scale, radiographs, the Gross Motor Function Measure (GMFM) and the need for floor reaction AFOs to manage crouch gait. Many studies

reported rates for recurrent equinus deformity or calcaneus gait/deformity. However the definitions for both equinus/calcaneus gait and equinus/calcaneus deformity varied from study to study and were not always clearly defined.

Effect of age at surgery on rates of recurrent equinus and calcaneus

The effect of age at index surgery on the rates of recurrent equinus or calcaneus were reported in 10 of the 35 studies[15,16,17,23,29,33,35,38,39,42]. Damron *et al*[17] found no clear relationship between age and outcome. Seven studies reported that a younger age at index surgery was associated with an increased risk of recurrent equinus [15,16,23,29,33,39,42] but one study found that age was not related to recurrent equinus[35]. One study[38] utilising gait analysis found that velocity and stride length improved most in younger patients. Twenty five of the 35 studies made no comment on the effect of age on either the rate of recurrent equinus or calcaneus.

Recurrent equinus deformity and type of surgery

Twelve of the 35 studies stated explicit criteria for the definition of equinus deformity and/or recurrent equinus[13,15,16,17,23,25,26,33,35,38,39,44]. Definitions for equinus deformity were either based on clinical examination and/or sagittal ankle kinematic data. In an attempt to increase the relevance of this heterogenous body of evidence, results were grouped according to the surgical zone as illustrated in Figure 1. Where results by sub-group were not specifically stated and could not be confidently derived from presented data, they are indicated to be not available. Recurrence rates varied from 0-43% overall but this increased up to 62% when specific sub-groups were considered. The majority of studies reporting three dimensional gait analysis had short term follow-up and reported much lower rates for recurrent equinus deformity than the clinical studies, which all included a mean follow-up of at least five years[26,28,30,36,40,44].

The rate of recurrent equinus deformity for procedures involving lengthening of the Achilles tendon were given in 13 studies[14,15,17,23,25,26,28,32,33,35,39,41] (Table 2).

Seven studies reported on the rates of recurrent equinus deformity following surgical lengthening in Zone 2 (the Baker and Vulpius procedures) [11,15,17,24,29,41,43] (Table 3).

Eleven studies reported a Zone 1 procedure, three Baumann and eight Strayers. Six studies reported on the rates of recurrent equinus following surgery [16, 22, 25, 26, 36, 39] (Table 4). No recurrent equinus was found at a mean follow-up of just over two years[36]. The only other Zone 1 procedure, the Strayer distal gastrocnemius recession was reported in combination with Achilles tendon lengthening or in sub-groups of studies reporting multiple surgical procedures.

Outcome and topographical distribution of cerebral palsy

Thirteen studies reported on the rate of recurrent equinus deformity in children with spastic diplegia[11,15,22,24,26,28,29,33,35,36,39,42,43.] Additionally the study by Kay et al, whilst not providing an overall recurrence rate in spastic diplegia, reported no significant difference in recurrence rates when children with diplegia who had gastrocnemius recession were compared to those who had tendo Achilles lengthening (Table 5).

Ten studies reported on the rates of recurrent equinus deformity in children with hemiplegia[15,23,24,26,29,33,35,39,42,43] (Table 6). Again, Kay et al[25] reported no difference in

the rate of recurrent equinus deformity in children with hemiplegic cerebral palsy who had either gastrocnemius recession or lengthening of the Achilles tendon.

Calcaneus deformity, calcaneus gait and crouch gait by topographical distribution

Nine of 35 studies reported explicit criteria for calcaneal gait, calcaneal deformity, or crouch gait[15,17,18,20,25,26,36,38,44]. The definitions included the inability to passively plantarflex the foot to neutral as a definition of calcaneus deformity and the need for a floor reaction orthosis for crouch gait. In terms of kinematic criteria, peak dorsiflexion in stance more than 2 standard deviations above the reference range was quoted in several studies and in kinetic terms, reduced push off or A_2 power generation was also quoted. Using these variable terms, the rates of calcaneus varied from 0 to 36% overall but up to 80% in specific sub-groups.

Thirteen studies reported the rate of calcaneus for operative procedures in Zone 3[15,17,18,23,25,28,26,32,33,35,39,41,42]. One study reported on the development of spastic flat foot and knee flexion deformity as postoperative complications[42] (Table 2).

Nine studies reported the rate of calcaneus following surgical procedures in Zone 2 [11,13,15,17,24,29,38,41,43] (Table 3). Five studies reported the rate of calacaneus following Zone 1 surgical procedures[16,25,26,36,39] (Table 4). The report by Saraph et al[36] on the Baumann procedure reported no calcaneus in 22 children and adolescents with spastic diplegia who had multilevel surgery including a Baumann gastrocsoleus lengthening.

Twelve studies reported the rates of calcaneal gait in diplegic children [15,17,18,19,24,28,29,33,36,38,43,44]. Kay et al[25] reported no significant difference in the rates of calcaneus gait in children with spastic diplegia who had either a gastrocnemius recession or an Achilles tendon lengthening (Table 4).

Thirteen studies reported the rates of calcaneus gait in hemiplegic children [11,13,15,17,18,24,28,29,33,36,38,39,43] Again, Kay et al[25] found no significant difference in calcaneus gait in hemiplegic children who had either a gastrocnemius recession or a lengthening of the Achilles tendon (Table 5).

The majority of studies were scored at level 4 evidence according to the Oxford classification for evidence based medicine (Table 1). Only 4 studies met the criteria for 3b level of evidence, demonstrating a comparable control group for comparison. Therefore, the MINORS index was applied to each study. In this review, MINORS scores ranged from 3 to 16 demonstrating a significant amount of methodological heterogeneity and quality within the studies.

Fifteen studies recorded results of equinus surgery in the setting of SEMLS (Table 6). On average, seven surgical procedures were preformed on each patient including calf lengthening. Overall trends demonstrated and increase in ankle dorsiflexion through initial contact and a decrease in the degree of knee flexion at initial contact (Table 6).

DISCUSSION

Cerebral palsy type and outcome

Given that the rate of calcaneus in children with spastic hemiplegia averaged only 1% (range 0-7%) and the rate of calcaneus in spastic diplegia averaged 15% (ranging from 0-41%) it would appear that the topographical distribution of cerebral palsy may well be the strongest determinant of outcome. It is also of note that several of the studies utilising instrumented gait analysis reported high rates of calcaneus gait and the clinical studies with the longest to follow-up also reported the highest rate of calcaneus gait. The outcomes in terms of recurrence versus calcaneus in children with hemiplegia versus children with diplegia are so different as to merit separate studies.

There is very little information on the movement disorder. It is very likely that a number of children with dystonia or mixed movement disorders have been included in these studies but have not been clearly identified. The outcome of surgery including surgery for equinus deformity, in children with dystonia and mixed movement disorders is usually considered to be more unpredictable but no information from the studies which have been reviewed is available.

The choice of studies for this review in two main groups, those with a mean follow-up of more than five years and those with instrumented gait analysis, give two quite different perspectives on both recurrent equinus deformity and calcaneus gait.

Gait analysis is expensive and the majority of studies report a preoperative and a single postoperative study usually within 1-2 years after surgery. Although the kinematic parameters are objective and sensitive in the definition of under-correction and over-correction, the brevity of follow-up, dramatically weakens the value of these studies. The one study which combines instrumented gait analysis and a medium term follow-up is that by Segal et al[38]. Segal and colleagues reported a 30% prevalence of calcaneal gait but there were only 20 children in their study and there were five different combinations of surgical procedures[38].

The two clinically based outcome studies with the longest follow-up and the largest number of cases of children with diplegia were the studies by Borton et al[15] and Dietz et al[18]. Borton and colleagues reported risk factors for both recurrent equinus and calcaneus in a large heterogenous population of children with cerebral palsy who were followed clinically for an average of seven years (range 5-10 years). They found a very high incidence of calcaneus gait leading to crouch gait in both spastic diplegia and spastic quadriplegia especially after percutaneous lengthening of the Achilles tendons. Dietz and colleagues confirmed these findings but added an objective measure of crouch gait, the need to prescribe a floor reaction brace[18]. Their study reports a single surgeon, case series with a highly standardised operative protocol, follow-up programme and consistency in the diagnosis and management of crouch gait. Again, results were best in patients with either hemiplegia or subjects who required lengthening on only one side. They recommended consideration of non-surgical treatment or more conservative surgical treatment such as gastrocnemius fascial lengthening[18].

There is a clear consensus from the studies that the type of cerebral palsy matters a great deal and should be carefully considered in the choice of surgical procedure and overall management. Only four studies reported Gross Motor Function Classification

System (GMFCS) levels[11]. In children with bilateral cerebral palsy, traditionally described as having spastic diplegia or spastic quadriplegia, the use of the GMFCS might well prove to be very valuable. It is possible that outcomes might vary considerably across GMFCS levels and this would be worthy of examination in future studies.

Effect of age at index surgery on rates of recurrent equinus deformity and calcaneus gait

There was widespread agreement in the studies reviewed that early surgery was a major risk factor for recurrent equinus deformity and possibly for unpredictable outcomes in general[14,15,18,29,39,44]. This led several authors to recommend delaying age at index surgery by the use of non-operative measures such as injection of Botulinum toxin A, serial casting and the use of orthoses[1,3,15,18]. No study to date has reported on the outcome of sequential non-operative management of equinus using Botulinum toxin injections followed by surgical lengthening at appropriate age. However several studies on the use of Botulinum toxin A have reported successful short term outcomes and the ability to delay surgery until age more than six years in the majority of children. A more widespread application of this policy might well improve surgical outcomes by reducing the risk of recurrent equinus and perhaps calcaneus.

Effect of surgical procedure on the rate of recurrent equinus deformity and calcaneus gait

A total of 10 different operative procedures or combinations were reported in the articles in this review. Some studies focused on a single operative procedure[23,29,36], others on comparison of multiple procedures[15,32,39] , and some in the setting of SEMLS [11,12,18,21,22,25,26,28,34,36,37,40,44]. The reason for choice of various procedures was often surgeon preference or frequently not clearly stated. The operative procedures range from very proximal procedures in Zone 1 of the gastrocsoleus to very distal procedures on the Achilles tendon. Despite a great emphasis on the importance of the differences in the surgical techniques, the evidence for difference in outcomes is weaker for surgical procedure than for cerebral palsy type or age at index surgery. In particular, several studies utilising three dimensional gait analysis found little difference between Zone 2 surgical procedures (Baker and Vulpius) and surgical lengthening of the Achilles tendon. Within the Achilles tendon, a wide range of procedures was utilised from open Z lengthening to percutaneous slide lengthening. There were no studies in which children were randomised to receive one surgical procedure versus another and the conclusions reported in most studies are questionable and highly susceptible to selection bias. Despite the paucity of evidence, opinions on the appropriate choice of surgical procedure are highly polarised. The lack of equipoise means that the design of randomised trials in which different procedures are compared is contentious and difficult.

Only one of the included studies was prospectively designed and no studies were randomised with a blinded control group. The majority of studies were Level 4 quality of evidence according to the Oxford Centre for Evidence Based Medicine Levels. Therefore, these results at best could lead to only a Grade C treatment recommendation.

Given the well recognised difficulties of designing, recruiting, funding and implementing randomised surgical trials consideration should be given to designing

better cohort studies. Improvements in study design might include improved definition of equinus gait and associated functional problems using a combination of instrumented gait analysis, functional testing and quality of life measures. Surgical populations should be described much more rigorously in terms of cerebral palsy type (movement disorder, topographical distribution and GMFCS level). The surgery for equinus deformity should be clearly described avoiding eponymous terms. The need for a clear description of postoperative management, bracing and night splinting is also obvious. In addition, for children with spastic diplegia it is essential to fully describe all additional procedures in multilevel surgical protocols.

Prospective studies with balanced outcome measures at prescribed intervals would greatly improve the evidence base for equinus surgery. A comparison of outcomes at baseline, 12 months and 5 years after surgery with appropriate outcome measures would be a major advance on the current poor evidence base.

CONCLUSION

Equinus deformity can be primary, secondary, either fixed or functional, and both impacts upon and is affected by the motor development of the child in question. A systematic review revealed the literature to be dominated by retrospective case series studies with no control groups, and a lack of prospective randomised controlled trials. The current literature is particularly weakened by the frequent treatment of patient groups with differing motor distributions as a homogenous population, and inadequate follow up to detect true complication rates. There is insufficient evidence to state clearly a preference for any single procedure over another. However, the literature indicates greater recurrence in children with hemiplegia regardless of procedure, and greater calcaneus in children with diplegia, particularly following procedures to the Achilles tendon.

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TABLE 1 35 Articles Fulfilling Criteria for Systematic Review

Author(s)	Surgical Procedures	SEMLS	No of subjects	Age (years) Mean (range)	Gender M F	CP Type H,D,Q	Outcome Measures	Follow-up (yrs/range)	OX LOE	MINORS Score
[11]Abel et al (1999)	Vulpius	Yes	30	8.7 (4-20)	-	30 D	3DGA, GMFM Goniometry	2 (1.6-3.1)	4	10
[12]Adolfsen et al (2007)	Vulpius, TAL(NS)	Yes	31	8.5 (5-15)	-	10H, 20D, 1Q	3DGA, EMG, Clinical exam	1.9 (0.7-6.4)	4	6
[13]Baddar et al (2002)	Vulpius	No	34	7.2 (2.6-15.2)	17M 17F	34 D	3DGA/EMG Clinical exam	1.3 (0.8-1.9)	4	8
[14]Banks (1976)	White TAL	No	200 feet	-	-	-	OGA	10 (-)	4	3
[15]Borton et al (2001)	Hoke TAL(Perc) Z TAL, Baker	No	134	7.6 (2-18)	78M 56F	45H, 65D, 24Q	3DGA, PRS Goniometry	6.9 (5-10)	4	11
[16]Craig & Van Vuren (1976)	Strayer ± White TAL	No	59	6 (1-15)	-	-	Clinical exam	6 (-)	4	8
[17]Damron et al (1994)	Z TAL,Baker, Vulpius	No	59	4.8 (2-11)	31M 28F	2H, 18D, 28Q	Goniometry Functional scale	7 (-)	3b	9 (14)
[18]Dietz et al (2006)	Hoke TAL(O), Z TAL	Yes	79	7.7 (2-47)	-	23H, 34D, 15Q	OGA GRAFO	7 (-)	4	11
[19]Engsberg et al (2005)	HCA, Vulpius, White TAL	No	32	7 (-)	19M 13F	32D, (GMFCS I)	Goniometry 3DGA, GMFM	* 1 (-)	4	11
[20]Fabry et al (1999)	TAL(O)(NS), TAL (P)(NS)	No	15	15.7	8M 7F	15D	2DGA Clinical exam	9.5	4	4
[21]Gough et al (2004)	Strayer	Yes	12	9.8	8M 4F	12D	3DGA, EMG Clinical exam Functional status	1.5 (-)	3b	9 (15)
[22]Gough et al (2008)	Strayer	Yes	13	6.4 (5.9-6.9)	-	13D	3DGA, GGI, EMG Clinical exam	2 (2-5)	3b	10 (16)
[23]Graham & Fixsen (1988)	White TAL	No	35	6.9 (3-15)	18M 17F	35H	Goniometry VRS	13 (9-16)	4	9
[24]Javors & Klaaren (1987)	Vulpius	No	47	6.8 (2-14)	-	15H, 23D, 9Q	Clinical exam	5.7 (1-12)	4	3
[25]Kay et al (2004)	Strayer, TAL(NS)	Yes	54	9.8 (4-16)	28M 27F	23H, 26D, 6Q	3DGA, PRS, Ashwoth	1.5 (1-2)	4	8
[26]Lofterod & Terjesen (2008)	Strayer, Z TAL	No	15	8.8 (6-14)	8M 7F	6H, 9D (GMFCS I + II)	3DGA Clinical Exam	* 3 (1.1-4.6)	4	6
[27]Lofterod & Terjesen (2008)	Strayer	Yes	47	11.5 (5-19)	29/18	13H 34D (GMFCS I+II+III)	3DGA exam GMFCS FMS	* 1.2 (.8-1.5)	4	8
[28]Lyon et al (2005)	Z TAL, Hoke TAL	Yes	14	9.1 (4-17)	6M 8F	14D	3DGA	* 1.5 (0.5-2.5)	4	6
[29]Olney et al (1988)	Baker	No	156	5.5 (2-14)	-	90H, 108D, 21Q	Clinical exam Recurrence	7.5 (3-14)	4	9

TABLE 1 35 Articles Fulfilling Criteria for Systematic Review (continued)

Author(s)	Surgical Procedures	SEMLS	No of subjects	Age (years) Mean (range)	Gender M F	CP Type H,D,Q	Outcome Measures	Follow-up (yrs/range)	OX LOE	MINORS Score
[30]Orendurff et al (2002)	TAL (NS)	No	9	-	-	-	3DGA Muscle length	* 1 (-)	4	5
[31]Park et al (2006)	Vulpius, Z TAL(NS)	No	16	8 (3-16)	11M 5F	16H	3DGA	* 1.25 (0.75 – 2)	4	5
[32]Pierrot & Murphy (1974)	HCA, White TAL, Z TAL	No	41	7 (3-10)	23M 18F	16H, 9D, 13Q	VRS	6 (3-10)	4	9
[33]Rattey et al (1993)	Z TAL	No	57	5.4 (1.4-14.7)	-	27H, 30D	Clinical exam Recurrence	9.6 (5-15)	4	9
[34]Rose et al (1993)	Baker	Yes	20	6 (4-26)	-	5H, 15D	Clinical exam 3DGA	* 1 (-)	4	7
[35]Sala et al (1997)	Hoke TAL	No	27	5 (2-10)	15M 12F	4H, 17D, 6Q	Clinical exam VRS, MAS	(2-9)	4	9
[36]Saraph et al (2000)	Baumann	Yes	22	12.6 (7.4-16.6)	-	22D	Clinical exam 3DGA	* 2 (2.1-4)	4	5
[37]Saraph et al (2005)	Baumann	Yes	32	11.1 (8.7-13.5)	-	32D	Clincal exam, 3DGA, EMG	* 4.4(1.0-5.4)	4	9
[38]Segal et al (1989)	Hoke TAL(P), White TAL, Z TAL, HCA, Vulpius	Yes	20	5.2 (2.7-8.2)	9M 11F	20D	Clinical exam 3DGA	5.8 (1.1-11)	4	11
[39]Sharrard & Bernstein (1972)	Z TAL, Strayer	No	92	7 (-)3	-	45H, 20D, 27Q	Clinical exam OGA	8.9 (-)	4	7
[40]Steinwender et al (2001)	Baumann	Yes	29	14.4 (-)	-	17D	Clinical exam 3DGA	* 3.9 (-)	3b	5 (13)
[41]Truscelli et al (1979)	TAL(NS), Vulpius	No	58	-	-	-	Clinical exam	14 (2-27)	4	4
[42]Uyttendale et al (1984)	Z TAL, Scholder	No	32	19 (14-30)	-	13H, 19D/Q	Clinical exam OGA	-	4	5
[43]Yoshimoto et al (2005)	HCA ± Vulpius	No	17	10 (5-17)	9M 8F	11H, 6D	Clinical exam OGA, Xray	8 (5.7-9.11)	4	11
[44]Yngve & Chambers (1996)	Z TAL, Vulpius	Yes	33	9 (-)	-	14H, 17D, 2Q	Clinical exam 3DGA	* 1 (-)	4	8
[45]Zwick et al (2001)	Strayer	Yes	17	11.2 (5.7-16.4)	-	17D	Clinical exam 3DGA	* 3.8 (2.6-5.7)	4	9

OX LOE – Oxford Levels of Evidence **HCA** – Heel Cord Advancement **G/S –** Gastrosoleus **3DGA**- 3D Gait Analysis
VRS – Visual Rating Scale **MAS** – Modified Ashworth Scale **OGA** – Observ

TABLE 2: Zone 3: Achilles Tendon Lengthening: Recurrent Equinus and Calcaneus

Author	Procedure	Feet	Hemis	Dis	Quads	Recurrent equinus %	Calcaneus %	F/up <5 yrs	3DGA
[12]Adolfsen et al	TAL (NS)	6	-	-	-	-	-		x
[14]Banks	White TAL	200	-	-	-	27%	-		
[15]Borton et al	Z TAL Hoke TAL (P)	40 98	-	-	-	23% 20%	35% 39%		x
[17]Damron et al	Z TAL	80	-	-	-	2%	2%		
[18]Dietz et al	Hoke TAL (O) Z TAL	56 56	-	-	-	- -	43% 53%		
[19]Engsberg et al	White TAL	10	0	10	0	-	-	x	x
[20]Fabry et al	TAL (O)(NS) TAL (P)(NS)	11 6	0	15	0	-	-	x	x
[23]Graham & Fixsen	White TAL	35	35	0	0	43%	0		
[25]Kay et al	TAL (NS)	17	11	4	2	29%	36%	x	x
[26]Lofterod et al JCP	Z TAL	10	2	8	0	0%	0 %		x
[28]Lyon et al	Z TAL Hoke TAL	21 2	0	12 2	0	35%	Most *	x	x
[30]Orendurff et al	TAL (NS)	12	-	-	-	-	-	x	x
[31]Park et al	Z TAL (NS)	13	13	0	0	-	-	x	x
[32]Pierrot & Murphy	White TAL Z TAL	9 13	7	4	5	46%	0%		
[33]Rattey et al	Z TAL	77	29	34	0	26%	5%		
[35]Sala et al	Hoke TAL	27	4	17	6	22%	-		
[38]Segal et al	Hoke TAL (P) White TAL Z TAL	12 6 12	0	6 3 7	0	-	25% 25% 25%	x	x
[39]Sharrard & Bernstein	Z TAL	77	37	12	24	23%	0%		
[41]Truscelli et al	TAL (NS)	72	-	-	-	35%	3%		
[42]Uyttendaele et al	Z TAL	25	10	15	0	20%	-		
[44]Yngve & Chambers	Z TAL	27	11	16	0	-	-	x	x

* avg df in stance 24.1 x = <5 years average follow up

TABLE 3: Zone 2: Mid Calf Gastrocsoleus Recession

Author	Procedure	Feet	Hemis	Dis	Quads	Recurrent equinus %	Calcaneus %	F/up <5 yrs	Gait analysis
[11]Abel et al	Vulpius	42	0	21	0	0%	0%		x
[12]Adolfsen	Vulpius	33	-	-	-	-	-		x
[13]Baddar	Vulpius	22	0	11	0	-	0%	x	x
[15]Borton et al	Baker	57	-	-	-	26%	32%		x
[17]Damron et al	Baker, Vulpius	38	-	-	-	12%	0%		
[19]Engsberg et al	Vulpius	10	0	10	0	-	-	x	x
[24]Javors & Klaaren	Vulpius	79	15	46	18	4%	4%		
[29]Olney et al	Baker	219	80	108	21	48%	0%		
[31]Park et al	Vulpius	3	3	0	0	-	-	x	x
[34]Rose et al	Baker	24	5	15	0	-	-	x	x
[38]Segal et al	Vulpius	2	0	2	0	-	0%	x	
[41]Truscelli et al	Vulpius	22	-	-	-	23%	36%		
[43]Yoshimoto et al	Vulpius (+HCA)	20	11	6	0	0%	0%		
[44]Yngve & Chambers	Vulpius	22	3	16	3	-	-	x	x

* avg df in stance 24.1 x = <5 years average follow up

TABLE 4: Zone 1: Proximal Calf Gastrocnemius Recession

Author	Procedure	Feet	Hemis	Dis	Quads	Recurrent equinus %	Calcaneus %	F/up <5 yrs	Gait analysis
[16]Craig & Van Vuren	Strayer	100	-	-	-	9%	3%		
[21]Gough et al	Strayer	22	0	12	0	-	-		x
[22]Gough et al	Strayer	21	0	12	0	25%	-		
[25]Kay et al	Strayer	38	12	22	4	19%	22%		x
[26]Lofterod & Terjesen	Strayer	10	4	6	0	10%	10%		x
[27] Lofterod & Terjesen	Strayer	42	-	-	-	-	-		x
[36] Saraph et al	Baumann	28	0	22	0	0%	0%		x
[37] Saraph et al	Baumann	23	0	32	0	-	-		x
[39] Sharrard & Bernstein	Strayer	53	5	25	23	15%	2%		
[40]Steinwender et al	Baumann	34	0	17	0	-	-		x
[45] Zwick et al	Strayer	11	0	6	0	-	-		x

* avg df in stance 24.1 x = <5 years average follow up

TABLE 5: Equinus Surgery Outcomes in Spastic Diplegia

Author	Procedure	Diplegic Feet	Recurrent equinus %	Calcaneus %	F/up >5 yrs	Gait analysis
[11]Abel et al	Vulpius	42	0%	0%	x	x
[12]Adolfsen et al	Vulpius , TAL (NS)	20	-	-		x
[13]Baddar et al	Vulpius	22	-	0%	x	x
[15]Borton et al	Baker Z TAL Hoke TAL(P)	110	16%	40%		x
[17]Damron et al	Baker, Vulpius Z TAL	37	-	0%		
[18]Dietz et al	Hoke TAL(O), Z TAL	34pts	-	41%		
[19]Engsberg et al	HCA Vulpius White TAL	12 10 10	-	-	x	x
[20]Fabry et al	TAL(O)(NS) Z TAL(P)(NS)	11 6	-	-	x	x
[21]Gough et al	Strayer	22	-	-	x	x
[22]Gough et al	Strayer	22	25%	-	x	x
[24]Javors & Klaaren	Vulpius	46	0%	0%		
[25]Kay et al	Strayer TAL (NS)	22 4	-	-	x	x
[26]Lofterod et al	Strayer Z TAL	14	7%	-	x	x
[27]Lofterod et al	Strayer	34	-	-	x	x
[28]Lyon et al	Hoke TAL, Z TAL	23	35%	?High*	x	x
[29]Olney et al	Baker	108	44%	0%		
[33]Rattey et al	Z TAL	56	18%	2%		
[34]Rose et al	Baker	19	-	-	x	x
[35]Sala et al	Hoke TAL	17	24%	-		
[36]Saraph et al	Baumann	28	0%	0%	x	x
[37]Saraph et al	Baumann	23	-	-		x

TABLE 5: Equinus Surgery Outcomes in Spastic Diplegia (continued)

Author	Procedure	Diplegic Feet	Recurrent equinus %	Calcaneus %	F/up >5 yrs	Gait analysis
[38]Segal et al	Vulpius Hoke TAL (P) Z TAL White TAL HCA	2 12 12 6 6	-	30%		x
[39]Sharrard & Bernstein	Z TAL Strayer	12 25	33% 4%	0% 4%		
[40]Steinwender et al	Baumann	34	-	-		x
[42]Uyttendaele et al	Z TAL	15	33%	-		
[43]Yoshimoto et al	Vulpius ± HCA	6	0%	0%		
[44]Yngve & Chambers	Vulpius Z TAL	16 16	-	-	x	x
[45]Zwick et al	Strayer	11	-	-		x

* avg df in stance 24.1

TABLE 6: Equinus Surgery Outcomes in Spastic Hemiplegia

Author	Procedure	Hemis	Recurrent equinus %	Calcaneus %	F/up >5 yrs	Gait analysis
[12]Adolfsen et al	Strayer TAL(NS)	10	-	-		x
[15]Borton et al	Baker Z TAL Hoke TAL(P)	45	38%	4%		x
[17]Damron et al	Baker, Vulpius Z TAL	4	-	0%		
[18]Dietz et al	Hoke TAL(O), Z TAL	23	-	0%		
[23]Graham & Fixsen	White TAL	35	43%	0%		
[24]Javors & Klaaren	Vulpius	15	4%	0%		
[25]Kay et al	Strayer TAL(NS)	12 11	-	-	x	x
[27]Lofterod & Terjesen	Strayer	13	-	-	x	x
[26]Lofterod & Terjesen	Strayer Z TAL	6	0%	-	x	x
[29]Olney et al	Baker	90	62%	0%		
[31]Park et al	Z TAL(NS) Vulpius	13 3	-	-	x	x
[33]Rattey et al	Z TAL	29	41%	7%		
[34]Rose et al	Baker	5	-	-	x	x
[35]Sala et al	Hoke TAL	4	50%	-		
[39]Sharrard & Bernstein	Z TAL Strayer	37 5	27% 40%	0% 0%		
[42]Uyttendaele et al	Z TAL	10	20%	-		
[43]Yoshimoto et al	Vulpius + HCA	11	0%	0%		
[44]Yngve & Chambers	Vulpius Z TAL	3 11	-	-	x	X

TABLE 7: Single Event Multi-Level Surgery (SEMLS) Articles

Author(s)	Equinus Procedures	Additional Procedures	No of subjects (avg or)	Age (years) Mean (range)	Preop DF IC	Postop DF IC	Preop Max DF	Postop Max DF	Preop KF IC	Postop KF IC	Outcome Measures	Follow-up (yrs/range)
[11]Abel et al	Vulpius	ADD R, REC T, HAM L, STF	27(7)	8.7 (4-20)	0	0	3	10	40	25	3DGA, GMFM	2 (1.6-3.1)
[12]Adolfsen et al	Strayer, TAL(NS)	REC T, HAM L, POTB	31(6)	8.5 (5-15)	-5	-2	7	12	31	21	3DGA, EMG, Clinical exam	1.9 (0.7-6.4)
[18]Dietz et al	Hoke TAL(O), Z TAL(	POTB, HAM L, REC T, ADD R	79(6)	7.7 (2-47)	-	-	-	-	-	-	OGA GRAFO	7 (-)
[21]Gough et al	Strayer	VDRO, HAM L, POTB, REC T, CO	12(6.4)	9.8	-	-	-5	15	-	-	3DGA, EMG Clinical exam	1.5 (-)
[22]Gough et al	Strayer	HAM L, POTB, CO, REC T	12(5)	6.4 (5.9-6.9)	-	-	-	-	-	-	3DGA, GGI, EMG Clinical exam	2 (2-5)
[25]Kay et al	Strayer, Z TAL(NS)	POTB, HAM L, CO, VDO, SMO, REC T, FTT	54(5)	9.8 (4-16)	-	-	0.6	16	-	-	3DGA, PRS, Ashwoth	1.5 (1-2)
[26]Lofterod & Terjesen	Strayer, Z TAL	POTB, REC T, VDO, HAM L	47(3.9)	11.5 (5-19)	-21.9	-7	-6	10.9	23.9	18.4	3DGA exam GMFCS FMS	* 1.2 (.8-1.5)
[28]Lyon et al	Z TAL, Hoke TAL	HAM L, REC T, SMO, VDO, FTT, ADD R	14(5.6)	9.1 (4-17)	-6.8	10.8	10.1	24.1	40.5	32.9	3DGA	* 1.5 (0.5-2.5)
[34]Rose et al	Baker	VDO, SMO, POTB, ADD R, HAM L, REC T	20(5.3)	6 (4-26)	-5	-3	4	12	25	16	Clinical exam 3DGA	* 1 (-)
[36]Saraph et al	Baumann	POTB, ADD R, HAM L, REC T, SMO, VDO	22(8)	12.6 (7.4-16.6)	-16.8	2.2	11.8	7.1	27.2	18.9	Clinical exam 3DGA	* 2 (2.1-4)
[37]Saraph et al	Baumann	POTB, ADD R, HAM L, REC T, SMO, VDO, FF	32(8)	11.1 (8.7-13.5)	-3.9	4.6	6.4	16	-	-	Clincal exam, 3DGA, EMG	* 4.4(1.0-5.4)
[38]Segal et al	Hoke(P) TAL White/ZTAL, Vulpius, HCA	POTB, ADD R, HAM L, VDO, SMO	20(4)	5.2 (2.7-8.2)	-	1.45	-	20.3	-	10.7	Clinical exam 3DGA	5.8 (1.1-11)
[40]Steinwender et al	Baumann	POTB, ADD R, HAM L, REC T, SMO, VDO, FF	17(13.5)	14.4 (-)	-11	0	-6	7.5	28	21	Clinical exam 3DGA	* 3.9 (-)
[44]Yngve & Chambers	Z TAL, Vulpius	POTB, VDRO, ADD R, HAM L, REC T	33	9 (-)	-13	0	-5	11	29	21	Clinical exam 3DGA	* 1 (-)
[45]Zwick et al	Strayer	POTB, ADD R, HAM L, REC T, SMO, VDO, FF	17(8)	11.2 (5.7-16.4)	-3.6	1.4	-0.9	5.1	31.6	23.6	Clinical exam 3DGA	* 3.8 (2.6-5.7)

VDO – Varus Derotational Osteotomy Femur **ADD R** – Adductor Release **HAM L –** Hamstring Lengthening **STF**- Sub-Talar Fusion **FTT -** Foot Tendon Transfer
REC T – Rectus Femoris Transfer **POTB** – Intramuscular Psoas Lengthening **CO** – Calcaneal Osteotomy **SMO –** Supra Malleolar Osteotomy **FF -** Foot Fusion

ORTHOPAEDIC MANAGEMENT OF SPINAL DEFORMITY IN CEREBRAL PALSY

Jeremy Fairbank MA MD FRCS
Professor of Spinal Surgery, University of Oxford, UK
&
Adnan Zubovic MD
Nuffield Orthopaedic Centre, Oxford

In principal, any growing child with a significant neurological deficit is vulnerable to developing combinations of scoliosis, lordosis and kyphosis. The more severe the deficit, the more likely is spinal deformity to occur, the earlier the age of onset, and curves are likely to be more severe. Curve progression is most obvious during spinal growth, but continues in adult life. These observations are based on personal experience. The quality of the available literature is poor. The closest we have to documented natural history of spinal deformity is Saito et al's study,[1] which we believe is consistent with these assertions. In general if a child with CP can walk, then serious scoliosis is much less likely compared with a wheelchair dependent individual. The aim of their study was to clarify the natural history of scoliosis from childhood through to adulthood and provide objective data on proper surgical indications for such patients. Authors retrospectively analyzed 37 institutionalized patients with severe spastic palsy and scoliosis. They reviewed the series of radiographs with an average follow up of 17.3 years. Assessed were five factors with the effect on the progression of scoliosis: sex, degree of spasticity, initial physical capability, pattern of spinal curve and location of curve. Their findings included the one that the scoliosis usually started before the age of ten years and progressed rapidly during the growth period. They also found that the progression of the scoliosis occurred in many cases even after the growth had ended. The mean Cobb angle of the curves at final examination was 55˚. In 85% of patients with the curve of >40˚ before the age of 15 years, the scoliosis progressed to >60˚ by the time of the spinal examination. In contrast to that, in only 13% of patients who had a curve <40˚ at the age of 15 years did the scoliosis progress to >60˚. Severe scoliosis (>60˚) developed predominantly in those patients with the total body involvement (67%), were bedridden (100%) or had thoracolumbar curves (57%). Authors concluded that the risk factors for progression of scoliosis in spastic cerebral palsy are: having total body involvement; being bedridden; and having a thoracolumbar curve. Their opinion was that the patients with these risk factors might benefit from early surgical intervention to prevent the progression to severe scoliosis – Oxford CEBM B.

Cerebral palsy may be classified topographically (hemiplegia, diplegia and total body involvement etc) or neurologically (spastic, athetoid, ataxic, rigid or mixed). Deformities in cerebral palsy are mobile or fixed, and generally progress from mobile to become fixed. We may speculate on the cause of spinal deformity. It is generally assumed to be the lack of dynamic muscle control to counteract the deforming forces resulting from gravity. As the size of deformity increases so gravity takes an increasing role and hastens the progression of the deformity. There are strong biological drivers for the spine to grow "straight". It is likely that there is some variability in this, and it may be that some children are more vulnerable to external deforming forces than others. The evidence for this statement is limited to personal observations.

INTERVENTIONS TO PREVENT AND TREAT SPINAL DEFORMITY

Posturing

Sleep systems are widely used but we have difficulty in finding evidence of efficacy. Daytime posturing has observational data mainly focussed on the hips.

Seating

This is an important element of managing a child with 4 limb involvement CP. Because it is universally applied it is difficult to establish efficacy. Seating can be moulded or modular. Reclining position or tilt-in-space can be used when gravity loading is a problem. However, tilting can interfere with quality of life if a child cannot see, or wants to use a keyboard, or during feeding.

Bracing

Custom moulded braces are useful especially in the younger child. We have used "soft" braces for "collapsing" spines. The problem is that as the child gets older and larger the surface loads on the brace can cause problems in progressive curves. It is also difficult to combine moulded seating and brace treatments, although it is easier to combine braces and modular seating. Evidence for efficacy is hard to come by.

Surgery

Surgical treatment is considered where there is a need to control progressive curves, to stabilise and correct pelvic obliquity, to control pain related to sitting and relieved by lying down, and where spinal deformity is contributing to respiratory problems. In some cases improvement of body image (for either patient or carers or both) is a benefit. Some carers report that lifting and handling of the patient is easier after the surgery. These potential benefits have to weighed against serious risks attached to such major surgery on a child or young adult with poor nutrition, respiratory function, poor bone quality and often major learning difficulties and limited communication capacity. Risks include mortality, infection, spinal cord and nerve damage, implant failure, infection, pseudoarthrosis, revision surgery, and perhaps worst of all, ventilator dependence.

The decision to proceed to surgery often lies with the carers with limited or no contribution from the patient, which is particularly challenging for all concerned. The decision is based on estimate of quality of life now and in the future with or without the intervention. These can be some of the toughest decisions in spinal surgery. Help in decision making should be sought from all those involved in the care of the patient.

Over the last 20 years there have been important advances in the implants available to surgeons. Where possible surgery is done posteriorly, but in some cases anterior release and instrumentation will contribute to a stable outcome. The more severe curves require preliminary anterior release and posterior stabilization. If there is fixed pelvic obliquity, the fusion should be extended to sacrum. Leaving the L5-S1 segment free leaves some spinal mobility, but risks instability and the need for further surgery later.

Anaesthetic techniques have also improved enormously with better control of bleeding and respiratory function, post-operative care and pain control.

EVIDENCE BASE FOR SPINAL SURGERY

This remains based on observational studies and personal experience. Once a decision has been made to proceed to surgery there is a big incentive for both surgeons and carers to say that the intervention was worth it. Outcome measures, beyond the geometry of the curve before and afterwards and the presence of complications, are of poor quality. Scoliosis is common in children with cerebral palsy and occurs in 15-25% of patients. Most curve patterns are long thoracolumbar or lumbar.

Comstock et al. in 1998 analyzed the surgical treatment and patient and caregiver satisfaction in patients with the total-body-involvement cerebral palsy.[2] In their nonrandomized descriptive case series authors analyzed the data from 79 of 100 patients with total-body-involvement spastic cerebral palsy who underwent posterior Luque instrumentation, or anterior spinal fusion, or both. Functional status was evaluated by physical examination and a personal interview was conducted with the patient, parents and a primary caregiver. In a median follow-up of 4 years late progression of scoliosis (>10˚), pelvic obliquity (>5˚), and decompensation (>4cm) were noted in>30% of patients. More than 75% of patients with late progression were skeletally immature at the time of surgery and underwent a posterior procedure only. 21% of the patients required a revision procedure because of the disease progression. Progression was not noted in any patients who underwent anterior fusion (instrumented or non-instrumented) with posterior instrumentation from the upper thoracic spine to the pelvis. 85% of caregivers were very satisfied with the results of surgery and noted a beneficial impact on the patient's sitting ability, physical appearance, ease of care, and comfort. The authors concluded that to avoid late progression of trunk deformity in skeletally immature patients, anterior spinal release and fusion combined with posterior segmental spinal instrumentation and fusion from the upper thoracic spine to the pelvis are recommended. Skeletally mature patients with good curve flexibility can be treated with posterior instrumentation and fusion only. Skeletally mature patients with large fixed curves benefit from an anterior-posterior procedure for better correction of the scoliosis and pelvis obliquity. Despite the surgical complexity and expected complications, the overall good surgical results and high patient and caregiver satisfaction confirm that corrective scoliosis surgery is indicated and beneficial for most patients with total-body-involvement cerebral palsy and scoliosis – Oxford CEBM C.

Teli et al. in 2006 reported on the treatment of patients with cerebral palsy and neuropathic scoliosis with third-generation instrumented spinal fusion by Cotrel-Dubousset instrumentation.[3] In their retrospective study authors analyzed 60 patients with minimum follow-up of two years. Mean age of patients was 15 years at the time of surgery. There were 26 anteroposterior and 34 posterior-only procedures, with the correction of coronal deformity and pelvic obliquity of average 60% and 40% respectively. Major complications occurred in 13.5% of patients and included implant loosening, deep infection and pseudoarthrosis. Outcome questionnaires showed marked improvements in the areas of satisfaction, function and quality of life. Third-generation instrumented spinal fusion has enhanced correction and allows early ambulating and nursing. The limitations of this study included the retrospective design and the absence of an internal control group treated with second-generation implants. Authors observed that in their cohort group, percentage correction of scoliosis was very similar inpatients treated posteriorly and anteroposteriorly, although the latter group had by definition stiffer curves on preoperative imaging. Correction of pelvic obliquity was significantly higher in patients in patients treated

with the double approach, which might suggest a positive role of the anterior release of lumbar spine toward correction of pelvic deformity. The improvement of trunk balance and pelvic obliquity led to better sitting balance and the absence of pressure sores at follow-up. Patients with posterior-only procedures had the shortest hospital stay and operating time, but the highest incidence of complications, particularly superficial and deep infections. Authors concluded that segmental, third-generation instrumented spinal fusion provides lasting correction of spinal deformity and improved quality of life in patients with cerebral palsy and neuropathic scoliosis, with a lower pseudoarthrosis rate compared to reports on second-generation instrumented spinal fusion – Oxford CEBM C.

Tsirikos et al. compared one-stage versus two-stage anteroposterior spinal fusion in paediatric patients with cerebral palsy and neuromuscular scoliosis.[4] In this retrospective study the objective was to evaluate the outcomes and complications of two treatments in 45 paediatric patients and to document which procedure is more efficacious and provides better results. There was no statistically significant difference between one-stage and two-stage patients, considering age at surgery, preoperative scoliosis angle, pelvic obliquity, kyphosis angle, lordosis angle, levels of anterior release, percentage of scoliosis correction, radiographic follow-up, hospitalization time, and intensive care unit stay. Sequentially performed spinal procedures (one-stage group) were associated with increased intraoperative blood loss, prolonged operative time, and a considerably higher incidence of medical and technical complications, including two perioperative deaths. Authors of this study concluded that the one-stage procedure were associated with a considerable mortality rate, increased operative morbidity and higher risk of technical complications. They recommended two-stage anteroposterior spinal fusion as safer and more advantageous over the single-stage procedure, especially in individuals with very large curves and concomitant medical illness – Oxford CEBM C.

Tsirikos et al analyzed the life expectancy in patients with cerebral palsy and neuromuscular scoliosis following spinal fusion.[4,5] In their retrospective cohort study they analyzed 288 severely affected paediatric patients with spasticity and neuromuscular scoliosis who underwent spinal fusion. Mean age of patients was 13 years and 11 months at the time of surgery. Kaplan-Meier survival analysis was performed demonstrating a mean predicted survival time of 11 years and 2 months after spinal surgery for this group of globally involved children with cerebral palsy. The most significant determinants for reduced life expectancy in paediatric patients with cerebral palsy are the coexistence of other significant disorders such as respiratory malfunction, epilepsy, feeding disorders, the degree of neurological involvement, the presence of severe cognitive abnormality, and the level of ambulatory disability. Authors of this study observed that the preoperative degree of scoliotic deformity was not correlated statistically with survival rates. The days spent in the ICU appeared to be the most accurate predictive factor for the long-term survival of severely affected children and adolescents. They concluded that the number of days spent in the intensive care unit after surgery and the presence of severe preoperative thoracic hyperkyphosis were the only factors affecting the survival rates and that the parents of high-risk children should be counselled about the prognosis – Oxford CEBM C.

OUTCOME MEASURES FOR NEUROMUSCULAR SCOLIOSIS.

This section gives brief summaries of relevant papers.

The Effect of Spinal Surgery on Function in Children with Neuromotor Disorders and Scoliosis: A prospective study using standardised measures. Nicky Courtier, John O'Dowd, Elspeth Will, Jonathan Lucas, Khai Lam, Elizabeth Wraige. Deformity Service, Guy's and St.Thomas's Spinal Unit, London (unpublished).

Introduction: It is widely accepted that operating on children with neuromotor disorders and scoliosis who are full time wheelchair users is often to prevent/correct deformity with the primary goal of maintaining/ improving sitting ability. In those children who are ambulant, preventing further deformity is usually the objective with the effect on function being less clear. The purpose of this study was to assess the impact of spinal surgery on all aspects of function in children with neuromotor disorders using standardised functional measures.

Methods: A consecutive series of 20 children (age range 2 –18) who underwent posterior only or anterior and posterior spinal fusion for neuromuscular scoliosis. Patients were assessed pre-op (A1), post-op (A2), at 3 months (A3) and 12 months (A4). The Seated Postural Control Measure (SPCM) examined posture and function (A1 – A4). The Paediatric Evaluation of Disability Inventory (PEDI) recorded. functional ability in the domains of self-care, mobility and social function (A1, A3, and A4).

Results: The SPCM demonstrated an improvement in posture in 95% from A1 – A2 with 25% demonstrating some regression at A3. Over 70% maintained or further improved posture by A4. The PEDI measured an increase or maintenance of self-care in at least 50% patients at A3. All patients demonstrated a reduction in mobility at A3. At least 60% when measured at 1 year post-op had regained their pre-op levels of mobility and self-care.

Discussion: Although children within this group do improve their sitting ability post-op, many other areas of their function are also affected following spinal surgery. These may have more of an impact on the child's/families' life. Both measures used in this study had some limitations but offered the opportunity to repeat assessments of care and independence over time exploring other considerations when deciding about spinal surgery.

Longitudinal Parental Perceptions of Spinal Fusion for Neuromuscular Spine Deformity in Patients with Totally Involved Cerebral Palsy. Jones et al (JPO, 2003) John Hopkins [6]

Prospective study over 3.5 yrs, consecutive pts
N=17(of 25) 10-21yo (mean 14.2) 11M, 9F
Cobb angle Complications
PODCI questionnaire
Scope – Comment on overall risks and carer perceived benefits of surgery for this small, homogenous patient group

Process Measures and Patient/Parent Evaluation of Surgical Management of Spinal Deformities in Patients with Progressive Flaccid Neuromuscular Scoliosis (Duchenne's Muscular Dystrophy and Spinal Muscle Atrophy). Bridwell et al (Spine, 1999) Missouri [7]

Prospective study assessing results and their own questionnaire 1985-95
N=48(of 55) 7-18yo (mean 13.4) 29DMD, 16SMA

Cobb angle
Complications
Questionnaire
Scope
Objective and subjective outcomes identified in a medium sized sample
Tool for assessment tested in flaccid neuromuscular scoliosis patients

Spinal Fusion and Instrumentation for Paediatric Neuromuscular Scoliosis: Retrospective Review. Thacker et al (J Orth Surg (Hong Kong), 2002) Singapore
Retrospective study 1993-98 CP,DMD, SMA
N=24 6-14yo (mean 10.6) 5.5 yr F/U
Cobb angle
Complications
ITU stay
FEV1 Sitting Ability
Scope
Assessment of procedure safety

Standards in Anterior Spine Surgery in Paediatric Patients with Neuromuscular Scoliosis. Sarwahi et al (JPO, 2001) Chicago [8]
Retrospective study 1988-95 CP, Spina Bifida, NF
N=111 3-18yo (mean 12.3) 46M, 62F
Cobb angle
Complications
Diagnosis
Procedure
Operative blood loss
Scope
Identification of risk factors for complications in anterior spinal surgery

Results and Morbidity in a Consecutive Series of Patients Undergoing Spinal Fusion for Neuromuscular Scoliosis. Benson et al (Spine, 1998) Connecticut [9]
Retrospective study 1990-95 CP, Spina Bifida, SMA
N=50 4-29yo (mean 13.5) 22M, 28F 3.3yr F/U
Cobb angle
Degree of correction
Complications
Scope
Radiological results and complication rates recorded

Goal setting in neuromuscular scoliosis surgery. Is it important? Nicky Courtier, John O'Dowd, Elspeth Will. Deformity Service, Guy's and St. Thomas' Spinal Unit, London (unpublished data).
Introduction: In adolescent idiopathic scoliosis the decision to operate is based on the cosmetic appearance of the deformity. With neuromuscular scoliosis, the objective of surgery is to maintain/improve sitting ability. However, the effect of surgery on other aspects of function in this group is often overlooked. On this basis, the overall success/satisfaction of the surgical intervention is dependent on the realistic expectations from the family/carers of what the surgery can offer.
Method: A group of 20 children who underwent spinal fusion for neuromuscular scoliosis were assessed using a postural and functional measure pre-op, post-op, at 3 months and 12 months post-op. In addition, each patient was asked to record three

goals for undergoing the surgery. At one year post op, patients were asked to grade on a scale of 0 – 10, how satisfied they were that the goals had been achieved.

Results: Nineteen patients had clear pre-op goals for the surgery relating to functional activities. The most frequent goals stated for the non-ambulant children were: Sitting for longer periods (7/46), making dressing easier (7/46) and sitting more upright (6/38). There were 15 other functional goals stated. The ambulant children stated: Appearing straighter (3/12), increasing in confidence (2/12), reducing pain (2/12) and maintaining respiratory function (2/12). There were 3 other functional goals stated. Seventeen patients completed the study, 2 were lost to follow up, 1 died. The average satisfaction rate from goals achieved 1 year post-op was 7.9/10.

Discussion: Establishing goals that are realistic and contribute positively to the functional ability or practical management of the child with neuromuscular scoliosis undergoing spinal surgery, encourages the family to be central in the decision making process. It also allows unrealistic expectations to be discussed pre-op and may appear to be a key factor in the overall success and satisfaction of the surgical intervention.

Conclusions:
Bracing is first line treatment for CP related spinal deformity. The evidence basis is poor, and in each case treatment is experimental. This has to be linked with seating and sleep systems with an emphasis on standing where this is possible in a frame or with splints and walking aids.
Surgery is used as part of the management of CP related spinal deformity only when non-operative treatment has failed. A careful discussion of risks and benefits is needed with all concerned parties, recognising that surgery is dangerous and takes many months to get over. The position of the Pelvis in coronal and sagittal planes is crucial. Patients with limited hip movement require particular consideration. The objective has to be a comfortable sitting posture. The reward is a patient whose pain has been relieved. Bonuses are when the rate of respiratory infections is reduced and when appetite is restored. Risks are of serious operative complications particularly of infections and repeat surgery. The surgeon must be constantly self critical and aware of their limitations. Surprising successes may encourage surgery against the odds and must be constantly guarded against. Non-surgeons should have the courage to question surgical decision making at all stages.

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A SYSTEMATIC REVIEW OF THE USE OF ORTHOSES IN THE MANAGEMENT OF PATIENTS WITH CEREBRAL PALSY: HIP, TRUNK, SPINE AND UPPER LIMB.

Phil Stevens, MEd, CPO, FAAOP
Specialized Prosthetic and Orthotic Technologies
Salt Lake City, USA

The purpose of this article is to provide a comprehensive review of the role of orthoses in the management of the hips, trunk, spine and upper extremities in persons with cerebral palsy. The management of the lower extremities is treated in a separate literature review. The majority of the referenced studies have been published since the first ISPO consensus conference on this topic in 1994, reflecting both newer treatment methods and enhanced experience in more traditional treatment approaches. Additionally, the larger acceptance of recent surgical, pharmaceutical and/or therapeutic modalities in the management of this complex population has created new adjunctive applications within the field of orthotics that are addressed. Finally, where historical foundations were considered beneficial, older publications have also been referenced. In reviewing this literature, the following questions were developed and answered according to the best available evidence:

- Do spinal orthoses improve posture, balance and associated control of the head neck and upper extremities in children with cerebral palsy when compared to unbraced conditions?
- Do spinal orthoses have a negative impact on pulmonary or circulatory function in children with cerebral palsy compared to unbraced conditions?
- Do spinal orthoses prevent or delay curve progression compared to natural history in children with cerebral palsy?
- Do hip orthoses prevent progressive hip subluxation compared to natural history in children with cerebral palsy?
- Do daytime hip orthoses improve gross and/or fine motor function in children with cerebral palsy compared to unbraced controls?
- Do daytime orthoses increase muscle activity in hip adductors in children with cerebral palsy compared to unbraced muscle activity?
- Do the combined modalities of dynamic assist upper extremity orthoses and functional stimulation of antagonist muscle groups decrease upper limb spasticity and/or improve upper limb function in children with cerebral?
- Do dynamic assist upper extremity orthoses maintain upper extremity range of motion more effectively than static splinting modalities in the treatment of patients with cerebral palsy?
- Are there established benefits associated with the bracing of the wrist joint with static and/or dynamic splints in children with cerebral palsy?
- Are there established benefits with respect to upper extremity function associated with the use of lycra garments in children with cerebral palsy?
- Are there established benefits associated with the use of lycra garments with respect to lower limb function in children with Cerebral Palsy?
- Are there established benefits associated with the use of Johnstone pressure splints in the management of children with cerebral palsy?

METHODS

A search of the literature was conducted in April and May, 2007 using the Cochrane Central Register of Controlled Trials, Ovid Medline, CINHAL and RECAL Legacy. The following search terms were combined with the base term "cerebral palsy:" "orthoses," "orthotic devices," "brace," "bracing," "splints," "splinting" "spinal curvatures," "scoliosis," "kyphosis," "jacket," "TLSO," "hip," and "upper extremity." In addition, references in original articles were examined to verify that no relevant literature had been overlooked. References were hand searched if they were published between 1994 and the present date. Articles were selected for further review if they addressed the role of orthotic modalities with respect to the spine, trunk, hip and upper extremities of persons with cerebral palsy. Publications focused solely on surgical techniques were not reviewed. Likewise, articles focused on seating and positioning were not reviewed unless they made specific reference to spinal or hip orthoses. When relevant, studies pertaining to adjuvant surgeries, therapies and other interventions were included. Relevant historic literature was also evaluated as appropriate to establish historical foundations where appropriate.

In the "research design" section of the tables, the following phrases are utilized.

Randomized controlled trial: A prospective experimental study in which subjects are *randomly* assigned to either a control or intervention group. Outcome measures are assessed after an appropriate follow-up time and results are compared between the control and intervention groups.

Cohort study: A prospective or retrospective observational study of subjects that may develop a specific condition. Subjects may be sub-divided into groups based on exposure to factors that may influence occurrence of the condition. Incidence of the condition is assessed after an appropriate follow-up time and subjects are compared to predict influential factors.

Case Series: A descriptive, observational study of the diagnosis, prognosis, treatment and/or outcomes of a subject group with the same (or similar aspects of a) condition.

Interrupted time series trial: A prospective experimental study in which multiple subjects are assigned only to an intervention group. No control group is formed; instead subjects serve as their own control. Subjects are evaluated *multiple times* before and after one or more interventions. *Repeated outcome measures* are assessed at known intervals and results are compared between the studied conditions.

Controlled before-and-after trial: A prospective experimental study in which multiple subjects are assigned only to an intervention group. No control group is formed; instead subjects serve as their own control. Subjects are evaluated *once before and once after* one or more interventions. Outcome measures are assessed at known intervals and results are compared between the studied conditions.

Qualitative Study: A descriptive, observational study in which a subject group is evaluated through subjective, open-ended questions and interview techniques.

Convenience Sample: Subjects were chosen on the basis of availability.

Levels of Evidence were determined according to the Oxford Centre for Evidence-based Medicine Levels of Evidence (May 2001)

SPINE

Do spinal orthoses improve posture, balance and associated control of the head neck and upper extremities in children with cerebral palsy when compared to unbraced conditions?

The use thoracolumbar sacral orthoses (TLSOs) to treat spinal deformities in persons with cerebral palsy was first described in a preliminary report in 1977, in which the use of "removable orthoplast jackets" was found to temporarily halt the curve progressions with low instances of both complications and patient rejection[1].

Fifteen years later, citing high rates of pressure sores and skin irritation, as well poor general tolerance of rigid spinal orthoses among patients with neuromuscular scoliosis (NMS), authors detailed their preliminary use of the "Soft Boston Orthosis" among NMS patients, the majority of which were patients with quadriplegic cerebral palsy[2]." The construct of this device was reported as two layers of Aliplast, with high density polyethylene panels compressed at intervals between the two layers. Responding to questionnaires, staff members reported improved head and trunk control and enhanced postural position and sitting stability associated with the use of the device. Subsequent investigations have reported similar benefits with both custom molded polypropylene and polyethylene TLSOs as well as "TLSOs with non-rigid SIDO frames" (essentially a posterior TLSHO which allows flexion and limited rotation of the trunk), including improved sitting balance and resultant postures of the head, trunk and extremities[3,4] (See Table 1). Such improvements have also been suggested within several expert opinion publications[5,6].

Do spinal orthoses have a negative impact on pulmonary or circulatory function in children with cerebral palsy compared to unbraced conditions?

Addressing concerns of respiratory compromise with the use of the Soft Boston Orthoses, the authors cited earlier reported no appreciable compromise in oxygen saturation levels associated with the use of the spinal orthosis[2]. In reference to this last concept, careful distinction needs to be drawn between the reported negative impacts of spinal bracing on neuromuscular scoliosis due to myopathies and those observed with the bracing of neuromuscular scoliosis due to neuropathy. Reports of reduced vital capacity historically observed in patients with muscular dystrophies and spinal muscle atrophies[7] should not be generalized to the cerebral palsy population. The sole publication addressing this concept within the cerebral palsy population found no significant differences in any of the major indices of pulmonary mechanics when patients with spastic quadriplegia were assessed with and without their Soft Boston Orthosis in sitting and supine[8] (See Table 2).

Do spinal orthoses prevent or delay curve progression compared to natural history in children with cerebral palsy?

The ability of spinal orthoses to retard curve progression and delay or prevent scoliosis is poorly understood. In a study whose population was said to exclude all but the most severely spastic quadriplegic patients, the use of custom molded

Wilmington orthoses failed to impact group curve progression rates or magnitudes compared to those of an unmatched cohort of unbraced controls[9]. Unfortunately, the magnitude and flexibility of the curves at the time when bracing was initiated were not reported. In a later study, children with spastic quadriplegia were considered within a larger cohort of NMS in which prefabricated Boston type braces were used. Subjects were monitored for an average of 3 years following the discontinuation of bracing. In nine of the 37 children with quadriplegic cerebral palsy, bracing was found to be successful, with curves failing to progress more than 10° during the observation period[10]. When subjects with athetosis are excluded from these findings, seven of 25 cases met the authors' criteria for success. Among the broad diagnosis of NMS, successful bracing outcomes were related to shorter, smaller curves. Unfortunately, given the mixed diagnoses reported on in the publication, these characteristics can only be cautiously generalized to the cerebral palsy population.

A final publication reported on retrospective analysis of 86 patients with spastic quadriplegic cerebral palsy who were managed with custom molded TLSOs[3]. The mean Cobb angle at the initiation of bracing for this cohort was 68°. As with the Wilmington orthoses, researchers found that as a group, the rate of curve progression within their braced subjects was similar to reported natural history values[11]. However, their report included a small cohort of patients, 16% of those under the age of 15 and 64% of those 15 or older, whose braced curves progressed less than 1° per year. Their numbers are strikingly similar to those suggested by a commonly sited source for expert opinion which observed that no more than 15% of braced curves stop progressing[12]. Regression analysis found that neither the Cobb angle at initiation of bracing, the type of scoliosis or the amount of observed pelvic obliquity was predictive of successful outcomes with bracing. The only variable associated with slowed curve progression was initial in-brace correction. Among their conclusions, the authors suggest that greater correction in the orthosis seems to be the only changeable factor associated with curve progression. However, given that the orthoses in the study were all made by the same team of experienced clinicians, clinical experience suggests that the variable outcomes observed could also have resulted from differences in curve flexibility (See Table 3).

Adjunctive Considerations

Ginsburg and Lauder recently reported their observations of rapid progressions of scoliosis in patients with spastic quadriplegia following the insertion of an intrathecal baclofen pump[13]. Reporting on 19 consecutively observed patients, the authors report a mean preoperative Cobb measurement of 10.2°. The average postoperative Cobb measurements for the same group, taken at an average of 20.9 months after pump insertion was 25.0°. Further analysis found that subjects were experiencing a 6-fold increase in the curve progression rate following the pump insertion. As a result, the authors report that they are now recommending spinal bracing once pumps are inserted with in-brace radiographs taken at 4 month intervals. As the use of intrathecal baclofen pumps becomes more common in the treatment of spasticity, the impact of these pumps on scoliotic curve progression rates and the implications with respect to spinal bracing will warrant further investigation.

Similarly, multiple articles have been published addressing selective dorsal rhizotomy within the diplegic and quadriplegic cerebral palsy populations with increased incidences of scoliosis[14-19], hyperlordosis[14-16,18-19], spondylolysis[16],

spondylotisthesis[14,16-18], and abnormal kyphosis[18-19]. The extent and nature with which this phenomenon will affect spinal bracing remains to be seen.

Conclusions and Recommendations: Spine

The primary benefits associated with the use of TLSO's in the cerebral palsy population appear to be related to improved balance and stability with positive secondary affects in the form of improved control of the extremities, head, neck and trunk. Concerns regarding possible respiratory compromise associated with the use of TLSO's appear to be unfounded. Though spinal curves appear to progress in most patients with cerebral palsy, there is a minority of patients for whom TLSO's appear to be effecting in slowing or halting curve progression. Curve flexibility may be a good predictor of the efficacy of TLSO's in slowing the progression of spinal curvatures. The adjunctive treatment modalities of intrathecal baclofen pumps and dorsal rhizotomies appear to influence the natural history of spinal deformities in this population. The extent to which this will affect bracing recommendations and outcomes remains to be seen.

HIP

Hip instability is a universally acknowledged challenge in the management of patients with cerebral palsy. In addition to the associated pain, compromised sitting stability and risk for pressure ulcers, hip subluxation has also been closely linked with the development of pelvic obliquity which is, in turn, closely associated with the presence of progressive scoliosis[1-3]. A recent systematic review has identified those patients with quadriplegic cerebral palsy as being at the greatest risk for hip instability, while those ambulatory patients with hemiplegia and diplegia are at a far lesser risk[4]. Similarly, gross motor function levels appear to be strongly associated with hip displacement rates, with lowest levels of motor function predictive of the highest rates of hip displacement[5]. Hip surveillance programs have been advocated, in which regular radiographs are taken for earlier detection of displacing hips and preventative surgeries are performed. Such programs have resulted in decreases in late hip dislocations and their described sequelae[6-7].

Various nonoperative modalities, including assorted bracing approaches and alignment systems have been described, both to discourage initial hip displacement and maintain proper hip alignment postoperatively[8-12]. However, patient compliance and the general efficacy of such treatments have been justifiably questioned[6,9,13-14]. Clinical trials with respect to hip orthoses and the management of hip displacement are currently limited to a single publication[15] (See Table 4).

Do hip orthoses prevent progressive hip subluxation compared to natural history in children with cerebral palsy?

The publication in question was a randomized, controlled evaluation of the combined effects of regular botulinum toxin A (Botox A) injections to the adductors and hamstrings and sustained regular use of a variable abduction hip orthosis on reducing hip displacement rates compared to matched controls. Both populations were followed for three years, with regular radiographs documenting hip displacement levels. At the conclusion of the trial period, progressive hip displacement was observed in both groups with the rate of hip displacement in the treatment group being

only slightly reduced relative to that of the control group[15]. The observed benefits largely failed to justify to temporal, procedural and financial costs of the treatment.

Do daytime hip orthoses improve gross and/or fine motor function in children with cerebral palsy compared to unbraced controls?

In addition to the questioned effects of variable abduction hip orthoses combined with Botox A injections on hip displacement rates, an earlier investigation was performed to evaluate the effects of the same treatment on gross motor function levels[16] (See Table 5). In this randomized, controlled trial, gross motor function was assessed in experimental and controlled populations at 12 month intervals. Groups were comprised of children with bilateral spastic cerebral palsy between the ages of 1-4 years old at enrollment, the majority of which were classified at Gross Motor Function Classification System (GMFCS) levels IV and V, consistent with lower motor function and greater disability[17]. As with the previously described trial, controls received regular Botox A injections at the hip adductors and hamstrings. In addition, they were provided with a variable abduction hip orthosis which they were instructed to use 6-8 hours daily. Gross motor function was assessed using the Gross Motor Function Measure (GMFM) as described by Russell et al[18]. Hip orthoses were not used during either the initial or concluding assessments. Authors reported that GMFM scores improved comparably in both study groups, with the GMFM scores of experimental group failing to improve beyond the rates observed in the matched controls[16].

Relevant to the expert opinion claims that hip orthoses are poorly tolerated with questionable compliance, the parents of the experimental study subjects reported via questionnaires that the orthosis, the Standing Walking and Sitting Hip (SWASH) was comfortable, durable and easy to don and doff. Furthermore, the majority reported improved sitting and standing balance with the SWASH. Brace wear was reported at a mean value of 5.6 hours per day, suggestive of good compliance[16].

The findings of this study should be carefully considered against other publications which suggest that the outcome measure selected, the GMFM, may not have been appropriate to detect any existent changes in motor performance within the study's population. Five years has been suggested as the minimum age at which the GMFM should be used to assess motor function[18-19]. Additionally, the sensitivity of the GMFM among patients at lower functioning, GMFCS levels IV and V, has been questioned[19]. Therefore, the study cohort may have been both too young and too severely disabled for the GMFM to detect changes in gross motor function. Interestingly, a treatment affect was observed among experimental subjects at GMFCS level III. These findings may either reflect improved treatment outcomes in patients with lesser levels of disability or simply an increased sensitivity of the selected outcome measure in this population.

Do daytime orthoses increase muscle activity in hip adductors in children with cerebral palsy compared to unbraced muscle activity?

It has been suggested that hip bracing is poorly tolerated by children with cerebral palsy because the passive abduction in the brace activates stretch reflexes in the spastic hip adductors[6,14]. However, the sole quantitative evaluation of this hypothesis failed to support it[20] (See Table 6). Using surface electromyography (SEMG) in a cross-over design with children with spastic diplegia, SEMG amplitudes at the hip

adductors were found to be similar with and without the SWASH orthosis. The study suggests that clinical concerns regarding increased adductor muscle activity with the use of the SWASH orthosis seem unwarranted[20].

Adjunctive Considerations

There is a small body of literature which has investigated the components and relevance of a functional sitting position for children with cerebral palsy[21-23]. A hip orthosis, consisting of two thigh cuffs separated by a spring, was described as one means of creating the symmetrical weight bearing distribution necessary for functional sitting[21]. Authors later used SEMG to record values from various leg muscles in various seat inclinations, both with and without an abduction orthosis while study subjects engaged in an upper extremity task[22]. Results indicated that a horizontal, forward-leaning seat with the use of an abduction orthosis decreased lower extremity muscle activity and might facilitate improved upper extremity function[22]. A later investigation with similar methodology failed to support the findings of the preceding study, observing similar SEMG findings, both with and without the use of hip abduction orthoses[23]. The later study suggests that when the other components of the functional sitting position are maintained, a hip abduction orthosis may be unnecessary.

Conclusions and Recommendations: Hip

While widely utilized, there is very little evidence to support or oppose the use of hip orthoses in the treatment of cerebral palsy. Static hip orthoses and their impact on hip subluxation rates have not received objective scrutiny. Functional hip orthoses such as the SWASH do not appear to impact hip displacement rates, but may be better tolerated than their static predecessors. The assertion that functional hip orthoses may encourage increased use of the hip adductors appears to be unfounded. The basic purposes of functional hip abduction orthoses in discouraging scissoring gait patterns and improving sitting stability have received limited scrutiny. Hip abduction appears to be an important component of functional sitting postures. The use of hip orthoses in facilitating this alignment has been observed.

UPPER LIMB

Do the combined modalities of dynamic assist upper extremity orthoses and functional stimulation of antagonist muscle groups decrease upper limb spasticity and/or improve upper limb function in children with cerebral?

There is a small body of literature supporting the combined treatments of brief daily use of neuromuscular electrical stimulation (NMES) coupled with simultaneous dynamic bracing and concomitant static nocturnal bracing (See Table 7). The treatment is based on the premise that spasticity has both a static and dynamic component. It has been suggested that orthoses address the static component of shortened muscles while NMES addresses the dynamic component of abnormal tone and muscle imbalances[1]. The first of these trials reported retrospectively on the observed affects of the treatment on 19 patients with hemiplegia, mild to moderate proximal spasticity (scapula, shoulder and elbow) and moderate to severe spasticity of the wrist and fingers. NMES was applied for one hour a day over the elbow extensors and wrist and finger extensors while subjects were wearing dynamic upper extremity

bracing[1]. The later study was a prospective design in which 24 subjects were randomly assigned to one of three treatment groups, NMES only, dynamic bracing only, and NMES combined with dynamic bracing[2]. Similar orthoses were used in each endeavor, inclusive of elbow-wrist-hand-finger orthoses with dynamic elbow extension and metacarpophalangeal extension joints.

In the first endeavor, authors reported universal decreases in spasticity and improved wrist and finger postures according to the Zancolli's classification system[3]. Similar improvements within the same classification system were reported in the later study, but only among those patient's who received both NMES and dynamic bracing. Grip strength and an upper extremity dexterity measure, the Melbourne Assessment of Unilateral Limb Function[4] were also reported outcomes in the later trial. Again, improvements in these measures were restricted to those patients receiving the combined treatments. Importantly, in the later study, authors reported that all of the improvements observed in the combined treatment group lasted for only one to three months after the conclusion of the treatments[2].

Do dynamic assist upper extremity orthoses maintain upper extremity range of motion more effectively than static splinting modalities in the treatment of patients with cerebral palsy?

In addition to these trials, there have two reports on small case series in which dynamic bracing of the elbow was used in children with CP following injections of botulinum toxin A into the biceps and brachioradialis and several weeks of corrective serial casting (See Table 8). In the first of these, three children with quadriplegic cerebral palsy underwent the injections and four weeks of serial casts bilaterally. For each child, each arm was fitted with one of two "maintenance orthoses" which were worn for an average of 4 hours per day. One arm was randomly assigned to receive a bivalved long arm cast while the other was fit with an elbow-wrist-hand orthosis with dynamic elbow extension assist joints. The authors reported significant improvements in passive range of motion at the elbow following the injections and serials casts. However, these improvements were found to be temporary, with range of motion limitations returning as the injections wore off. The losses in range of motion during the maintenance period were not as great with the arms treated with the orthosis. However, given the small sample size, statistical power for this finding was lacking[5].

In the second case series, authors retrospectively reported their observations of six children fitted with a similar, dynamic elbow extension assist orthosis. Treatment protocols were poorly defined and reported. Similarly, the data from this series are poorly presented and suggest only that there may be a small clinical benefit in increasing range of motion at the elbow with such devices, but that it is highly dependent on the compliance of the patients and their families in using the prescribed devices[6].

Conclusions and Recommendations: Upper limb

The combined treatments of dynamic assist bracing and electrical stimulation appear to be effective in reducing upper extremity tone and improving dexterity and grip. However, the treatment effect has not been shown to endure once the modalities have been discontinued. The use of dynamic extension assist joints at the elbow as an adjunct to serial casting has been reported. Compliance with these bulky devices appears to impact their effectiveness.

WRIST/HAND

As with other body segments, function at the wrist and hand in children with quadriplegic and hemiplegic cerebral palsy is adversely affected by spasticity. Within this population, ulnar deviation, wrist flexion and thumb adduction posturing and contractures are commonly observed. The use of various hand splints have been suggested to prevent progressive contracture of these spastic muscles and to assist in motor control functions such as grip, pinch and dexterity.

Are there established benefits associated with the bracing of the wrist joint with static and/or dynamic splints in children with cerebral palsy?

Case studies and anecdotal reports of subjective changes associated with various wrist and hand orthoses have not been uncommon[1-6]. However, within the time parameters established for this review, there has only been a single clinical trial investigating the impact of wrist orthoses within this population[7] (See Table 9).

In this endeavor, ten children with hemiplegic cerebral palsy served as the experimental group while five age matched, non-affected children acted as controls. Grip strength, pinch strength, dexterity and surface EMG recordings at the muscles of the shoulder, elbow and wrist were recorded in three different conditions. These included a no-splint condition, a static splint condition, in which a custom static wrist-hand orthosis made in 15°-20° wrist extension was worn, and a dynamic splint condition in which a wrist-hand orthosis of spiral design was worn, fabricated in 15°-20° wrist extension, but allowing 30° of wrist movement.

Among the authors' key findings were the following: Children with hemiplegia demonstrated reduced grip, pinch and dexterity measures relative to controls. Children with CP demonstrated the greatest grip strength when wearing their dynamic splints. In contrast, static wrist splints compromised grip strength relative to the unbraced condition. The greatest values for lateral pinch were observed in the unbraced condition. Dexterity improved in both braced conditions, with the greatest improvements in the dynamic splint condition. Muscle activation was decreased in the static splint condition, suggesting a possible risk of disuse atrophy. Muscle activity at the shoulder was increased relative to controls in the static and no-splint conditions, suggesting compensatory proximal joint activity. In contrast, with dynamic splints, the muscle activity at the shoulder in children with CP was similar to that of controls.

Conclusions and Recommendations: wrist/hand

There appear to be trade-offs with the use of the various designs of wrist hand orthosis. It appears as though dynamic splints might facilitate better function in tasks that require grip and dexterity. However, splints may compromise pinch strength. While the use of static wrist-hand orthoses failed to suggest improvements in the functional outcomes studied, the trial did not evaluate the ability of such orthoses to prevent or delay contracture at the at-risk joints of the wrist and hand over time. While further inquiry is needed in this area, it would appear that decisions regarding the bracing of the wrist and hand should be made on a case by case basis, addressing the individual needs of each patient.

LYCRA GARMENTS

The theory behind the use of Lycra garments in children with cerebral palsy lies in the close relationship between proximal stability and distal control. Proponents of this intervention have suggested that such garments reduce tone in spastic muscles, decrease involuntary movement and improve trunk tone in children with postural hypotonia[1].

Are there established benefits with respect to upper extremity function associated with the use of Lycra garments in children with cerebral palsy?

Are there established benefits associated with the use of Lycra garments with respect to lower limb function in children with Cerebral Palsy?

There currently exists a small body of literature that has attempted to identify the potential benefits of the therapy, common deterrents to the therapy, and which populations may respond more positively to the therapy (See Tables 10 and 11). However, consensus conclusions are challenging to draw for several reasons.

The types of lycra garments used vary greatly from study to study, including "UPsuits[1]," "Camp lycra body suits[2]," "Camp full body suits[3]," "full suits, sleeved vests and gloves[4]," "total body," "shorts," "gloves," and "long-sleeved vests[5]," and "second skin upper limb garments[6]." Additionally, the outcome measures varied greatly across the various reports. Nicholson et al and Edmonson et al reported on vaguely defined "functional improvements" and "positive and negative effects," Nicholson et al and Rennie et al reported on everyday functional abilities as assessed with the Pediatric Evaluation of Disability Inventory or PEDI[7], Knox et al reported on gross motor abilities as assessed with the Gross Motor Function Measure or GMFM[8] and on function and movement quality of the upper limbs as assessed by the Quality of Upper Extremity Skills Test or QUEST[9], and Corn et al reported on the quality of upper limb movement as determined using the Melbourne Assessment[10]. Finally, sample sizes were both generally small, ranging from 2 to24 subjects, and extremely heterogeneous, including subjects with athetosis, dystonia, ataxia, hypotonia and spasticity, as well as subjects with quadriplegia, diplegia and hemiplegia. Functional improvements, when identified, were consistently very small and largely offset by challenges with the devices, including difficulties with donning and doffing, excessive heat and restrictiveness, difficulties with toileting and incontinence and respiratory compromise. Two of the three publications that reported on the usage of Lycra garments after the trials reported that only a single subject from each trial continued to use the modality[4-5].

Conclusions and Recommendations: Lycra

The use of Lycra garments in the management of cerebral palsy appears to be of some benefit in individual cases. Current literature suggests that hypotonic patients and patients with athetosis may derive the greatest benefit. However, these benefits were often found to be comparatively small and were often offset by other detrimental effects of the devices.

JOHNSTONE PRESSURE SPLINTS

Several decades ago, it was postulated that impulses from cutaneous receptors had a direct influence on motor neuron excitability[1]. It has been suggested that the application of neutral heat and pressure decrease the excitability of thermal and tactile neurons, thereby decreasing the excitability of motor neurons.

The application of these principles was first reported on clinical trials of adults with hemiplegia[1-3]. Decreased spasticity values were reported in these early trials.

Johnstone Pressure splints (JPS) have described as "transparent inflatable plastic pressure splints to support extremities in postures antagonistic to the spastic posture[4] (Kerem 01)." It has been suggested that they are used to 1) "stimulate proprioceptive and cutaneous receptors by application of deep pressure," 2) "provide required support for extremity stabilization during exercise," 3) "control combined motor patterns," and 4) "inhibit pathological reflexes." [2,4].

Are there established benefits associated with the use of Johnstone pressure splints in the management of children with cerebral palsy?

There is a small body of literature which has investigated the efficacy of these splints in children with Cerebral Palsy (See Table 12). The first of these was a randomized controlled trial in which thirty-four children with cerebral palsy underwent 3 months of physical therapy based on neurodevelopmental therapy (NDT). Seventeen of these children served as a control group. The remaining seventeen children, matched to the controls according to age and motor development levels, underwent the same physical therapy regimen, but with the addition of a defined set of JPS's, applied bilaterally for 20 minutes.

At the conclusion of the treatment period, authors reported significant improvements in lower extremity range of motion (ROM) in both groups with the ROM improvements of the treatment group being significantly higher than those of the controls. Further, while group means for spasticity values decreased in several tested muscles in the control group, these values decreased in all tested muscles in the treatment group. Finally, sensory function, as evidenced by latencies in somatosensory evoked potentials (SEP), appeared to improve in both groups, with the improvements of the treatment group reported to be significantly higher than those of the control group[4].

The second trial compared the clinical benefits derived from the use of JPS compared to those observed with the use of Botulinum Toxin A (BTX) in the medial hamstrings and hip adductors[5]. Both treatment groups received NDT exercises 3 times per week for 12 weeks. Outcomes measures were collected for both groups at the beginning and end of the treatment period. These included Gross Motor Function Measures, passive range of motion into hip abduction, modified Ashworth scale assessment of the spasticity in the hip adductors and the measured distance between the medial femoral condyles in maximum abduction. Relative to group means, statistically significant increases were reported in all outcome measures in both treatment groups. However, those in the BTX groups were consistently superior to those in the JPS group.

Conclusions and Recommendations: Johnstone Pressure splints

The use of JPS appears to have value as an adjunct to NDT physical therapy with respect to improvements in ROM, reduced spasticity and improved sensory function. However, because those benefits associated with JPS are only experienced during the comparatively brief treatment sessions themselves, these benefits do not appear to be as significant as those gained through the use of BTX injections.

LIMITATIONS AND CONCLUSIONS

The preceding review has a number of limitations which warrant further consideration. First of all, while the current literature provides some evidence regarding the efficacy of individual orthotic modalities on individual body segments, it provides little or no insight on the cumulative effect of multiple orthoses on the overall well being of the patient. The pervasive nature of this pathology, often affecting the head, trunk and multiple extremities, is such that the use of orthoses across every joint could be rationalized. Little has been done to elucidate a hierarchy of available benefits to provide guidance that might determine when the inconvenience, discomfort and/or expense of a given orthosis outweighs its benefits, particularly when multiple devices have already been prescribed or recommended. Further investigation along these lines would assist patients, families and rehabilitation professionals in deciding just how much to intervene.

Additionally, the modalities used in the management of persons with cerebral palsy continue to change. Interventions such as Botulinum toxin injections, neurodevelopmental therapies and dorsal rhizotomies are used with increasing frequency and acceptance. To the extent that these interventions alter the natural history of the pathology, they will impact the appropriateness of the various orthotic modalities. The potential benefits of spinal orthoses following dorsal rhizotomy or the use of orthoses to augment the effect ot Botox injections are two such examples. It is incumbent on the field at large to continue to recognize the effects of these rehabilitative modalities and the resultant implications and opportunities upon the field of orthotics.

Cerebral palsy is a broad diagnosis, with varying levels of associated disability. As such, recent attempts to further define subpopulations with respect to their needs and abilities, such as the Gross Motor Function Classification System and report upon the outcomes observed with a targeted modality upon various subpopulations has been highly informative. Future research should continue to report on outcomes with respect to clearly defined subpopulations, as these will provide better clinical guidance. Along these lines, continued investigation is needed to define which outcome measures are more appropriate in evaluating the various subpopulations.

With respect to the orthotic management of the cerebral palsied patient, the clinical treatment plan should be based on the collective goals of the medical professionals, the family and the patient. Such goals might include the prevention of initial or aggravated contracture, the facilitation of improved balance, and improved function of targeted extremities. Close communication with other health care providers is necessary to ensure that the provided treatments are complimentary and consistent with the established goals. An awareness of the limited findings of clinical trials to date can assist practitioners in recommending more affective orthotic approaches.

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Table 1: Do spinal orthoses improve posture, balance and/or associated control of the head, neck or extremities in children with cerebral palsy when compared to unbraced conditions?

Author	# of subjects	Type of Orthosis	Research Design	Evidence Level	Key Findings
Letts M et al, 1992	55 (41)*	"Soft Boston" TLSO	Case Series, Convenience sample, Qualitative study (caregiver questionnaire)	4	- improved head and trunk control - improved sitting stability - improved general appearance - improved patient handling
Terjeson T et al, 2000	86 (57)**	Custom molded polypropylene or polyethylene TSLO	Case Series, Convenience sample, Qualitative study (caregiver interview)	4	- improvement of sitting balance - better head/neck control - improved sitting function
Verkerdy Z et al, 2007	42	TLSO with non-rigid SIDO frame***	Case Series, Convenience sample, Controlled before-and-after trial, Qualitative study (family questionnaire)	4	- improved feeding - improved trunk posture - improved head posture - improved posture of the extremities

* 55 patients with neuromuscular scoliosis a varying etiology, 41 patients with quadriplegic cerebral palsy
** 86 patients in the case series, 57 of which were used to generate parent/caregiver feedback
*** essentially a posterior TLSHO which allows flexion and limited rotation of the trunk

Table 2: Do spinal orthoses have a negative impact on respiratory function in children with cerebral palsy compared to unbraced conditions?

Author	**# of subjects**	**Research Design**	**Protocols**	**Evidence Level**	**Key Findings**
Letts M et al, 1992	36 (55)*	Case Series, Convenience sample, Controlled before-and-after trial (A-B)	- Oxygen saturation levels measured before and after orthosis application	4	- no appreciable differences in oxygen saturation levels before and after the application of the orthosis.
Leopando M et al, 1999	12	Case Series, Convenience sample, Controlled before-and-after trial (A-B-C)	-Pulmonary mechanics of subjects were obtained in 3 postions -Sitting with TLSO -Sitting without TLSO -Supine without TLSO	4	- no appreciable differences in pulmonary resistance, compliance, tidal volume, minute ventilation, work of breathing, oxygen saturation or end-tidal CO_2 tension were measured with the subjects seated both with and without the orthosis and in the supine position without the orthosis. - Subjects tended to have an increased work of breathing when seated without the orthoses compared to the other two conditions.

*55 subjects were treated with the soft Boston orthosis, in-brace oxygen saturation levels were only measured in 36.

Table 3: Do spinal orthoses prevent or delay curve progression compared to natural history in children with cerebral palsy?

Author	# of subjects	Type of orthosis	Research Design	Evidence Level	Key Findings
Miller A et al, 1996	21(43)*	Wilmington, custom-molded	Cohort Study, Convenience sample (unmatched controls)	2b	- the use of the orthoses (mean duration = 67 months) was found to have no statistically significant effect on curve magnitude or on the rate of curve progression relative to unbraced, unmatched controls.
Olafsson Y et al, 1999	38(90)**	Prefabricated Boston-type TLSO	Case Series, Convenience sample	4	- bracing was found to be successful (spinal curves failed to progress more than 10° during the observation period) in 9/37 (24%) patients with cerebral palsy. - success rates improve to 7/27 (26%) when patients with athetosis are excluded
Terjesen T et al, 2000	86	Custom molded polypropylene or polyethylene TSLO	Cohort Study (retrospective), Convenience sample	2b	- in-brace curve progression rates were similar to reported natural history values (4.5°/year). - 16% of those subjects under the age of 15 had curve progression rates < 1°/year. - 64% of those subjects 15 y/o or older had curve progression rates < 1°/year. - neither the Cobb angle at initiation of bracing, scoliotic curve type nor degree of pelvic obliquity were predictive of successful bracing outcomes. - initial correction in the orthosis was found to significantly influence in brace curve progression rates.

* Twenty one subjects were received TLSOs, twenty-two subjects served as controls

** Ninety subjects with neuromuscular scoliosis, thirty-eight of which had spastic quadriplegia

Table 4: Do hip orthoses prevent progressive hip subluxation compared to natural history in children with cerebral palsy?

Author	# of subjects	Research Design	Protocols	Evidence Level	Key Findings
Graham H et al, 2008	47(91)*	Randomized Controlled Trial	- Treatment: BTX injections to the adductor and medial hamstring muscles every six months for three years, SWASH hip orthosis prescribed, to be worn for 6 hrs/day. - Control: No BTX or orthosis	1b	- Progressive hip displacement observed in both groups. - Very slight reduction in displacement rates (-1.4 % per year) was observed in the treatment group. - Clinical significance was considered questionable given the associated costs of treatment.

* forty-seven subjects were treated with hip orthoses, forty-four additional children served as matched controls

Table 5: Do daytime hip orthoses improve gross and/or fine motor function in children with cerebral palsy compared to unbraced controls?

Author	# of subjects	Research Design	Protocols	Evidence Level	Key Findings
Boyd R et al, 2001	19(39)*	Randomized Controlled Trial	- Treatment: BTX injections to the adductors and medial hamstrings every 6 months for one year, prescription of SWASH hip orthosis to be used 6-8 hours per day, regular physical therapy. - Control: Regular physical therapy only, without BTX or hip orthosis	1b	- Both groups demonstrated improvements in total GMFM scores. - No additional treatment effects were found for the treatment group. - The GMFM may not have been sensitive to changes in the studied populations.

* nineteen subjects were treated with the hip orthosis, twenty additional subjects served as controls

Table 6: Do daytime orthoses increase muscle activity in hip adductors in children with cerebral palsy compared to unbraced muscle activity?

Author	# of subjects	Research Design	Protocols	Evidence Level	Key Findings
Embrey D et al, 2006	10	Case Series, Controlled before-and-after trial (A-B)	- surface electromyography data collected from the hip adductor and hip abductor muscles during standing, walking and sitting, both with a without the use of the SWASH orthosis	4	- No statistically significant differences were observed with and without the use of the SWASH orthosis relative to surface electromyography data obtained from the hip adductor and hip abductor muscles in either sitting, standing or walking conditions.

Table 7: Do the combined modalities of dynamic assist upper extremity orthoses and functional stimulation of antagonist muscle groups decrease upper limb spasticity and/or improve upper limb function in children with cerebral palsy relative to controls?

Author	# of subjects	Research Design	Protocols	Evidence Level	Key Findings
Shecker F et al, 1999	19	Case Series, Convenience sample, Interrupted time series trial (A-B-B-B . . .)	- Electrical stimulation: applied over the wrist and finger extensors and triceps - Dynamic orthosis: wrist/hand: static support in wrist extension (10° short of end range) with an adjustable pan splint which locked the PIP and DIP joints in extension while allowing dynamic extension of the MCP joints - Dynamic orthosis: elbow: forearm positioned in maximum supination, dynamic elbow extension assist joint. - Electrical Stimulation and Dynamic orthosis were used simultaneously for 1 hour/day - Static wrist/hand orthosis used at night to maintain ROM	4	- Universal improvements observed according to Zancolli's classification of deformity, suggestive of improved wrist and finger function.
Ozer K et al, 2006	8(24)*	Randomized controlled trial	- Electrical stimulation: as per Shecker et al. - Dynamic orthosis: as per Shecker et al. - Group 1: Two 30-minute sessions of electrical stimulation to antagonist extensors per day. Nocturnal static brace. - Group 2: Two 30-minute sessions of dynamic bracing per day. Nocturnal static brace. - Group 3: Two 30-minutes sessions of electrical stimulation to antagonist extensors during concurrent use of dynamic bracing. Nocturnal static brace.	1b	- Group 3 demonstrated significant improvements in dexterity. - These improvements lasted only 1 month post treatment - Group 3 demonstrated a steady increase in grip strength during treatment which subsequently declined post treatment - Group 3 demonstrated the greatest improvements in Zancolli's classification system, but regressed to pretreatment values 3 months after treatment.

* Eight subjects were treated with the combined treatments, eight subjects were treated with electrical stimulation alone, eight additional subjects were treated with the dynamic orthosis alone.

Table 8: Do dynamic assist upper extremity orthoses maintain upper extremity range of motion more effectively than static splinting modalities in the treatment of patients with cerebral palsy?

Author	# of subjects	Research Design	Protocols	Evidence Level	Key Findings
Yasukawa A et al, 2003	3	Cohort study	-Pre-trial: BTX injections to biceps and brachioradialis bilaterally followed by 4 sequential serial casts until gains in range of motion had plateaued. Subjects fit with two types of "maintenance orthosis," one on each arm, to be worn 4+ hours daily. -Bivalve long arm cast -Ultraflex dynamic elbow extension assist elbow wrist hand orthosis.	2b	- Increases gained in elbow range of motion were found to be temporary - As the BTX wore off, each subject had difficulty wearing either maintenance orthosis, requiring replacement bivalve casts and adjustments to the orthoses. - Ultraflex orthoses were found to maintain elbow ROM longer than the bivalve cast. - Subjects' parents indicated a preference for the ultraflex orthosis over the bivalved cast.
Yasukawa A et al, 2008	6	Case series, Convenience sample	- Loosely defined: - subjects used a combination of oral baclofen, BTX injections or no medications - All subjects underwent serial casting at the wrist to improve ROM prior to the use of the orthosis - Orthoses were elbow-wrist-hand orthoses with static wrist joints and dynamic elbow extension assist joints	4	- Increases in passive range of motion in the upper extremity were correlated with compliance to the wearing scheduled of the orthosis. - Subjects and caregivers consistently reported that the brace was not used at night because the discomfort interfered with sleeping.

Table 9: Are there established benefits associated with the bracing of the wrist joint with static and/or dynamic splints in children with cerebral palsy?

Author	# of subjects	Research Design	Protocols	Evidence Level	Key Findings
Burtner et al, 2008	10(15)*	Case Series, Convenience Sample, Controlled before-and-after trial (A-B-C)	- Data collected during grip, pinch and peg-board tests in each of 3 conditions: - "static splint" immobilizing the wrist in 15°-20° wrist extension. - "dynamic splint" of spiral design, fabricated in 15°-20° wrist extension, but allowing 30° of wrist movement - "no-splint"	4	- children with hemiplegia demonstrated reduced grip, pinch and dexterity measures relative to controls. - children with CP demonstrate greater grip strength with dynamic splints. - static wrist splints compromised grip strength in children with CP compared to the unbraced condition. - the greatest values for lateral pinch were observed in the unbraced condition. - dexterity improved in both braced conditions, with the greatest improvements in the dynamic splint condition. - muscle activation is decreased in the static splint condition suggesting a possible risk of disuse atrophy - muscle activity at the shoulder was increased relative to controls in the static and no-splint conditions, suggesting compensatory proximal joint activity. - with dynamic splints, muscle activity at the shoulder in children with CP was similar to that of controls

* Ten subjects with CP, five age matched control subjects

Table 10. Are there established benefits with respect to upper extremity function associated with the use of lycra garments in children with cerebral palsy?

Author	# of subjects	Research Design	Protocols	Evidence Level	Key Findings
Blair et al, 1995	24	Case series, Convenience Sample, Interrupted time series trial (A-B-A-B), Qualitative Study.	- Subjects used the garments (UPsuits) according to the following schedule: - Weeks 1-3: without - Week 4: lycra introduced - Weeks 5-7: with lycra - Weeks 8-10: without - Weeks 11-16: with - Videos taken at the conclusion of weeks 3,7,10 and 13 in the corresponding condition	4	- Observers (parents, teachers therapists) tended to view the garments as having a positive effect on such dynamic functions as crawling, walking and both fine and gross motor function of the upper extremities. - Similar trends were reported with respect to static functions such as sitting, standing, the reduction of involuntary movement and maintaining an erect head posture. - Blinded video raters rated improvements in postural stability and the quality of upper-limb movement when subjects were wearing the lycra garments. - Researchers identified respiratory compromise and intractable peripheral cyanosis as medical contraindications for the lycra suits.
Edmonson et al, 1999	15	Case Series, Convenience Sample, Controlled before-and-after trial (A-B) as well as Qualitative Study	- Lycra garments (body suits) worn for at least 6hrs/day for 12 months.	4	- All children showed some functional improvements according to the authors' unspecified assessments of gross and fine motor function. - Parent responses were variable, citing both benefits and problems. - 7/15 subjects continued to use the suit after the trial.
Nicholson et al, 2001	12	Case Series, Convenience Sample, Controlled before-and-after trial (A-B) as well as Qualitative Study	- Lycra garments worn for at least 6 hours/day for 6 weeks. - Garments used included "full suit," "sleeved vest" and/or gloves - PEDI assessments made at the beginning and the conclusion of the trial. - Five subjects were selected for motion analysis of the trunk and upper extremities 8 weeks after receiving the garments.	4	- All subjects made mild improvements according to the PEDI following the 6 week wear period. - Improved proximal stability was observed in 4/5 children receiving motion analysis. - Garments increased smoothness of movement in children with athetosis and ataxia, but made movements more jerky in subjects with spasticity. - All children had problems wearing the garments, including toileting difficulties and urinary incontinence. - The parents of only 1/12 subjects wanted to continue use of the garment.

Table 10. Are there established benefits with respect to upper extremity function associated with the use of lycra garments in children with cerebral palsy? (continued)

Author	# of subjects	Research Design	Protocols	Evidence Level	Key Findings
Knox et al, 2003	8	Case series, Convenience sample, Controlled before-and-after trial (A-B) as well as Qualitative Study.	- Lycra garments (total body, shorts, gloves, and/or long-sleeved vest) worn 4 hrs/day for 4 weeks. - GMFM and QUEST outcomes assessed before and after the intervention. - Questionnaires completed by parents and subjects at the conclusion of the trial.	4	- 3/8 subjects withdrew from the trial citing discomfort of the suit, restrictiveness and donning difficulties. - Of the 4 subjects who remained in the trial, all showed improvements in either GMFM (3/4) or QUEST (2/4). - Observed benefits included improved sitting balance, grasping of objects and self-feeding. - Perceived disadvantages included the time required to don and doff, the heat and restrictiveness of the garment and the reduction of certain functions. - 1/8 subjects wore a second garment once the originals were outgrown.
Corn et al, 2003	2(4)*	Case series, Convenience sample. Interrupted time series trial (B-A)	- Subjects with CP had worn "second skin" upper limb garments for at least 12 months prior to the trial - Quality of upper limb movement established using the Melbourne Assessment.	4	- No changes of statistical significance were seen between the two conditions. - Trends were towards reduced performance with the "second skin." - Data collection for the 2 subjects was described as "problematic" due to compromisingly low assessment scores and required splint modifications.

* Four subjects were included in the trial. Only two of these were diagnosed with cerebral palsy.

Table 11. Are there established benefits associated with the use of lycra garments with respect to lower limb function in children with Cerebral Palsy?

Author	# of subjects	Research Design	Protocols	Evidence Level	Key Findings
Rennie DJ	7(8)*	Case Series, Convenience Sample, Controlled before-and-after trial (A-B) as well as Qualitative Study	- Lycra garments worn for at least 6 hours/day for 6 weeks. - Garments were full body suits, extending from the ankles to the neck and from the neck to the wrist joints. - PEDI assessments made at the beginning and the conclusion of the trial. - Subjects underwent motion analysis 8 weeks apart with root mean square error (RMSE) values between trials used as a measure of proximal and distal joint stability. - Parents completed a questionnaire on practicality and compliance of using the garment.	4	- 4/7 subjects showed reduced RMSE values at the pelvis with the lycra garments, suggestive of improved proximal stability. - 3/7 subjects showed reduced RMSE values at the hip, knee and ankle joints with the lycra garments, suggestive of improved distal stability. - With respect to proximal and distal stability, a subject with athetosis demonstrated the greatest improvements with the garments. - Other subjects demonstrated increased RMSE values with the garments, suggestive of compromised proximal and distal stability. - There were no statistically significant changes in RMSE before and after use of the garment. - 5/7 subjects improved in one aspect of the PEDI. - There were no significant changes in self-care, mobility, social function or the level of caregiver assistance required when the group was looked at as a whole. - Parents generally found the suits easy to don and no respiratory problems were reported. - 5/7 subjects experienced toileting difficulties. - The parents of 6/7 subjects indicated that they would not consider using lycra garments in the future, citing problems with toileting, excessive heat and discomfort. The parents of the remaining subject were undecided.

* Seven of the eight subjects in the study had cerebral palsy, a remaining subject had Duchenne muscular dystrophy.

Table 12. Are there established benefits associated with the use of Johnstone pressure splints in the management of children with cerebral palsy?

Author	# of subjects	Research Design	Protocols	Evidence Level	Key Findings
Kerem et al, 2001	17(34)*	Randomized Controlled Trial	- Control: Neurodevelopmental Therapy, five days a week for three months. - Treatment: As with controls with the additional use of various Johnstone pressure splints, applied bilaterally for 20 minutes each session.	1b	- Significant improvements in lower extremity range of motion (ROM) were seen in both groups - ROM improvements of the treatment group were significantly higher than those of the controls - Group means for spasticity values decreased in several tested muscles in the control group and in all tested muscles in the treatment group. - Sensory function, as evidenced by latencies in somatosensory evoked potentials (SEP), appeared to improve in both groups. - Improvements in SEP were significantly higher in the treatment group relative to those of the control group.
Hazneci et al, 2006	21(43)**	Randomized Controlled Trial	- BTX treatment group: 300 IU of Botox injected into adductors and medial hamstrings. NDT exercises 3 days/week for 12 weeks - JPS treatment group: long leg JPS applied 3 days/week during the same NDT exercises for 12 weeks.	1b	- At the conclusion of the treatment period, statistically significant improvements were seen in the group means of both treatment groups with respect to: - gross motor functions measurements (GMFM), - passive hip abductions ROM, - spasticity (as measured by the modified Ashworth scale) and - the measured distance between the medial femoral condyles in maximum abduction. - Improvements in the BTX groups were superior to those in the JPS group across all measured variables.

* Seventeen subjects were treated with Johnstone pressure splints, seventeen other children served as controls

** Twenty-one children were treated with Johnstone pressure splints, twenty-two others served as controls

A REVIEW OF THE EFFECTIVENESS OF LOWER LIMB ORTHOSES USED IN CEREBRAL PALSY

Roy Bowers Dip. Pros. Orth. Senior Lecturer in Orthotics
&
Karyn Ross BSc Prosthetist & Orthotist
National Centre for Prosthetics and Orthotics,
University of Strathclyde, Glasgow, UK

To produce this review, a systematic literature search was conducted for relevant articles published in the period between the date of the previous ISPO consensus conference report on cerebral palsy (1994) and April 2008. The search terms were "cerebral and pals* (palsy, palsies), "hemiplegia", "diplegia", "orthos*" (orthoses, orthosis) orthot* (orthotic, orthotics), brace or AFO. Papers were selected for review if they addressed the use of lower limb orthoses in cerebral palsy. Papers relating to adult onset pathology were rejected. Papers relating to the direct application of hip orthoses were excluded as this area is addressed in a separate review on the effects of orthoses on the hips, spine and upper limbs. Abstracts were rejected if their content was subsequently located in full research papers. Only English language papers were included. Databases searched were EMBASE (ovid), Science Direct, social services abstracts, psychINFO, Medline (ovid), APAIS Heath (informit), AMI, Cinahl, PubMed, Recal, the NHS Scotland e-library and Google Scholar. The literature review on orthotic management of cerebral palsy by Morris [1] was also consulted.

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As a result of reading and reviewing the literature, a number of questions were developed and answered according to the best available evidence.

- Do lower limb orthoses influence the temporal and spatial parameters of gait?
- What are the effects of orthoses on the ankle and foot?
- What are the indirect effects of orthoses on the knee?
- What are the effects of orthoses on the hip?
- What are the effects of orthoses on the pelvis?
- What effect do orthoses have on the metabolic and cardiopulmonary cost of walking?
- What effect do orthoses have on muscle activity?
- What effect do orthoses have on muscle length?
- How do orthoses affect function and ability?
- What are the benefits of tuning ankle-foot orthoses?
- Can lower limb orthoses influence the upper limb?
- What are the perceptions of users and parents regarding orthotic treatment?

A number of abbreviations are used in this review

ADL	Activities of daily living
AFO	Ankle-foot Orthosis
BTA	Botulinum Toxin A
CP	Cerebral Palsy
DAFO	Dynamic AFO (AFO with tone reducing features)
DST	Double Support Time
EEI	Energy Efficiency Index
EMG	Electromyography
FRAFO	Floor Reaction AFO (synonymous with GRAFO)
GRAFO	Ground Reaction AFO (synonymous with FRAFO)
GRF	Ground Reaction Force
GMAE	Gross Motor Ability Estimator
GMFCS	Gross Motor Function Classification System
GMFM	Gross Motor Function Measure
GMPM	Gross Motor Performance Measure
HAFO	Hinged AFO (also known as Articulated AFO)
MAS	Modified Ashworth Scale
PBS	Pediatric Balance Scale
PCI	Physiological Cost Index
PEDI	Pediatric Evaluation of Disability Inventory
PLS	Posterior Leaf Spring AFO
PODCI	Pediatric Outcomes Data Collection Instrument
ROM	Range of Motion
SAFO	Solid AFO (or rigid AFO)
SD	Standard Deviation
SMO	Supramalleolar AFO
SST	Single Support Time
STS	Sit to Stand
SWOC	Standardised Walking Obstacle Course
VVG	Video Vector Generator

Where appropriate, evidence from the reviewed scientific papers has been presented under separate headings reflecting the nature of the participants in the study in question. Studies that included a mixed cohort of subjects, irrespective of whether separate analysis was conducted, are included in the "mixed" section.

DO LOWER LIMB ORTHOSES INFLUENCE THE TEMPORAL AND SPATIAL PARAMETERS OF GAIT?

1. VELOCITY

Mixed

The majority of studies [2-7] conducted on mixed groups of subjects (i.e. hemiplegia and diplegia) reported an increase in velocity using a variety of AFOs, including SAFO, HAFO, PLS, DAFO, "standard polypropylene" and "wrap-around polyethylene". In one study where velocity reportedly increased [4] the type of AFO was not described. The increase in velocity was statistically significant in most studies [2-4, 6] but not in all [7]. While in one study [5] velocity increased in both

hemiplegic and diplegic subjects using SAFOs and DAFOs, the increase only reached statistical significance in the diplegic group. This study was one of only two [3, 5] which classified subjects by their GMFCS level. The other study [3] reported a statistically significant increase in all subjects with GMFCS I, II and III. Interestingly, this improvement was attributed to increased step length rather than cadence, which decreased slightly. One study [2] investigated the effect of Botulinum Toxin A (BTA) on the gait of predominantly spastic children whose velocity had increased when using PLS. No further increase was reported after BTA. A single study [8] investigating the effects of DAFOs and SAFOs found no change in velocity.

Hemiplegia

The majority of studies investigating the effects of AFOs on velocity in hemiplegic subjects reported statistically significant increases using a variety of designs [9-14]. Two studies [10, 11] reported a significant increase using HAFOs, while others found similar results using SAFOs [9, 12]. It is interesting to note that in the study by Brunner [12] velocity increased more when using a flexible AFO than using a SAFO. However, the authors report that with the SAFO the heel lost contact with the floor too early, suggesting inadequate tibial inclination in the orthosis, and a need for tuning (see Tuning AFOs for optimum function). It is also interesting to note that the "SAFOs" in the study by Thompson [9] were clearly not rigid. Increases in velocity have also been reported using PLS, Dual Carbon Fibre Spring AFO, and Orteam® AFO [13]. In one study [14] velocity increased significantly when using PLS and a Dual Carbon Fibre Spring AFO compared to the barefoot condition, but not compared to gait with shoes. A further study investigating the effects of HAFO, SAFO, PLS [15] found no significant change in velocity using orthoses when compared to barefoot gait.

Diplegia

Only one study [16] reported a significant increase in velocity in diplegic subjects using AFOs. This study investigated the effects of SAFOs, although as in the study by Thompson [9] the SAFOs were clearly not rigid. The majority of studies on diplegic children [17-24] reported no significant increases in velocity with a variety of AFO designs including SAFOs, SMOs, DAFOs, HAFOs and PLS. One of these studies [17] reported that gait with HAFOs was significantly slower in comparison to PLS. One study [25] reported a decrease in velocity with AFOs although this was not statistically significant. A further study [21], found a decrease in velocity when comparing SAFOs with shoes only, although again this was not statistically significant. One abstract [24] reported increase in velocity in two children, who also used assistive devices for ambulation, being faster in SMOs than HAFOs.

Conclusions

The majority of investigations into the effect of orthoses on velocity in hemiplegic gait report significant increases using orthoses. By contrast, the evidence for orthoses increasing velocity in diplegic subjects is equivocal, with some studies showing little or no effect on velocity. There is some evidence that the use of shoes alone can increase velocity. It is therefore important that future research isolates the effect of orthoses from that of footwear.

2. CADENCE

Mixed

Most studies on mixed subjects [3, 5, 6, 8, 26] report a decrease in cadence using a variety of AFO designs, including SAFO, DAFO, HAFO, PLS, standard polypropylene and wrap around polyethylene. However only three of these studies [6, 8, 26] found this decrease to be statistically significant in both the hemiplegic and diplegic subjects. While in the study by White [3] the decrease in cadence for the complete group was statistically insignificant, subgroup analysis revealed statistically significant decrease for the subjects with GMFCS levels I and II. Subjects with GMFCS Level III actually showed a small but statistically insignificant increase. There was no difference in the decrease in cadence between patients using SAFO and HAFO. One study [5] found a statistically significant reduction in cadence only in the hemiplegic group (GMFCS level 1), with no reported decrease in the diplegic subjects, while another [4] reported no change in cadence at all. A further study [27] investigated the effect of shoes alone on the gait of mixed subjects, reporting no significant change in cadence. A further statistically significant decrease in cadence was reported following BTA in patients described only as "predominantly spastic", using PLS [2].

Hemiplegia

A number of studies [9, 11-13, 15] report a significant decrease in cadence in hemiplegic subjects when using varying designs of AFOs, including HAFO, SAFO, PLS, flexible AFO, Dual Carbon Fibre Spring AFO, and an Orteam® AFO. As previously noted, the SAFOs in the study by Thompson [9] were clearly not rigid. In the study by Brunner [12] cadence decreased more with a SAFO than a with a flexible AFO, however the early heel rise observed with the SAFO may be indicative of inadequate tibial inclination. One study [14] reported a significant reduction in cadence when walking with shoes only in comparison to barefoot. Further non-significant reduction was observed using PLS and Dual Carbon Fibre Spring AFO compared to shoes only. It is also interesting to note that in this study. One study [10] reported no significant change in cadence with HAFO use.

Diplegia

Most studies into the effects of HAFO, SAFO, PLS, DAFO and SMO on cadence in diplegic subjects report no significant effect [16, 18-23]. Although Abel [16] found no change in cadence using SAFOs, it should be noted that the SAFOs in question could not truly be regarded as solid ankle design, as some dorsiflexion was allowed by buckling. However, statistically significant reduction in cadence was reported by Buckon [17] using HAFO, SAFO and PLS. Investigating the effect of SAFOs and SMOs vs. Shoes, Carlson [18] found the only significant difference was between shoes and SMOs.

Conclusions

The majority of studies that investigated the effect of orthoses on cadence in subjects with hemiplegia reported significant reductions when using a variety of AFOs**.** However, as with the findings relating to velocity, the effects of orthotic intervention on cadence in diplegia are more equivocal. While two studies on diplegic subjects reported reduction in cadence using orthoses, the majority found no effect. There is some suggestion that the use of shoes alone has a beneficial effect on cadence. It is therefore important that future research isolates the effect of the orthosis from that of

the footwear. There a suggestion that the use of BTA may further improve the effects of AFOs on cadence.

3. STEP LENGTH

Mixed

The majority of papers [2, 3, 5, 6, 28] investigating the effects of AFOs on step length in mixed groups of hemiplegic and diplegic subjects report statistically significant increases using a variety of orthoses (HAFO, SAFO, PLS, SMO, standard polypropylene and wrap-around polyethylene). It should be noted that in the studies by White [3] and Hayek [5] there was a statistically significant increase on both sides in the hemiplegic group, although the gait remained asymmetrical. De Groot [27] investigated the effect of shoes alone on gait. While no details of the footwear characteristics are reported, step length increased with the shoes only intervention. Huenaerts [2] investigated the effect of PLS before and after BTA in participants described only as "predominantly spastic". While the PLS alone increased step length, further increase was observed after the BTA.

Hemiplegia

All studies into the effect of orthoses on step length in hemiplegic subjects reported statistically significant increases [9, 11-15, 29]. These improvements were noted using HAFO, SAFO, DAFO, PLS, flexible AFO, Dual Carbon Fibre Spring AFO, and Orteam® AFO. It should be noted that the SAFOs used in the study by Thompson [9] were not rigid. In the study by Brunner [12] step length increased more when using a flexible AFO than using a SAFO. As previously observed, the early heel rise seen with the SAFO may suggest inadequate tibial inclination in the orthosis, and a need for tuning.

Diplegia

Buckon [17] described a trial investigating the effects of HAFOs, SAFOs and PLS on diplegic gait, reporting that step length increased with all orthoses ($p<0.005$).

Conclusions

The use of AFOs has been demonstrated to increase step length in both hemiplegic and diplegic subjects. Some of this effect may be due to the wearing of shoes, which have also been demonstrated to increase step length when used without orthoses.

4. STRIDE LENGTH

Mixed

All studies into the effect of AFOs on stride length conducted on mixed groups of hemiplegic and diplegic subjects report statistically significant increases [3-5, 26, 30]. Improvements were noted using SAFO, HAFO, DAFO, PLS, standard polypropylene, wrap around polyethylene and Klenzak AFOs. In the study by White [3] each of the subgroups investigated (SAFO, HAFO, hemiplegia, diplegia, GMFCS levels I, II, and III) showed statistically significant improvement. De Groot [27] investigated the effect of shoes alone on mixed subjects, reporting statistically significant increase in stride length simply by wearing shoes.

Hemiplegia

Statistically significant increases in stride length were reported in all studies investigating the effects of AFOs on subjects with hemiplegia [10-12, 15, 29].

Orthoses used included SAFO, HAFO, DAFO, PLS, and flexible AFO. In the study by Brunner [12] the same test orthosis was used in both trials, the SAFO having had its flexibility increased by removal of an anterior wedge of material. While both orthoses increased stride length, the increase was more pronounced in the flexible orthosis. This might suggest that the SAFO was inadequately inclined, and needed tuning (while the authors stated that equinus was accepted in cases where dorsiflexion was limited, they did not make it clear whether any wedging was added to the SAFOs to compensate for this). Additionally, the use of flexible AFOs (or free-dorsiflexion HAFOs) in the presence of a plantarflexion contracture is contraindicated, making the results questionable.

Diplegia

Statistically significant improvement in stride length was reported in a number of studies [16-19, 23, 31] using SAFO, HAFO, PLS, SMO and DAFO. In the study by Buckon [17] the SAFOs investigated were clearly not rigid and allowed some dorsiflexion as they deflected. The study by Rethlefsen [20] investigating the effects of SAFO and HAHO reported no change in stride length. Smiley [21] investigated the effects of SAFOs, HAFOs and PLS on the gait of children with spastic diplegia. While stride was slightly increased in the HAFOs and the PLS, it was slightly reduced in the SAFOs, possibly because of inadequate tibial inclination in the orthosis. None of the differences in stride length were significant. Kornhaber [24] investigated the effects of SMOs and HAFOs on spastic diplegic gait finding increase in stride length only in those who wore SMOs and used assistive devices for ambulation. Crenshaw [32] investigated the effects of tone reducing features on the gait of diplegic children. Subjects were tested wearing shoes only, a HAFO, a HAFO with tone-reducing features, and a SMO with tone-reducing features. Increase in stride length was only significant in the tone reducing HAFO.

Conclusions

The majority of papers that investigated the effect of orthoses on stride length reported improvement as a result of various types of AFO intervention. In a few cases the results using SAFOs were not as good as using AFOs that allowed dorsiflexion. This may indicate that the SAFO is resisting tibial advancement at too early a stage in stance, adversely affecting stride length. When using a stiff AFO, the issue of tuning the orthosis is very important, if unacceptable resistance to tibial progression is to be avoided. It is of interest that the SAFOs that were somewhat flexible produced results comparable to HAFOs or PLS, although such buckling should be considered undesirable as it will compromise triplanar control of the foot.

5. SINGLE SUPPORT

Mixed

White [3] reported statistically significant increase in single support time (SST) in a mixed group of patients using four AFO variations (SAFO, HAFO, standard polypropylene and wrap around polyethylene). All of the subgroups (SAFO, HAFO, hemiplegia, diplegia, GMFCS I or II, and GMFCS III) showed statistically significant increase in SST. The more involved subgroups (i.e. those with diplegia and those with GMFCS level III) demonstrated greater improvement. Interestingly, in the hemiplegic group, the affected and unaffected leg demonstrated the same increase in SST.

Hemiplegia
Balaban [10] reported improvement in SST using a HAFO when treating hemiplegic subjects. Brunner [12] investigated the effect of SAFO and a flexible AFO (this was the SAFO with an anterior wedge removed) reporting that the SAFO significantly increased SST, but there was no change with the flexible AFO. Romkes [11] found no significant change in SST using a HAFO.

Diplegia
Abel [16] improved SST using SAFOs (although the AFOs buckled under load).

Conclusions
The duration of single support appears to be increased in both hemiplegic and diplegic subjects by the use of a variety of AFO designs.

6. DOUBLE SUPPORT

Mixed
Hayek [5] compared SAFOs and HAFOs to barefoot in a mixed group of subjects who were examined in the orthosis they habitually used, rather than being fitted with both types of AFO. Double support time (DST) decreased in the diplegic group, while in the hemiplegic group, the number of patients achieving symmetry in the initial double support period of the gait cycle increased with AFO use.

Hemiplegia
While Romkes [11] found no significant change in DST in hemiplegic subjects using a HAFO, Balaban [10] reported significant decrease in DST with a HAFO on a similar patient group. Surprisingly, Brunner [12] reported an increase in double support time with a SAFO and a flexible AFO (SAFO > flexible FAFO). An explanation for this unexpected increase may lie in the fact that while an equinus position in the AFOs was accepted where dorsiflexion was limited, it is unclear whether any wedging was added to the SAFOs to compensate for this. Additionally, the presence of a plantarflexion contracture should be regarded as a contraindication to the use of flexible AFO allowing dorsiflexion.

Diplegia
Abel [16] reported that DST was decreased in diplegic subjects using a SAFO, although the orthosis in question allowed dorsiflexion by buckling. Rethlefsen [20] reported that DST was significantly longer using HAFOs than in SAFOs or shoes alone.

Conclusions
What little evidence there is appears to support the hypothesis that orthoses, particularly SAFOs can reduce DST, but suggests that to do so they must be well aligned.

CONCLUSIONS RELATING TO ALL TEMPORAL AND SPATIAL PARAMETERS

In general, the available evidence suggests that orthoses can have positive effects on all temporal and spatial parameters of gait, i.e. velocity, cadence, step length, stride length, single and double support.

WHAT ARE THE EFFECTS OF ORTHOSES ON THE ANKLE AND FOOT?

1. ANKLE KINETICS

Mixed

Ounpuu [33] found that the peak power-generating capabilities of the ankle were reduced when wearing a PLS, finding that more mechanical energy was absorbed during midstance and less produced during terminal stance. The authors concluded that while the PLS improved ankle function, it did not store mechanical energy, or return this in the propulsive phase of gait. Molenaers [6] also reported that ankle power at push-off was decreased with PLS use. Kirkeide [8] reported a significant decrease in forefoot force with DAFOs compared to barefoot, but a significant increase with a SAFO compared to shoes. Molenaers [34] investigated the effect of BTA combined with orthotic management and short leg casting, reporting an increase in power generation at ankle at push-off with all regimes.

Hemiplegia

Balaban [10] reported a significant decrease in ankle power in stance with a HAFO. Buckon [15] investigated the effects of a HAFO, a SAFO and a PLS, reporting significant decrease in power with all orthoses. Desloovere [14] investigated push-off using "identically tuned" PLS and dual carbon fibre spring AFO. The dual carbon fibre spring AFO allowed significantly better power generation in pre-swing while the PLS significantly improved power absorption in loading response. Romkes [29] reported significant reduction in power absorption with a HAFO and a DAFO, but no change in power generation.

Diplegia

Hassani [35] compared HAFOs and DAFOs to barefoot, finding that both orthoses produced significant differences in peak ankle plantarflexion moment, but no significant difference in power. Crenshaw [32] investigated the effects of HAFOs, HAFOs with tone reducing features and SMOs. Both HAFO variants increased plantar flexion moments when compared to SMO and shoes only. Ankle power absorption was increased significantly only with HAFOs, while greatest power was generated with shoes only. Radtka [19] found significant differences in pre-swing peak ankle power between no orthosis and SAFOs and between HAFOs and SAFOs. Peak ankle power was reduced by SAFOs, while using HAFOs it remained similar to that with no orthosis. Rethlefsen [20] also reported lower power generation with SAFOs compared to HAFOs. Chambers [36] reported that "standard AFOs" resulted in significant reduction in power generation at push-off due to reduced plantar flexion, and that using DAFOs with contoured footplates allowed ankle power generation that was similar to shoes only. Carlson [18] found that neither SAFOs or SMOs increased ankle power at push-off, although only the SAFOs improved propulsive moments. Abel [16] found that SAFOs significantly reduced ankle power, an effect which would have presumably been greater had the SAFOs not buckled. Lam [23] found that both

SAFOs and DAFOs improved maximum plantar flexion moment in comparison to barefoot, with SAFOs enabling generation of a significantly higher 2nd peak of the GRF. Investigating the effects of HAFOs, SAFOs and PLS, Buckon [17] reported that power generation in stance decreased significantly, with the greatest decrease seen in the SAFO, and least in the PLS. The timing of peak power generation also changed significantly, being later and closer to normal in all three AFO configurations compared with barefoot. Power absorption, showed slight change towards normal, particularly with the SAFO but this was not significant. Bennett [37] investigated the effect of "AFOs" on mechanical energy recovery, reporting a significant increase in peak push-off moments.

Conclusions

Orthoses that restrict ankle joint motion have been demonstrated to reduce power generation and absorption at the ankle. This reduction in power is inevitable, and the value of this finding is of questionable clinical value. There is a suggestion that carbon fibre construction may improve power generation without sacrificing control of unacceptable ankle kinematics. Further research is required to properly investigate this. Loss of power generation with some orthoses may be an acceptable compromise in order to optimise other parameters of gait. SAFOs may improve ankle joint moments in the propulsive phase of gait and help increase the 2nd peak of the ground reaction force in late stance.

2. ANKLE KINEMATICS

Mixed

Investigating the effect of PLS Ounpuu [33] reported a statistically significant improvement in ankle dorsiflexion in both swing and end stance. Molenaers [6] also reported improvements in ankle position throughout stance with PLS. Radtka [26] significantly reduced plantarflexion using SAFOs and DAFOs with plantarflexion stop. Kirkeide [8] found that SAFOs and DAFOs significantly reduced ankle excursion and timing. Heel contact time was significantly increased by both orthoses when compared to barefoot, with the greater effect seen in DAFOs, which also produced plantar dynamics closest to normal. Hayek [5] reported significant improvements in dorsiflexion in swing and at initial contact using SAFOs and HAFOs, with the type of AFO having borderline significance. There were no significant changes to loading response with either orthosis. Hainsworth [38] also found that SAFOs and HAFOs with 90^0plantarflexion stop improved ankle kinematics. Hobbs [28] compared the effects of SAFOs, HAFOs and SMOs, reporting that HAFOs and SMOs demonstrated "improved" dorsiflexion at terminal stance. Desloovere [30] found that Klenzack AFOs, SAFOs, HAFOs and PLS led to more dorsiflexion at initial contact and less plantarflexion at push-off.

Hemiplegia

Balaban [10] and Romkes [11] reported significant correction of equinus using a HAFO, as did Thompson [9] using a SAFO. Buckon [15] investigated the effects of a HAFO, a SAFO and a PLS finding that ankle dorsiflexion at initial contact was significantly increased in all AFOs compared to barefoot, with greatest improvement noted in the HAFO, and least in the PLS, whereas one might have expected this from the SAFO. The authors acknowledged that the SAFOs deformed into dorsiflexion in almost all cases, which may infer that plantarflexion could also have been occurring. Peak stance dorsiflexion was significantly increased in all AFO configurations compared to barefoot, particularly the HAFO. Ankle dynamic range was significantly

decreased in all orthoses, and was understandably significantly less in the SAFO. Improvements in ankle dorsiflexion with the knee extended of the order of 2^0 were noted only in the HAFO and the PLS, leading the authors to conclude that "this refutes claims that stretch of plantarflexors increases spasticity". It is questionable whether a change of 2^0 is clinically significant. Brunner [12] reinstated heel-toe pattern with both a SAFO and a flexible AFO. The reported early heel rise suggests that the SAFO may have benefitted from tuning. It is also interesting to note that the flexible AFO and the SAFO allowed almost the full range of functional dorsiflexion. Van Gestel [13] improved dorsiflexion angle at initial contact with a PLS, a Dual Carbon Fibre Spring AFO, and an Orteam® AFO, although only the improvement with the Carbon Fibre orthosis was significant. The carbon fibre spring orthosis reportedly produced the most physiological 3rd rocker. Excessive stance phase dorsiflexion of the ankle was noted in both the PLS and Orteam® AFO, suggesting inadequate resistance to dorsiflexion, despite the reported tuning. Dorsiflexion was also considered excessive in swing in these two devices, possibly suggesting poor alignment. Desloovere [14] significantly improved 1st and 2nd ankle rocker and foot clearance in swing using a PLS and a dual carbon fibre spring orthosis. The dual carbon fibre spring orthosis allowed better ankle motion during push off, and significantly increased ankle angular velocity in prewsing, while the PLS significantly improved power absorption at loading response and produced a slightly more normal ankle moment. Significant improvement in ankle range of motion and ankle angular velocity was also reported with shoes only. Romkes [29] found that both a HAFO and a DAFO significantly increased dorsiflexion at initial contact and peak dorsiflexion, with the HAFO (which unlike the DAFO had a plantarflexion stop) producing a more normal ankle angle.

Diplegia

Abel [16] significantly improved ankle excursion with a SAFO, while acknowledging that the "SAFOs" were actually designed to allow dorsiflexion by buckling. Lucareli [39] reviewed the effect of hinged FRAFOs, reporting significant reductions in ankle dorsiflexion in those with maximum stance knee extension greater than 15^0. Rethlefsen [20] reported increased dorsiflexion at initial contact and terminal stance using both SAFOs and HAFOs, with terminal stance dorsiflexion greatest in HAFOs. Interestingly this was reported to be greater than the amount of passive dorsiflexion available, leading the authors to conclude that this additional dorsiflexion may be allowing a stretch to the triceps surae. An alternative interpretation is that the additional dorsiflexion used up gastrocnemius length at the expense of knee extension, which was reportedly slightly less in the HAFOs. Some of the subjects in SAFOs exhibited considerable hyperextension of the knee at terminal stance, suggesting inadequate tibial progression and a need for tuning. Radtka [19] also compared SAFOs and HAFOs, finding that both reduced excessive plantarflexion throughout stance. Again, dorsiflexion at terminal stance was greater in the HAFOs compared to SAFOs.

Buckon [17] significantly improved dorsiflexion at initial contact and swing using SAFOs, HAFOs and PLS, while dynamic ankle range significantly decreased. The increase in peak stance dorsiflexion was greater in HAFOs than SAFOs. Using the same orthoses, Smiley [21] found that all designs appeared to produce an abnormal amount of dorsiflexion across the gait cycle. Hassani [35] found that compared to barefoot, both HAFOs and DAFOs produced significant differences in peak stance dorsiflexion, and peak swing plantarflexion, with no statistical significant differences between the two designs. Carlson [18] found that compared to shoes only, equinus

was improved by SAFOs but not by SMOs. Lam [23] found that SAFOs and DAFOs both significantly improved ankle position and peak mean stance dorsiflexion, while reducing plantar flexion at push off and overall ankle ROM. Crenshaw [32] reported that both HAFOs and tone reducing HAFOs significantly reduced plantarflexion at heel strike and toe-off compared to shoes only and SMOs.

Chambers [36] found that DAFOs with contoured footplates and "standard" restricted ankle motion during walking, reporting a tendency towards "toe-in" with both designs. All subjects in this study had equino-valgus foot deformity, correction of which would be expected to internally rotate (adduct) the mid tarsal joint, potentially highlighting more proximal transverse plane abnormalities. By contrast, Dole [40] reported a significant reduction in the angle of toe-in using HAFOs.

Conclusions
It is fairly clear that AFOs of different designs can impact positively on ankle kinematics. Solid ankle designs or those with plantarflexion stops are best at preventing equinus. Excessive dorsiflexion, as seen in crouch gait, is best addressed with SAFOs or AFOs with dorsiflexion stops. The increases in late stance dorsiflexion that have been reported using HAFOs should be considered in the context of gastrocnemius length and the possible adverse effect on knee and hip kinematics in late stance, if dorsiflexion is allowed to occur when this muscle is short.

3. EFFECT OF ORTHOSES ON FOOT ALIGNMENT

Only one study [41] was identified that addressed the important issue of the ability of AFOs to influence foot and ankle alignment. X-rays and quantitative segmental analysis were used to investigate the effect of AFOs on mixed triplanar foot deformities in a mixed group of ambulant children. AFO designs investigated in this study included SAFO, PLS, HAFO and GRAFO. While the study showed statistically significant changes in all measurements of segmental alignment, the authors concluded that as the actual changes were small ($<10^0$) they were clinically insignificant, and that AFOs had failed to improve static foot alignment in the majority of feet. Correction was most successful in the pronated (valgus) foot, possibly due to the greater flexibility commonly associated with this type of deformity. It is possible that the age of subjects may have adversely affected the results, as some subjects were skeletally quite mature (up to 18 years old).

Conclusions
It is difficult to draw conclusions from the little evidence that exists for orthotic effect on foot alignment. While statistically significant improvements in foot alignment have been reported, the suggestion that corrections of the order of up to 10^0 are clinically insignificant is perhaps open to question. Control of alignment in the growing child is critical if progression to skeletal deformity of the foot is to be resisted. More research is required in this area, particularly on the feet of those who have yet to develop fixed deformities.

WHAT ARE THE INDIRECT EFFECTS OF ORTHOSES ON THE KNEE?

Mixed
Hayek [5] investigated the effect of SAFOs and HAFOs reporting significant reduction in knee flexion at initial contact in the hemiplegic group, but no change in the diplegic group. Surprisingly, neither orthosis had any significant effect on knee

kinematics in loading response, single support or swing phase, raising questions about the alignment and design characteristics of the orthoses. Desloovere [30] analysed the effects of Klenzack AFOs, SAFOs, HAFOs and PLS on gait. Compared to barefoot, AFO use significantly increased knee flexion during loading response, with SAFOs producing a more pronounced knee extension moment during stance. Lampe [42] studied the influence of orthopaedic shoes, insoles, DAFOs, ring orthoses and AFOs on the kinetics and kinematics of gait in a mixed group of 18 children including some with CP. Barefoot, one child with diplegia had a 15^0 knee extension deficit throughout stance, which increased to 25^0 using orthopaedic shoes. Duration of knee joint extension was also increased markedly by these shoes, while extension moment in initial stance decreased slightly. The use of a shoe with an orthotic insole, compared to barefoot, resulted in improved knee extension in early stance phase in one hemiplegic child. Use of an AFO in a diplegic child improved knee extension and knee stability in stance and increased swing knee flexion.

Molenaers [34] investigated the effect of BTA combined with short leg casting and orthotic management. All children, except those who already had casting prior to BTA, demonstrated improved stance knee kinematics post BTA. Huenaerts [2] investigated the effects of a PLS before and after BTA. Although the appropriateness of using PLS in the presence of spasticity may be questionable, the PLS after BTA improved knee kinematics compared to barefoot walking. However, knee kinetics with the PLS after BTA showed that the alignment of the ground reaction force to the knee was not always appropriate, suggesting that retuning of AFOs is of major importance after BTA.

Hemiplegia

Balaban [10] reported significant reduction in knee flexion at initial contact using a HAFO, but no significant change in maximum stance knee extension or peak swing knee flexion. Romkes [29] also reported no improvement in knee hyperextension using a HAFO, although timing of peak knee extension was significantly delayed by both a HAFO and a DAFO. Peak knee flexion was also significantly improved with both orthoses. Romkes [11] reported that a HAFO improved loading response knee kinematics and led to more dynamic knee movement, and creation of a flexion-extension pattern, suggesting that these findings might be the result of the increased walking speed. Brunner [12] found that a SAFO reduced maximum knee extension (compared to barefoot) and reduced the knee range of motion throughout gait, whereas a flexible AFO had only a slight influence on the knee. Buckon [15] reported greater improvement in knee hyperextension with a HAFO than with a SAFO, which is surprising as the orthoses were made from the same plaster model. However, it was acknowledged that most SAFOs deformed into dorsiflexion, so it is possible that they also deformed into plantarflexion, compromising knee hyperextension control. Jagadamma [43] investigated the effect on knee joint kinetics and kinematics of incrementally wedging a SAFO. Neither the SAFO nor was the footwear was adequately described, both of which influence outcomes. For the child in this study, a 13^0 angle of tibial inclination (achieved using an 8^0 wedge) produced optimal knee alignment. Desloovere [14] reported that compared to barefoot, both a PLS and a dual carbon fibre spring AFO significantly improved knee shock absorption and maximum stance knee flexion, and changed the amount and timing of maximum swing knee flexion. Compared to shoes however, maximum knee flexion and timing of peak swing knee flexion were unchanged, although maximum knee flexion in stance remained significantly higher. The PLS was also significantly associated with minimal knee hyperextension. Although both orthoses significantly increased knee

flexion velocity at toe-off, this effect was considerably greater with the PLS, perhaps indicating inadequate resistance to tibial progression. Van Gestel [13] reported decrease in maximum stance knee extension in using a PLS and an Orteam®AFO, whereas a Dual Carbon Fibre Spring AFO actually encouraged hyperextension, suggesting that the carbon fibre orthosis was not optimally tuned. Knee velocity in swing was also increased in all orthoses, as was peak knee flexion, and knee shock absorption. Maximal knee flexion was decreased, but remained higher than normal. Desloovere [14] found that using shoes only (compared to barefoot) led to significant changes in knee motion, increasing knee shock absorption, flexion in swing and maximal knee extension in stance.

Diplegia

Abel [16] (1998) reported significant improvement in knee excursion using a SAFO, but found that the effect on the knee in stance was not significant, perhaps due to the fact that the SAFO was clearly not rigid. Farmer [44] used SAFOs as part of a programme of targeted training for crouch posture. Using SAFOs to support the lower legs in an inclined alignment on a sprung "wobble board" the interaction between the hip and knee flexors and extensors was enhanced by throwing and catching a ball. After three months the angle of tibial inclination was reduced, and a 10^0 reduction in knee flexion reported at six months. It is interesting that improved control of standing was demonstrated prior to measurable increase in hamstring length, challenging the view that muscle length must be gained as a precursor to improving control. Further research is required to investigate this effect. Wesdock [45] investigated the effect of 90^0 SAFOs on patients with crouch gait. The orthoses were used with shoes alone and also following the addition of heel wedges. No significant improvements were made in knee extension using these interventions. Possible reasons for this include poor foot progression angle in approximately half of the subjects, and gastrocnemius shortening which was not accommodated in the SAFOs. Proximal muscle weakness and/or spasticity were also suggested as possible factors, with the authors noting that the amount of knee flexion demonstrated by all subjects exceeded the amount of contracture measured at the knee. The wedge was the only footwear modification, with no attempt made to interrupt anatomical 3rd rocker by stiffening the sole of the shoes. There was no mention of SAFO stiffness, another critical factor. The fact that the addition of wedges made no difference to knee extension is perhaps not surprising, as the effect of adding wedges when the heel is off the ground is to realign the GRF posteriorly, actually reducing the knee extension moment. Lucareli [39] reported significant reduction of knee flexion hinged FRAFOs (with dorsiflexion stop) particularly in the more involved subjects. Rethlefsen [20] reported no significant differences in knee kinematics between SAFOs and HAFOs. It is, however, interesting to consider whether the greater angle of terminal stance knee flexion seen in the HAFO was linked to the observation that dorsiflexion in the HAFO at this stage of gait exceeded the passive range recorded in the gastrocnemius. Ankle dorsiflexion in the absence of adequate gastrocnemius length will limit knee extension. Also of interest is the observation that some subjects in the SAFO exhibited considerable hyperextension of the knee at this stage, suggesting inadequate tibial progression and a need for the SAFOs to be tuned. Radtka [19] also found that excessive knee flexion in barefoot stance was unchanged with either HAFOs or SAFOs, nor were knee moments in stance. The authors concluded that the concerns expressed by some clinicians regarding the possibility of increased knee flexion in crouched gait resulting from using HAFOs, rather than SAFOs were unsubstantiated. However, had the SAFOs blocked ankle dorsiflexion as intended, the external knee extension moments generated would have been greater, and differences between the two AFO designs

may have been evident. Buckon [17] reported no change in knee kinematics, with SAFOs, HAFOs and PLS, although the early stance knee extensor moment was significantly increased with HAFOs. Buckling of the SAFOs was reported, which must have negatively affected their function. Smiley [21] reported statistically significant differences in maximum knee flexion in swing using HAFOs, and in knee flexion in mid swing using HAFOs and SAFOs, but not PLS. Chambers [36] compared the effects of DAFOs and "standard" AFOs reporting that the "tall standard" AFO made from thicker plastic was best at restricting dorsiflexion, with significant improvement in mean stance knee extension. Lam [23] found that DAFOs significantly increased knee flexion at initial contact compared to barefoot, while SAFOs reduced this. Hassani [35] reported that compared to barefoot there were significant differences in peak knee flexion in stance and swing using HAFOs and DAFOs, but no significant difference in peak knee extension in stance or swing with either orthosis.

Conclusions

The indirect effects of AFOs on the kinetics and kinematics of the knee joint have been reported by a number of authors using a number of different orthosis designs. Knee hyperextension can be influenced by SAFOs or by HAFOs with plantarflexion stops, while excessive flexion is best addressed using SAFOs or AFOs with dorsiflexion stops. The alignment of the AFOs is critical if optimum effect is to be achieved at the knee, with tuning of the AFOs likely to be beneficial.

WHAT ARE THE INDIRECT EFFECTS OF ORTHOSES ON THE HIP?

Mixed

The indirect effects of AFOs on hip joint kinematics in mixed groups of subjects have been investigated by a number of authors [2, 5, 6, 27, 28, 30, 34]. Molenaers [6] reported improvement in hip extension throughout stance using PLS. Desloovere [30] analysed the effects of Klenzack AFOs, SAFOs, HAFOs and PLS compared to barefoot, finding that use of PLS and HAFOs increased hip extension at terminal stance and decreased hip abduction during swing, but had no effect on hip rotation. Hayek [5] found that SAFOs and HAFOs had minimal effect on hip kinematics when compared to barefoot. Hobbs [28] reported that SAFOs significantly increased both hip extension during stance and hip flexion in swing, whereas use of HAFOs and SMOs led to excessive hip flexion, relative to normal. Huenaerts [2] investigated the effects of PLS before and after BTA on the gait of children described only as "predominantly spastic", reporting that PLS significantly decreased internal rotation of the hip at initial contact. At midstance and terminal stance there was further significant improvement after BTA. Hip extension also significantly increased after BTA. Molenaers [34] investigated the effect of multilevel BTA combined with daytime AFOs, night time KAFOs and serial casting if required, finding that those children without casting showed greater hip extension at terminal stance following BTA injections. The incidence of internal hip rotation in stance and swing significantly decreased for all patients after treatment.

De Groot [27] investigated the effect of shoes only on gait, reporting a significant decrease in minimum hip flexion in stance and a significant increase in hip range of motion.

Hemiplegia

Romkes [11] reported an increase in hip flexion at initial contact using a HAFO, compared to walking with no orthosis. The authors suggest that this increase may have been related to increases in velocity and step length, which have been demonstrated to increase peak hip flexion and extension in healthy children. Brunner [12] investigating the effects of a SAFO and a flexible AFO found that the SAFO increased hip flexion, extension and abduction, but decreased adduction. Changes to abduction and adduction were more pronounced in the flexible AFO. Perhaps surprisingly, the reported arcs of hip flexion and extension were similar in both orthoses. This similarity should however be considered in the context of the amount of ankle dorsiflexion reported in the two different orthosis designs. The SAFO clearly buckled, showing an average of 8.4^0 dorsiflexion compared to 11.9^0 in the flexible AFO. With similar ankle kinematic graphs, it is reasonable to expect similar orthotic effect at the hip in the two test conditions. Investigating a PLS, a Dual Carbon Fibre Spring AFO, and an Orteam® AFO, Van Gestel [13] reported significant improvements in hip moments and power, except for maximal abduction moment in stance and maximal power generation at pre-swing. Although there were only small differences between the different orthoses, the Dual Carbon Fibre Spring AFO created a significantly higher maximal hip flexion moment in stance. It was reported that maximal hip extension in stance increased with the PLS and the Dual Carbon Fibre Spring orthosis, the Orteam® orthosis decreased it, although neither change was significant. Desloovere [14] reported that compared to barefoot gait, using shoes only increased coronal hip angle and pelvic obliquity in swing. While these changes were stated to be statistically significant, they were of the magnitude of only a few degrees, and therefore perhaps not clinically significant.

Diplegia

Using SAFOs, Abel [16] demonstrated significant improvement in hip excursion, even though the SAFOs allowed some dorsiflexion by buckling, while Van Rooijen [31] reported significant decrease in minimum hip flexion in stance. Lucareli [39] reviewed the effect of hinged FRAFOs, finding no significant changes to maximum hip extension. Buckon [17] reported no changes at the hip using HAFOs, SAFOs and PLS. Lam [23] evaluated the effects of SAFOs and DAFOs reporting that DAFOs significantly increased hip flexion at initial contact compared to barefoot. Hassani [35] reported significant differences in peak hip flexion in stance with both HAFOs and DAFOs, compared to barefoot. There was however, no significant difference to peak hip extension in stance or peak hip flexion in swing. Crenshaw [32] investigated the effects of tone reducing features in lower limb and reported that the AFOs without these features showed significant increase in maximum extension moment and total power generation at the hip.

Conclusions

There is some evidence that hip joint kinetics and kinematics can be influenced by AFOs. In order to achieve indirect orthotic effect at the hip, the AFOs must be successful in controlling ankle kinematics. Control of the permitted arc of ankle motion in flexible AFOs and HAFOs, and the stiffness of SAFOs are critical factors if the ground reaction force is to be successfully manipulated. Tuning of the orthoses is important if the hip is to be positively influenced. There is some evidence that the use of shoes only can influence hip kinematics.

WHAT ARE THE INDIRECT EFFECTS OF ORTHOSES ON THE PELVIS?

Mixed

A number of authors [2, 6, 30, 34] investigated the indirect effect of AFO use on the pelvis in mixed subjects. Two studies [6, 30] investigated the effects of a range of AFOs including, SAFOs, HAFOs and PLS, reporting decreased pelvic motion in the transverse place, compared to barefoot, in all but the Klenzack AFOs. The other two studies [2, 34] investigated the effect of BTA when used in conjunction with orthoses. Molenaers [34] used 3D gait analysis and EMG to investigate the effects of BTA combined with daytime AFO use and KAFOs at night (using short leg casting when indicated). Increased pelvic stability was reported, with greatest improvement seen in those who had short leg casting before BTA. Huenaerts [2] investigated the effects of a PLS before and after BTA reporting no changes in pelvic kinematics as a result of either.

Hemiplegia

Brunner [12] investigated the effect of a SAFO and a flexible AFO on hemiplegic gait. The SAFO improved pelvic obliquity, had no effect on pelvic tilt, but adversely affected (i.e. increased) pelvic rotation. The flexible AFO improved pelvic obliquity, slightly increased pelvic tilt, and had an adverse effect on pelvic rotation less than the SAFO. Romkes [11] found no significant change at the pelvis using a HAFO.

Diplegia

Abel [16] reported significant improvement in pelvic excursion using a SAFO (although the AFO did buckle under load). Buckon [17] reported no changes at the pelvis in diplegic subjects using HAFOs, SAFOs and PLS.

Conclusions

Little evidence exists to support the hypothesis that AFOs can positively impact on pelvic motion, although there is a suggestion that this may be possible. If using SAFOs, the rigidity and the alignment of these is important if the effect at the pelvis is to be positive, rather than negative.

What effect do orthoses have on the metabolic and cardiopulmonary cost of walking?

Hemiplegia

Two studies investigated the effect on oxygen consumption of different AFOs including HAFO, SAFO and PLS. Buckon [15] compared the effects of all three designs on hemiplegic subjects. At self selected speed, there were no significant changes in oxygen consumption or energy cost (energy consumption/velocity), however in all three AFO configurations self selected speed significantly increased. In fast walking oxygen consumption was unchanged, but energy cost was significantly decreased when wearing the HAFO. Balaban [10] reported a significant reduction in oxygen consumption with HAFO use, testing subjects at the same speed, rather than at self-selected speed.

Diplegia

A number of studies [17, 21, 25, 46] investigated the effects of a number of AFO designs including HAFO, SAFO, PLS and “conventional plastic AFOs” on the gait of children with diplegia. Mossberg [25] investigated the effects of bilateral

"conventional plastic AFOs" on energy expenditure at self-selected speed. Most subjects demonstrated a statistically significant decrease in Physiological Cost Index (PCI) when using AFOs. Maltais [46] investigated the effects of HAFOs on the metabolic and cardiopulmonary cost of walking. Net oxygen uptake was significantly reduced at both slow and fast speed, but not at self-selected walking speed. Significant reduction in net pulmonary ventilation was reported at slow speed. There was no effect on heart rate or respiratory exchange ratio at any of the three speeds, or on any physiological variable at self-selected walking speed. Smiley [21] compared the effect of HAFOs, SAFOs, and PLS to shoes alone on energy cost at self selected walking speed. The measure used was the Energy Efficiency Index (EEI), which is defined as heart rate minus resting heart rate divided by speed. No significant differences were found in the EEI between the three AFOs, but it is unclear whether the EEI was different between the AFOs and the shoes alone condition. Buckon [17] also investigated the effects of HAFO, SAFO, and PLS, reporting significant reduction in energy cost at self selected and fast walking speeds with all designs when compared to shoes only. Oxygen consumption was significantly reduced using the HAFO and SAFO at self selected and fast walking speeds. Reduction in oxygen consumption with the PLS was not statistically significant due to the resulting increase in velocity.

A further study by Van de Walle [7] failed to describe the type of subjects fitted with PLS. In this study the slight increase in velocity was associated with increase in energy consumption, leading the authors to conclude that there was no benefit in terms of gait efficiency when using AFOs.

Conclusions

There is some evidence that the metabolic and cardiopulmonary cost of walking may be improved by the use of AFOs. In some studies where walking speed has been standardised, reduction in oxygen consumption has been reported with AFO use. In other studies at self-selected walking speed, oxygen consumption has been unchanged by AFO use, but self-selected walking speed has increased.

WHAT EFFECT DO ORTHOSES HAVE ON MUSCLE ACTIVITY?

Mixed

Dumas [47] investigated the effects of an AFO on the reflex and non reflex components of hypertonia, in one hemiplegic and one diplegic subject. Unfortunately, no details were given regarding either child, or of the AFO design employed. After six months of AFO use, the resistive forces for the non-reflex component were reduced by 19.7% in the diplegic child, and by 44.5% in the hemiplegic child. These forces had returned to pre-AFO values two months after discontinuing AFO use, suggesting a temporary effect. The reduction in the reflex component, seen only in the hemiplegic child at six months was also lost two months after discontinuing AFO use. These effects were consistent with the Modified Ashworth Scale (MAS) scores which were initially 4 in both children, decreasing to 2 in the diplegic and 1 in the hemiplegic at six months. Two months after discontinuing AFO use, only the hemiplegic MAS increased (to 2). In both children passive dorsiflexion was increased after six months AFO use, but this gain was also lost within two months of discontinuation. Lampe [42] studied the effects of orthopaedic shoes, insoles, DAFOs, ring orthoses and AFOs in a mixed group which included few CP children. One diplegic child used both a "long and short orthosis" (not described), with minimal influence on quadriceps muscle tone. Neither orthosis had any effect on EMG signal intensity of biceps femoris or gluteals. In the hemiplegic subject, use of a shoe plus

orthotic insole resulted in improved knee extension in early stance phase, which correlated with increased activity of rectus femoris, which reduced through mid and late stance. Due to the small number of patients, the findings have limited applicability. Following BTA combined with short leg casting and orthotic management, Molenaers [34] reported persistence of premature gastrocnemius activity in stance, with most subjects showing lower levels of gastrocnemius activity at initial contact with all regimes. The frequency of tibialis anterior activity during terminal swing and loading response increased after BTA. Radtka [26] found that neither SAFOs nor DAFOs changed abnormal muscle timing.

Hemiplegia

Romkes [11] used EMG to investigate the effect of a HAFO on muscle activity, compared to barefoot. Tibialis anterior showed the greatest EMG reduction, with average peak activity reducing by 36.1% during initial contact/loading response, and by 57.3% just after toe off. While slight changes were detected in rectus femoris, semitendinosis, biceps femoris, vastus medialis and lateralis during swing, these changes resulted in no notable functional effects. Surprisingly, no data was collected relating gastrocnemius or soleus. Matthews [48] investigated the effect of a HAFO with neurophysiological features in a single subject, reporting reduction in upper limb spasticity and night cramps in the calf. Unfortunately, the comparison was with a SAFO without any neurophysiological features, so any specific effect of neurophysiological modifications was not isolated.

Diplegia

Van Rooijen [31] reported that compared to non-disabled controls, barefoot diplegic gait showed significantly higher coactivation (hamstrings and rectus femoris; tibialis anterior and gastrocnemius medialis). Wearing SAFOs, significantly decreased the amplitude of rectus femoris and tibialis anterior, significantly increased the amplitude of medial hamstrings and had no effect on the amplitude of gastrocnemius medialis. While there were no significant effects on coactivation over gait cycle as a whole, there was significant increase of coactivation in the shank during loading response. While this study implies that SAFOs have only small effects on EMG amplitudes and little influence on coactivations, the authors suggest that further modifications to the SAFO and/or footwear (i.e. tuning) may have resulted in greater improvements in gait and lower levels of co-activation. Lam [23] reported that when compared to non-disabled controls, median frequency of EMG signal indicated extremely high firing in the lower limbs of diplegic subjects, resulting in tiredness. While neither SAFOs nor DAFOs changed total muscle contracting duration compared to barefoot, SAFOs significantly reduced median frequency of EMG signal while the DAFOs did not. This suggests potential for improvement of walking endurance when wearing SAFOs. Radtka [19] found no change in the timing of muscle activity (pretibial, triceps surae, quadriceps femoris, and hamstrings) using SAFOs and HAFOs. Rethlefsen [20] (1999) found no differences in peak EMG amplitude of calf muscles using HAFOs and SAFOs.

Conclusions

There is limited evidence that muscle activity may be positively affected by the use of AFOs, indicating a clear need for further research. When such effect has been reported, it has been noted to be restricted to the period of orthosis use, or slightly longer. The issue of whether orthoses can influence the physiological properties of muscle has interested researchers for some time. Williams [49] reported changes in

muscle following periods of immobilisation, including modification of the number of sarcomeres and the degree of overlap of myosin and actin filaments. Resultant improvement in muscle tension was reported, especially in younger children. Tardieu has demonstrated that at least six hours of immobilisation per day is required to change resistance to passive stretch and decrease tone in the soleus. Given the prolonged daily periods of orthotic use for many patients, it is possible that SAFOs may indeed confer this effect, albeit to the muscles actually immobilised by the orthosis. Pharmacological interventions may be required to influence tone in the proximal muscles so that the orthosis can optimise proximal joint kinetics and kinematics [34, 50].

Ronan [50] cautions against the use of ankle joints that assist dorsiflexion in case these elicit a spastic response in the plantarflexors. However, significant stretch of the plantarflexors can also occur in stance phase using free-dorsiflexion ankle joints. Some orthoses incorporate the features of so-called "inhibitory casts" which are thought to encourage reflexive relaxation by stimulation of one of the tonic reflexes of the foot [51]. Mills [52] has demonstrated changes in EMG activity of spastic muscles using such inhibitory casts.

WHAT EFFECT DO ORTHOSES HAVE ON MUSCLE LENGTH?

Mixed

One study [47] investigated the effects of an AFO on the reflex and non reflex components of hypertonia. Only two participants were studied, one hemiplegic and one diplegic. No details were given regarding either child, or of the AFO design(s) employed. In both children passive dorsiflexion was increased after six months AFO use, but this gain was lost within two months of discontinuation.

Hemiplegia

One study [9] investigated whether proximal muscle length in subjects with hemiplegia is modifiable by the use of AFOs. Joint angles calculated from Vicon gait analysis were used to model muscle length throughout the gait cycle, using Software for Interactive Musculoskeletal Modelling (Musculographics, Evanston, IL, USA). While maximum length of semimembranosus, semitendinosus, biceps femoris, rectus femoris, iliacus, and psoas were calculated throughout gait, the only significant increase reported was in the length of the hamstrings and rectus femoris. This effect was less marked in the more involved children in the study (GMFCS level II and III) than in the group as a whole. No modelling of gastrocnemius or soleus was undertaken in this study. The study results may have been affected by the use of comparative data from adults, which the authors acknowledged as less than ideal.

Diplegia

Two studies [17, 44] investigated the effect of AFOs on muscle length in subjects with diplegia. Buckon [17] reported no changes in passive dorsiflexion range using HAFOs, SAFOs and PLS. In a single case study Farmer [44] investigated the use of SAFOs as part of a programme of targeted training for a child with crouch posture. With the child standing on a sprung "wobble board," inclined SAFOs were used while interaction between the hip and knee flexors and extensors was enhanced by throwing and catching a ball in such a way that perturbation was ensured. The angle of tibial inclination was reduced after 3 months. At 6 months the popliteal angles had reduced by 10^0, indicating hamstring lengthening. Knee flexion in standing and walking was also reduced by approx 10^0.

Conclusions
There is limited evidence of beneficial effect of AFO use on muscle length, although there is a suggestion that this may be positively influenced by orthosis use. Further research is required.

HOW DO ORTHOSES AFFECT FUNCTION AND ABILITY?

Russell [53] investigated whether the Gross Motor Function Measure (GMFM-88) is sensitive to within-child changes in function arising from the use of ambulatory aids, including orthoses. Paired *t*-tests comparing barefoot and aided assessments revealed that the GMFM-88 total scores were significantly higher for assessments using AFOs, ambulatory aids, and AFO plus ambulatory aid, demonstrating that the GMFM-88 is sensitive to functional changes of the magnitude conferred by aids and/or orthoses. With regard to the changes seen in the orthoses group, the significance varied by GMFCS level, with the GMFM-88 sensitive enough to detect changes in levels I, III, and IV but not level II. This may have been because of the small sample size at level II and insufficient power to detect a difference, even if one existed. The authors concluded that the GMFM-88 might provide a clinically useful tool to help in understanding the impact of ambulatory aids and orthoses on gross motor skills of children with CP. Evans [54] used the GMFM and the Gross Motor Performance Measure (GMPM) to evaluate the effects of lower extremity orthoses on pre-ambulatory children with spastic CP. Although significant improvement was observed in the group using orthoses in addition to physiotherapy, no significant difference in gross motor function as measured by the GMFM-88 dimensions D (Standing) and E (Walking, Running, and Jumping) or in GMPM was found. In the study by Bjornson [55] significant improvements were detectable in crawling/kneeling, standing, and walking/running and jumping skills in children with spastic CP wearing DAFOs, using the GMFM 88 and the Gross Motor Ability Estimator 66. Kott [56] investigated orthotic effect on performance using the Standardized Walking Obstacle Course (SWOC), the Pediatric Balance Scale (PBS), performance on individualized goals related upright function, and self-reports of comfort and stability. The SWOC and PBS detected no significant differences in performance with and without orthoses. However, some children performed better on an individualized goal with their orthoses, while approximately half reported feelings of improved comfort and stability while wearing their orthoses. The results of this study do not reveal improved ambulation and balance wearing orthoses, when measured in a functional context.

Hemiplegia
Buckon [15] investigated the effects of HAFOs, SAFOs and PLS on functional tasks in children with hemiplegia, using the GMFM and GMPM. Functional mobility was assessed using Functional Skills-Mobility Dimension of the Pediatric Evaluation of Disability Inventory (PEDI). While the GMFM showed no changes between conditions, the GMPM showed improved performance in coordination and weight shift, but not alignment and stability. Functional skills, assessed using the PEDI, improved significantly with all three orthoses. The PEDI results reinforced the GMFM/GMPM findings indicating that AFO use enhanced performance of already mastered skills rather than attainment of new skills.

Diplegia
Maltais [46] also found no change in these GMFM dimensions in a study of children with spastic diplegia using HAFOs. Hassani [57] evaluated the effect of HAFOs and DAFOs in children with spastic diplegia, reporting that while gait and temporal stride

measures were sensitive to changes between braced and unbraced conditions, no significant differences were detectable with either the GMFM or the Pediatric Outcomes Data Collection Instrument (PODCI), which correlated strongly with each other. Buckon [17] investigated the effects of HAFOs, SAFOs and PLS on functional tasks in diplegic subjects using the GMFM, reporting no improvement within the standing dimension but significant improvements to the walking/running/jumping dimensions. No significant improvements were detected using either the GMPM or PEDI with any AFO. Ferdjallah [58] investigated the relationship between postural stability, gait, and GMFM goal scores in a study of the effectiveness of HAFOs and DAFOs on children with spastic diplegia. GMFM goal total scores increased when children were assessed with orthoses compared to barefoot, but did no information was provided on which dimensions of the GMFM were used. The authors recommended that a combination of measures should be used to evaluate the effects of AFOs, including measures of postural stability, gait, and GMFM assessment. Kornhaber [24] investigated the effects of SMOs and HAFOs on gross motor function in children with spastic diplegia. All scored higher on the GMFM in SMOs than in HAFOs for tasks that required ankle mobility. Performance in mat mobility and STS improved in younger children. Older children improved in more difficult tasks such as stair climbing and jumping. Most children scored lower for standing on one foot, but not on other tasks that required ankle stability.

SITTING BALANCE

Only one study [59] investigated the effects of SAFOs on seating position in non ambulatory children. Seating position was standardised, with 15 degrees of hip abduction achieved using a wedge. The only significant change observed was a decrease in kyphosis in all four children. The small sample size, coupled with the fact that this study relied on comparison of tracings of projected images, must bring the value of the results into question.

Conclusions

It is not possible to draw any conclusions from the very limited research on the effect of orthoses on sitting balance.

STANDING BALANCE

Diplegia

Wesdock [45] investigated the effect of 90^0 SAFOs on standing balance, and the influence of adding heel wedges. Balance time increased when SAFOs with wedged shoes were used compared to no orthosis, but without statistical significance. There was also an increase in balance time using SAFOs with wedged shoes compared to SAFOs without, but again the difference was not statistically significant. However, it is interesting to note that balance time in subjects who were already able to stand without orthoses for at least 15 seconds, significantly improved when using a SAFO with wedged shoes compared to no orthosis, and when using a SAFO with wedged shoes compared to a SAFO without. Standing balance was best in the SAFO plus wedge combination. Subjects with GMFCS level of II or III demonstrated greater improvements using SAFOs with wedged shoes compared to SAFOs alone, contrasting with the results of those at GMFCS IV, who demonstrated very small differences, supporting the hypothesis that children who exhibited better balance to begin with derive greater benefit from the addition of wedged shoes to their AFOs. Farmer [44] investigated the effects of targeted training and SAFOs on crouch posture

in a single child with diplegia. Interaction between the hip and knee flexors and extensors was enhanced by throwing and catching a ball while standing with SAFOs on a sprung "wobble board". After three months standing posture had improved, with the child able to maintain an erect posture.

Burtner [60] investigated the effect of SAFOs and carbon fibre AFOs of a spiral design on perturbed stance in children with diplegia. It was found that their preferred pattern of muscle-recruitment was well established and not altered by either AFO design. The SAFOs however, led to decreased activation of distal musculature (gastrocnemius) as well as decreased recruitment of ankle strategies (which are seen in normally developing children) for balance. These trends were not found with carbon fibre AFOs. The authors concluded that spiral design carbon fibre AFOs may be advantageous for children with spastic CP when balance control is required during unexpected perturbations in stance. However, the small sample size (four) in this study makes generalizing results difficult. Näslund [61] (2005) investigated whether DAFOs improved standing stability in children with diplegia GMFCS levels III and IV, reporting that that DAFOs did not improve standing posture, or distribution of body weight, compared to shoes only, although some showed improvement in weight distribution between the feet and improved sagittal plane alignment of the knee. Carmines [62] investigated the effect of DAFOs on static balance in spastic diplegia, using non-disabled barefoot children and barefoot CP children as controls. Mean centre of pressure was analysed, and it was reported that use of the DAFOs created an irregular M/L shift in the base of support, and a significant posterior shift. Jesinkey [63] investigated the effects of DAFOs compared to shoes only, on coordination of standing posture and reaching movement in spastic diplegia. With DAFOs a greater number of subjects showed anticipatory divergence of centre of pressure. DAFOs also led to improvement in weight distribution between the feet and sagittal plane alignment of the shank, but not lateral stability. Compensatory postural adjustments with proximal to distal muscle activation (which is considered to be an immature response) were reduced with DAFOs. The study was limited by the small sample (four) and the potential training effect from wearing DAFOs for three years prior to testing. Crenshaw [32] investigated standing balance using HAFOs, HAFOs with tone-reducing features, and SMOs with tone reducing features in diplegic subjects, compared to shoes alone, reporting no significant difference between test conditions. Jones [64] investigated whether AFOs would improve standing balance in children with GMFCS < II, using non-CP children as a control. Standing balance was assessed using an instrumented Romberg test, and postural sway parameter was measured during 20-second trials. While AFOs did not significantly reduce postural sway in the CP group when eyes were open, there was a significant reduction in sway when used with eyes closed.

Conclusions

There is some evidence to support the hypothesis that orthoses can improve standing balance. Further research is needed in this area.

SIT TO STAND

The effect of HAFOs and SAFOs on performance of the sit to stand (STS) manoeuvre in subjects with diplegia has been investigated in a number of studies [65-67]. In a study on perambulatory children, Wilson [65] compared the effect of a 90^0 SAFO and a HAFO with 90^0 plantarflexion stop reporting that those whose performance was already comparable to normal performed better without AFOs, which actually

hindered STS. Patients more than one standard deviation (SD) slower than normal showed significant improvements using HAFOs. Those patients with equinus in barefoot standing derived most benefit from HAFO use. SAFO use produced no significant improvement. Although the authors stated that "control of equinus using HAFOs" significantly improved the speed of STS, the SAFO provided the same equinus control as the HAFO, so the difference is clearly not to do with equinus control per se. Haideri [66] also investigated the effect of SAFOs and HAFOs in subjects with dynamic equinus, again reporting that subjects whose performance was already close to normal were better without AFOs, and performed worst using SAFOs. As in the study by Wilson [65] improvements were reported in the subset who were more than one SD slower than normal.

Park [67] used a motion analysis system to investigate the effect of HAFOs in subjects who were able to perform STS without using their upper limbs, unlike the study of Wilson [65] where this was allowed. The control condition was barefoot. Total duration of STS was significantly shortened by use of a HAFO. Significant increases were noted in initial knee flexion angle, and initial and final angle of ankle dorsiflexion with the HAFO. Maximal moment and power at the hip and knee were significantly increased, whereas maximal moment and power at the ankle were not significantly changed. The authors suggested that the increase in the speed of performing the task may have contributed to the changes in power at the knee and hip.

Conclusions

It is perhaps understandable that no papers were identified that related to STS manoeuvre in hemiplegic subjects. In diplegic subjects, there is some evidence that HAFOs can improve performance of the STS manoeuvre, but that this benefit may be limited in those whose performance is already close to normal. A number of authors have suggested that the ability to dorsiflex the ankle is important, with Wilson [65] identifying dorsiflexion as the only consistent strategy for STS. However, it should be remembered that in the presence of gastrocnemius shortening, the use of free dorsiflexion ankle joints is contraindicated. The use of HAFOs to bring about improvements in STS may be detrimental to walking performance. An alternative way to consider this issue is that it may actually be the angle of tibial inclination that is important, and that SAFOs that prevent tibial inclination, i.e. SAFOs that maintain the tibia in a position too close to vertical, may impede STS. It is possible that SAFOs that have been tuned to a position of some tibial inclination may be more effective. Further research is needed to clarify these issues.

Stair Climbing

Only one paper [68] was identified that investigated the impact of different AFO designs on stair locomotion in subjects with spastic hemiplegia. All subjects, who were able to ascend and descend stairs in a reciprocal fashion without assistance, were evaluated using a HAFO, a SAFO, a PLS, and barefoot. The stair-specific items from the Pediatric Evaluation of Disability Inventory (PEDI) were used to evaluate function, and a motion analysis system evaluated kinematics. Velocity ascending and descending stairs, as compared to non-disabled peers, improved using all AFOs, with the greatest improvement noted when using HAFO, and least with PLS. Differences were not statistically significant for velocity during stair ascent and descent, or in single limb stance. The results of this study suggest that stair ambulation in children with spastic hemiplegia is not impaired by the use of an AFO.

Conclusions
There is no evidence that AFO use impairs stair climbing in children with cerebral palsy. What little evidence there is appears to support the view that AFOs actually improve stair performance.

WHAT ARE THE BENEFITS OF TUNING ANKLE-FOOT ORTHOSES?

In the study by Owen [69] on independently ambulant children using SAFOs, all orthoses were tuned using the Orthotics Research and Locomotor Assessment Unit transportable Video Vector Generator (VVG). Tuning involved making fine adjustments to the angle of tibial inclination of each AFO using wedges under the heel, in order to manipulate and optimise the relationship of the ground reaction force (GRF) to the knee and hip. Regardless of whether the AFO had been cast in a dorsiflexed, neutral or plantarflexed alignment, optimal knee and hip kinetics were always achieved with some angle of tibial inclination. It is interesting to note that the mean angle of tibial inclination was 11.86^0 which approximates to the angle observed at midstance in normal gait. Stallard [70] also used the portable VVG to investigate the effect of tuning SAFOs, reporting that tuning improved the biomechanical alignment in the majority of subjects. Based on this experience the authors suggest that tuning with kinematic/kinetic monitoring should become routine clinical practice, but stress that if the alignment of the GRF is to be successfully influenced, the AFOs must have appropriate mechanical properties, i.e. they must be appropriately stiff. Butler [71] also concluded that tuning SAFOs can improve knee kinetics and kinematics, advocating that pre-bracing knee kinematics is a good predictor of those most likely to benefit from tuning. Greatest improvements were strongly associated with knee flexion no greater than 20^0 in the first third of stance, combined with movement towards knee extension in the second third of stance to 10^0 flexion or less. Attempts at tuning those with knee flexion of greater than 35^0 in the first third of stance and greater than 15^0 degrees in mid stance were reported to be unsuccessful. A popliteal angle in excess of 45^0 was a poor prognostic sign, as was hip flexion contracture greater than 15^0. The authors conclude that it may be necessary to address proximal problems of tightness and control at hip and knee before attempting tuning. They also suggest that the presence of ataxia was a barrier to successful tuning. In a single case study, Jagadamma[43] used gait analysis to investigate the effects of an incrementally wedged SAFO on hemiplegic knee joint kinetics and kinematics. Unfortunately, no details were provided on the characteristics of either the SAFO or footwear, which are critical issues when tuning. In this study, optimal knee alignment was found with tibial inclination of 13^0. Van Gestel [13] reported that each of the three AFOs fitted to hemiplegic subjects was “optimally tuned”, defined as having the shank in an alignment ranging from neutral to maximum 10 degrees inclined. How the orthoses were judged to have been successfully tuned in this context is not described. As dorsiflexion was permitted by all the orthoses investigated, the tuning process can only have been of benefit in early stance phase.

In some cases, footwear modifications may assist in optimising terminal stance kinetics [72]. Optimal GRF alignment at this stage of gait is anterior to the knee and posterior to the hip, as seen in normal gait. With excessive tibial inclination, as seen in crouch gait, this GRF alignment may become impossible, thereby compromising stability. While a rigid SAFO can help retard tibial progression, it may still prove inadequate if the normal anatomical 3rd rocker allows the knee centre to pass in front of the GRF too early in terminal stance. Modification of the footwear, to block the normal 3rd rocker, replacing it with a more anteriorly placed “point-loading rocker”,

has been shown to further improve resistance to tibial progression, and enable optimal knee and hip kinetics. These point loading rockers were most successful with their apex located in front of the metatarsal-phalangeal joints. Nuzzo [73] successfully used an extended posterior heel flare shoe adaptation, with a SAFO set at an angle of 7 – 10 degrees of tibial inclination, to treat knee hyperextension at initial contact, although the approach was unsuccessful in the presence of athetosis.

Conclusions

The issue of tuning AFOs to optimise their function has been addressed by a number of investigators. There is emerging evidence that tuning AFOs can significantly improve their effectiveness, with beneficial effects reported at the proximal joints. However, in order to achieve benefit, the orthoses must be cast in a position that accommodates any gastrocnemius shortening, and they must be appropriately stiff, or must block ankle motion at appropriate angles, so that the ground reaction vector can be successfully manipulated. Some angle of tibial inclination appears to be most appropriate, regardless of whether the AFOs have been cast in a plantarflexed or dorsiflexed angle. Proximal contractures and/or spasticity may need to be addressed prior to tuning. Footwear modifications can also be beneficial in this process, in order to optimise entrance to and exit from stance.

CAN LOWER LIMB ORTHOSES INFLUENCE THE UPPER LIMB?

Very few studies [12, 17, 48] have reported changes in upper limb function when subjects wear AFOs. Brunner [12] investigated the effect of a SAFO and a flexible AFO on hemiplegic gait, reporting improvement in upper limb movement during gait only with the SAFO. Matthews [48] investigated the effect of a HAFO incorporating neurophysiological features and a SAFO on a single hemiplegic subject. While upper limb spasticity was reportedly reduced, it is unclear whether this effect was attributable to the hinged rather than solid design, or to the neurophysiological features in the HAFO. Only Buckon [17] actually measured the effect on performance of upper limb functional tasks in subjects with diplegia using HAFOs, SAFOs and PLS. Upper limb coordination, speed and dexterity all significantly improved with all AFO designs.

Conclusions

It is inappropriate to draw conclusions from the very small amount of evidence that exists in this area.

WHAT ARE THE PERCEPTIONS OF USERS AND PARENTS REGARDING ORTHOTIC TREATMENT?

Only one study [74] was identified that investigated the perceptions of parents of children receiving lower limb orthotic treatment. The parents of children with spastic diplegia using DAFOs were invited to open-ended interviews in order to elicit their responses. The parents' perceptions were that the DAFOs had contributed to mechanical changes in posture, making the foot and ankle more stable, and enabling better postural control and alignment. They also felt that they had contributed to improvements in ADL. Additionally, some felt that the DAFOs had a beneficial effect on the leg muscles, which required less stretching. These generally favourable results may have been biased by the fact that the interviews were conducted by physiotherapists involved in the treatment.

Conclusions
It is important to ascertain the opinions of orthosis users and their carers. Little evidence exists at present, and further research is required in this area.

OBSERVATIONS ON THE REVIEW

The authors of this review feel compelled to make a number of disappointing observations on the available literature. There is evidence that lower limb orthoses can confer a number of beneficial effects when treating cerebral palsy. However, before clinicians can attempt to make use of this (or indeed any) evidence in the treatment of patients, there are a couple of very important questions that must be addressed:

- Are the patients to be treated the same as (or similar to) the study patients?
- Can the orthotic intervention in the study be accurately reproduced?

Unfortunately, many of the reviewed papers failed to provide important clinical details about the subjects in the study. In the worst cases, subjects are described as "children with CP" as if this was a homogeneous population or as having hemiplegia, or diplegia. Differentiating between types (e.g. hemiplegia and diplegia) is only a start in visualising the subjects, but more information is clearly needed. Clinicians appreciate the critical importance of many interrelated factors when prescribing and fitting lower limb orthoses, including joint range of motion, contracture, muscle tone and spasticity. Too many papers failed to provide adequate information on these factors. In studies where range of motion was discussed, the focus was primarily on ankle dorsiflexion, and often it was not made clear whether this was achieved with the knee flexed or extended. This information is of critical importance. If dorsiflexion (to 90^0 for example) can only be achieved with the knee flexed due to shortening or hypertonus in the gastrocnemius, then an AFO that positions the ankle at this angle will actually limit knee extension. It was disappointing to note that a number of studies reported that subjects with plantarflexion contractures were provided with AFOs that held the ankle at 90^0. Furthermore, when HAFOs are used in cases like this, any dorsiflexion that occurs can only do so at the expense of knee extension. Ignoring gastrocnemius shortening in this way is inevitably detrimental to the kinetics and kinematics at the knee and at the hip. High tone and/or spasticity in gastrocnemius can have similar effects, if not addressed. Proximal contractures, whether true or dynamic, also have a profound influence on outcomes, particularly contractures in the biarticular muscles. All too frequently these issues are not discussed. Confusion is compounded in those studies on non-homogenous groups in which results are not presented separately.

Remarkably, many studies provided inadequate information on the orthosis being investigated, making reproduction of the intervention a challenge. In the worst examples, the intervention was only described as "an AFO". Some papers used confusing terms to describe the type of orthosis being investigated, such as "conventional AFO" and "standard AFO". Such terms are ambiguous, meaning different things to different people, and should be avoided. To some the term "dynamic AFO" clearly means a supramalleolar orthosis with tone reducing features such as a contoured footplate, while to others it means a "spring-like" orthosis, perhaps of a spiral design, rendering the term potentially misleading. The term "independent ambulation" is also ambiguous. In most studies this term was used to

describe ambulation without any walking aids, but in some studies the "independent ambulators" used crutches or walking sticks.

A large percentage of the reviewed papers failed to explicitly state the angle of plantarflexion or dorsiflexion at which the orthosis positioned the foot and ankle, and very few specifically mentioned how the orthosis controlled the subtalar joint or the midtarsal joint. In general, there appeared to be little or no attempt to quantify "fit" which underpins optimal orthotic treatment. Information on fitting complications and rejection rates was rarely provided. Information on the materials used in the construction of the orthosis was seldom provided. The quality of a number of papers studying SAFOs was compromised by the fact that these "solid" AFOs were able to deflect into dorsiflexion (and perhaps even plantarflexion). Rarely were attempts made at measuring this deflection, making the results questionable. Worryingly, some authors actually described this deflection as intentional, ignoring the fact that as the AFOs buckle, the intimacy of their fit is inevitably compromised, with detrimental effects on the alignment of unstable subtalar and midtarsal joints. In the papers investigating the use of HAFOs, there was often inadequate detail on the actual arc of motion permitted by the ankle joints. In many cases there was no specific statement that the HAFO blocked plantarflexion, merely a suggestion that this might be the case.

A number of papers correctly highlighted the fact that footwear has an impact on biomechanics, but only rarely did papers provide adequate information on the footwear being used with the orthoses. Some authors made no mention of the footwear at all, while others simply stated that "tennis shoes" or "training shoes" were used. The effective heel height of the footwear, i.e. the difference between the thickness of the heel and the sole, is an important factor but this information was rarely provided. Knowledge of the effective heel height, combined with the AFO angle, is essential if we are to appreciate the resulting tibial inclination angle, which is the critical issue when considering whether the orthoses are appropriately "tuned". The angle of tibial inclination is only rarely mentioned in the literature. Little detail was provided on footwear modifications, which can also have a significant impact on biomechanics. Some papers did not explicitly state whether controls wore shoes or were barefoot and there appeared to be no consensus regarding this.

RECOMMENDATIONS

To ensure that future research has optimal influence on clinical practice, it is important that detailed information is routinely provided on the subjects being studied. This should include information on:

- diagnosis
- age
- passive range of motion at all lower limb joints
- "dynamic" range of motion i.e. range observed when tested at speed to elicit the effects of tone and spasticity
- detailed information on the multi-joint muscles (gastrocnemius, hamstrings and rectus femoris) with the gastrocnemius being perhaps the most critical when designing AFOs
- torsional abnormalities affecting the foot progression angle
- prior surgery
- use of BTA within previous six months

Details of the orthotic intervention should be comprehensive and explicit, and should include:

- the type of the orthosis
- the angle at which the orthosis positions the ankle joint, and how this relates to the length of the gastrocnemius
- the alignment of the subtalar and midtarsal joints
- the range of ankle joint motion (plantarflexion and/or dorsiflexion) being permitted in HAFOs, but also in other flexible AFO designs
- information on the rigidity of SAFOs, and details of any motion that is occurring
- the type and thickness of the materials used to construct the orthosis
- details on footwear specification and modification (if any), specifically, the effective heel height (i.e. the difference between heel and sole thickness).

Details on the research methodology should also be provided, including

- whether the fit of different orthoses has been standardised by fabricating from the same plaster casts
- order of testing
- whether tests have been conducted on the same or different days
- period of acclimatisation
- subgroup analysis, if appropriate
- whether the subjects have been investigated with or without walking aids
- whether the control group has been investigated with or without shoes

If this level of detail on the subjects and interventions under investigation is routinely provided, and ambiguous terminology can be avoided, then research can be more effectively converted into clinical practice, to the benefit of all orthosis users.

It is particularly disappointing to realise that many of these same recommendations were made in the ISPO "*Report of a consensus conference on the orthotic management of stroke patients*" published in 2004.

Table 1. Do lower limb orthoses influence the temporal and spatial parameters of gait?

First Author, year	Number and age range of subjects	Type of subjects	Type of Orthosis	Research Design	Evidence Level	Key Findings
Hemiplegia						
Balaban, 2007	N=11 mean age 7.18±1.16	Hemiplegia	HAFO	Prospective Within subject comparison study	4	velocity improved cadence unchanged stride length improved single support improved
Brunner,1998	N=14 median age 11.42 (range 6.46-20.08)	Hemiplegia	SAFO Flexible AFO	Prospective Crossover study	4	velocity improved (both) cadence decreased (both) step length increased(both) stride length improved (both); greater improvement in flexible AFO single support increased SAFO; no change flexible AFO
Buckon, 2001	N=30 mean age 9y4m (range 4-18)	Hemiplegia	HAFO SAFO PLS	Prospective Crossover study	4	velocity no significant change cadence slight decrease step length improved stride length improved
Desloovere, 2006	N=15 mean age 5.86 years ±1.76	Hemiplegia	PLS Dual carbon spring AFO	Prospective Crossover study	4	velocity improved both designs vs. barefoot; not significant vs. shoes cadence reduced both vs. barefoot; not significant vs. shoes; shoes alone cadence decreased step length improved both vs. barefoot; not significant vs. shoes stride length increased
Romkes, 2002	N=12 mean age 11.9±4.9	Hemiplegia	HAFO DAFO	Prospective Crossover study	4	step length improved in both; no difference between orthoses stride length improved in both; no difference between orthoses
Romkes, 2006	N=10 mean age 9.7±1.6	Hemiplegia	HAFO	Retrospective Within subject comparison study	4	velocity improved cadence decreased step length improved, stride length improved single support unaffected
Thompson, 2002	N=18 mean age 8y5m range 5y8m-11y	Hemiplegia	SAFO	Prospective Within-subject comparison	4	velocity improved cadence decreased step length increased
Van Gestel, 2008	N=36 mean age 8y5m±2y8m range 4-14	Hemiplegia	PLS Dual carbon spring AFO Orteam® AFO	Retrospective Cohort study	2b	velocity improved all cadence decreased all step length increased all

Table 1. Do lower limb orthoses influence the temporal and spatial parameters of gait? (continued)

First Author, year	Number and age range of subjects	Type of subjects	Type of Orthosis	Research Design	Evidence Level	Key Findings
Diplegia						
Abel, 1998	N=35 mean age 8.7 range 2.5-19	Diplegia	SAFO	Retrospective Within subject comparison study	4	velocity improved cadence unchanged stride length improved single support improved double support decreased
Buckon, 2004	N=16 mean age 8 y4m ± 2y4m (range 4y4m-11y6m)	Diplegia	HAFO SAFO PLS	Prospective Crossover study	4	velocity no significant change in; slower in HAFOs than PLS cadence decreased all step length increased all stride length improved all
Carlson, 1995 (abstract)	N=11 age range 4-11	Diplegia	SAFO SMO	Prospective Crossover study	4	velocity no significant effect cadence no significant effect
Carlson, 1997	N=11 age not stated	Diplegia	SAFO SMO	Prospective Crossover study	4	velocity no significant change cadence decreased SMO stride length improved both
Crenshaw, 2000	N=8 age 8.9±2.4 (range 4-11)	Diplegia	HAFO HAFO + tone reducing features SMO + tone reducing features	Prospective Crossover study	4	stride length increased in HAFO + tone reducing features
Kornhaber, 2006 (abstract)	N=4 age range 4-15	Diplegia	SMO HAFO	Prospective Crossover study	4	children using assistive devices faster in SMOs than HAFOs stride length improved in children using assistive devices
Lam, 2005	N=13 average age 5.9±1.81 range 3.3-9.7	Diplegia	SAFO DAFO	Prospective Crossover study	4	velocity no significant effect cadence no effect stride length improved both
Mossberg, 1990	N=18 mean age 8.3±2.8 range 3-14	Diplegia	AFOs	Retrrospective Within subject comparison study	4	faster without AFOs, not significant

Table 1. Do lower limb orthoses influence the temporal and spatial parameters of gait? (continued)

First Author, year	Number and age range of subjects	Type of subjects	Type of Orthosis	Research Design	Evidence Level	Key Findings
Radtka, 2005	N=12 average age 7.5±3.83 range 4-16	Diplegia	SAFO HAFO	Prospective Crossover study	4	velocity no significant effect cadence no significant effect stride length improved both
Rethlefsen, 1999	N=21 average age 9.1±2.2 range 5.3-13.5	Diplegia	SAFO HAFO	Prospective Crossover study	4	velocity no significant effect cadence no significant effect stride length no significant effect double support increased HAFOs
Smiley, 2002	N=14 average age 10.7 range 6.9-16	Diplegia	SAFO HAFO PLS	Prospective Crossover study	4	velocity slight increase HAFOs and PLS, slight reduction SAFO; not significant cadence slightly reduced HAFO and SAFO, slightly increased PLS; not significant stride length improved HAFO and PLS; SAFO slightly reduced
Van Rooijen, 2006 (abstract)	N=12 mean age 11.1	Diplegia	SAFO	Retrrospective Within subject comparison study	4	stride length improved
Mixed						
De Groot, 2006 (abstract)	N=13 mean age 11.6 range 7-16	Mixed	Shoes only	Prospective Within subject comparison study	4	cadence unaffected stride increased step length increased stride length increased
Dursun, 2002	N=24 mean age 6.6±0.73	Mixed	AFO not described	Prospective Within subject comparison study	4	velocity increased cadence no effect stride length increased
Hayek, 2007	N=56 mean age 8.8 ± 3.5 range 4-17	Mixed	SAFO HAFO	Retrrospective Within subject comparison study	4	velocity increased both; marginally in hemiplegics cadence decreased in hemiplegic, not diplegic step length increased both stride length increased both double support decreased in diplegic group

Table 1. Do lower limb orthoses influence the temporal and spatial parameters of gait? (continued)

First Author, year	Number and age range of subjects	Type of subjects	Type of Orthosis	Research Design	Evidence Level	Key Findings
Hobbs, 2003 (abstract)	N=24 age not stated	Mixed	SAFO HAFO SMO	Retrrospective Within subject comparison study	4	step length increased
Huenaerts, 2004 (abstract)	N=23 age not stated	Mixed	PLS (effect of BTA)	Retrospective Within subject comparison study	4	velocity improved PLS velocity no change after BTA step length increased after BTA cadence decreased after BTA step length increased PLS, further improvement post BTA
Kirkeide, 1999 (abstract)	N=5 mean age 5.2	Mixed	DAFO SAFO	Study design unclear		velocity no change cadence reduced both (vs barefoot)
Molenaers, 2006 (abstract)	N=30 mean age 9.0±4.9 range 4-14	Mixed	PLS	Retrospective Within subject comparison study	4	velocity increased cadence decreased step length increased
Radtka, 1997	N=10 average age 6.5±1.86 range 3.5-8.5	Mixed	SAFO DAFO	Prospective Crossover study	4	Significant decrease in cadence with both. Both increased stride length
Van de Walle, 2005 (abstract)	N=9 age range 6-10	Mixed	PLS	Within subject comparison study	4	velocity increased slightly, not significant
White, 2002	N=115 mean age 9 range 5-15	Mixed	HAFO SAFO Standard Polypropylene Wrap-around polyethylene	Retrospective Within subject comparison study	4	velocity improved all cadence slight decrease in (not significant); greatest in GMFCS I and II step length improved all stride length improved all single support improved all

Table 2. What are the effects of orthoses on the ankle and foot?

First Author, year	Number and age range of subjects	Type of subjects	Type of Orthosis	Research Design	Evidence Level	Key Findings
Hemiplegia						
Balaban, 2007	N=11 mean age 7.18+1.16	Hemiplegia	HAFO	Prospective Within subject comparison study	4	ankle power decreased equinus corrected
Brunner, 1998	N=14 median age 11.42 (range 6.46-20.08)	Hemiplegia	SAFO Flexible AFO	Prospective Crossover study	4	heel toe pattern reinstated both
Buckon, 2001	N=30 mean age 9y4m (range 4-18)	Hemiplegia	HAFO SAFO PLS	Prospective Crossover study	4	ankle power decreased all equinus improved all (HAFO & SAFO best) peak stance dorsiflexion improved all ankle dynamic range improved all dorsiflexion greater in all peak dorsiflexion greatest HAFO
Desloovere, 2006	N=15 mean age 5.86 years ±1.76	Hemiplegia	PLS Dual carbon spring AFO	Prospective Crossover study	4	power generation better in pre-swing dual carbon spring AFO; better power absorption in loading response PLS equinus and 1st and 2nd rocker improved both angular velocity in pre-swing better dual carbon orthosis ankle moment and power absorption in loading response more normal PLS ankle motion and angular velocity also improved shoes
Romkes, 2002	N=12 mean age 11.9±4.9	Hemiplegia	HAFO DAFO	Prospective Crossover study	4	power absorption significantly reduced both power generation unchanged by either dorsiflexion at initial contact and peak dorsiflexion increased in both
Romkes, 2006	N=10 mean age 9.7±1.6	Hemiplegia	HAFO	Retrospective Within subject comparison study	4	equinus corrected

Table 2. What are the effects of orthoses on the ankle and foot? (continued)

First Author, year	Number and age range of subjects	Type of subjects	Type of Orthosis	Research Design	Evidence Level	Key Findings
Thompson, 2002	N=18 mean age 8y5m range 5y8m-11y	Hemiplegia	SAFO	Prospective Within subject comparison study	4	equinus corrected
Van Gestel, 2008	N=36 mean age 8y5m±2y8m range 4-14	Hemiplegia	PLS Dual carbon spring AFO Orteam® AFO	Retrospective Cohort study	2b	equinus corrected in all; only significant dual carbon spring 3rd rocker most normal dual carbon spring
Diplegia						
Abel, 1998	N=35 mean age 8.7 range 2.5-19	Diplegia	SAFO	Retrospective Within subject comparison study	4	ankle power decreased ankle excursion improved
Bennett, 2005 (abstract)	N=15 Mean age (experiment 1) 9y8m (experiment 2) 7y11m	Diplegia	AFO not described	Prospective Within subject comparison study	4	peak push off moment increased
Buckon, 2004	N=16 mean age 8 y4m ± 2y4m (range 4y4m-11y6m)	Diplegia	HAFO SAFO PLS	Prospective Crossover study	4	power generation greatest reduction SAFO; least in PLS timing of peak power generation improved in all
Carlson, 1997	N=11 age not stated	Diplegia	SAFO SMO	Prospective Crossover study	4	propulsive moments improved SAFO ankle power at push off increased neither power generated and absorbed decreased SAFO equinus improved SAFO, not SMO
Chambers, 1999 (abstract)	N=9 mean age 8.2 range 6-11	Diplegia	DAFO "Standard AFO"	Prospective Crossover study	4	power generated at push off decreased "Standard AFO" power generation in DAFO similar to shoes only ankle motion restricted both, standard AFO better (difference between orthoses not statistically significant)

Table 2. What are the effects of orthoses on the ankle and foot? (continued)

First Author, year	Number and age range of subjects	Type of subjects	Type of Orthosis	Research Design	Evidence Level	Key Findings
Crenshaw, 2000	N=8 age 8.9±2.4 (range 4-11)	Diplegia	HAFO HAFO + tone reducing features SMO + tone reducing features	Prospective Crossover study	4	plantarflexion moments increased HAFO and tone reducing HAFO power absorption increased HAFO power generation greatest in shoes only plantarflexion significantly reduced both HAFO designs vs. SMO or shoes
Dole, 1997 (abstract)	N=1 age 5	Diplegia	HAFO	Single case study	5	foot progression angle improved
Hassani, 2004	N=16 mean age 7.5±2.9	Diplegia	HAFO DAFO	Prospective Crossover study	4	significant effect on plantarflexion moment with both significant increase in peak stance dorsiflexion and peak swing plantarflexion in both no significant difference between orthoses
Lam, 2005	N=13 average age 5.9±1.81 range 3.3-9.7	Diplegia	SAFO DAFO	Prospective Crossover study	4	maximum plantarflexion moment improved both higher 2nd peak of GRF enabled in SAFO quines improved both (SAFO better) and peak mean dorsiflexion
Lucareli, 2007	N=71 average age 12.2±3.9	Diplegia	Hinged FRAFO	Retrospective within subject comparison study	4	dorsiflexion decreased
Radtka, 2005	N=12 average age 7.5±3.83 range 4-16	Diplegia	SAFO HAFO	Prospective Crossover study	4	increased power generation HAFOs vs. SAFOs peak ankle power with HAFOs similar to no AFO dorsiflexion improved both dorsiflexion at terminal stance greater HAFO
Rethlefsen, 1999	N=21 average age 9.1±2.2 range 5.3-13.5	Diplegia	SAFO HAFO	Prospective Crossover study	4	power generation higher HAFOs dorsiflexion improved at initial contact both dorsiflexion at terminal stance greater in HAFO
Smiley, 2002	N=14 average age 10.7 range 6.9-16	Diplegia	SAFO HAFO PLS	Prospective Crossover study	4	all designs appeared to produce an abnormal amount of dorsiflexion across gait cycle

Table 2. What are the effects of orthoses on the ankle and foot? (continued)

First Author, year	Number and age range of subjects	Type of subjects	Type of Orthosis	Research Design	Evidence Level	Key Findings
Mixed						
Desloovere, 1999 (abstract)	N=47 age range 5-12	Mixed	Klenzak AFO SAFO HAFO PLS	Retrospective Within subject comparison study	4	dorsiflexion increased at initial contact plantarflexion reduced at push off
Hainsworth, 1997	N=12 mean age 4y8m range 3y11m-7y5m	Mixed	SAFO HAFO	Prospective Within subject comparison	4	ankle kinematics improved
Hayek, 2007	N=56 mean age 8.8 ± 3.5 range 4-17	Mixed	SAFO HAFO	Retrrospective Within subject comparison	4	dorsiflexion improved in both; type of AFO had borderline significance
Hobbs, 2003 (abstract)	N=24 age not stated	Mixed	SAFO HAFO SMO	Retrrospective Within subject comparison	4	dorsiflexion in terminal stance increased HAFO and SMO
Kirkeide, 1999 (abstract)	N=5 mean age 5.2	Mixed	DAFO SAFO	Study design unclear		forefoot force decreased DAFO; increased with SAFO; ankle excursion and timing reduced both heel contact time increased both (DAFO better)
Molenaers, 1999 (abstract)	N=33 age range 4-9	Mixed	AFO not described BTA	Retrospective Cohort study	2b	power generation increased post BTA
Molenaers, 2006 (abstract)	N=30 mean age 9.0±4.9 range 4-14	Mixed	PLS	Retrospective Within subject comparison	4	ankle power decreased ankle position improved
Ounpuu, 1996	N=31 mean age 10.6±3.8	Mixed	PLS	Retrospective Within subject comparison	4	mechanical energy not stored or returned in propulsive phase terminal swing and end stance dorsiflexion improved peak stance dorsiflexion no effect
Radtka, 1997	N=10, age 6.5±1.86 range 3.5-8.5	Mixed	SAFO DAFO	Prospective Crossover study	4	plantarflexion decreased with both
Westberry, 2007	N=102 mean age 10 range 5.1-18.7	Mixed	SAFO PLS HAFO GRAFO	Retrospective Within subject comparison study	4	all measurements of segmental alignment changed (statistically significant) improvements considered clinically insignificant by authors correction most successful in pronated feet

Table 3. What are the indirect effects of orthoses on the knee?

First Author, year	Number and age range of subjects	Type of subjects	Type of Orthosis	Research Design	Evidence Level	Key Findings
Hemiplegia						
Balaban, 2007	N=11 mean age 7.18+1.16	Hemiplegia	HAFO	Prospective Within subject comparison study	4	knee flexion reduced at initial contact maximum knee flexion in stance, or in swing no change
Brunner,1998	N=14 median age 11.42 (range 6.46-20.08)	Hemiplegia	SAFO Flexible AFO	Prospective Crossover study	4	maximum knee extension reduced SAFO (vs barefoot) knee range of motion reduced throughout gait SAFO flexible AFO had only a slight influence
Buckon, 2001	N=30 mean age 9y4m (range 4-18)	Hemiplegia	HAFO SAFO PLS	Prospective Crossover study	4	knee hyperextension improvement greater with HAFO vs. SAFO
Desloovere, 2006	N=15 mean age 5.86 years ±1.76	Hemiplegia	PLS Dual carbon spring AFO	Prospective Crossover study	4	knee shock absorption and maximum knee flexion in stance vs. barefoot improved both amount and timing of maximum knee flexion in swing changed both vs. barefoot maximum knee flexion and timing of peak knee flexion in swing unchanged vs shoes minimal hyperextension allowed in PLS knee flexion velocity at toe-off increased both, particularly PLS knee shock absorption, flexion in swing and maximal knee extension in stance increased in shoes only vs barefoot
Jagadamma, 2007 (abstract)	N=1 age 12	Hemiplegia	SAFO	Within subject comparison study	5	knee alignment improved by addition of wedging under SAFO optimum result with 13^0 of tibial inclination
Romkes, 2002	N=12 mean age 11.9±4.9	Hemiplegia	HAFO DAFO	Prospective Crossover study	4	timing of peak knee extension significantly delayed by both peak knee flexion improved in both
Romkes, 2006	N=10 mean age 9.7±1.6	Hemiplegia	HAFO	Retrospective Within subject comparison study	4	knee kinematics improved in loading response flexion-extension pattern created

Table 3. What are the indirect effects of orthoses on the knee? (continued)

First Author, year	Number and age range of subjects	Type of subjects	Type of Orthosis	Research Design	Evidence Level	Key Findings
Van Gestel, 2008	N=36 mean age 8y5m±2y8m range 4-14	Hemiplegia	PLS Dual carbon spring AFO Orteam® AFO	Retrospective Cohort study	4	maximum stance knee extension decreased PLS and an Orteam®AFO hyperextension encouraged in Dual Carbon Fibre Spring AFO knee velocity in swing and peak knee flexion increased in all orthoses maximal knee flexion decreased, but remained higher than normal. knee shock absorption improved.
Diplegia						
Abel, 1998	N=35 mean age 8.7 range 2.5-19	Diplegia	SAFO	Retrospective Within subject comparison study	4	knee excursion improved effect on knee in stance not significant
Buckon, 2004	N=16 mean age 8 y4m ± 2y4m (range 4y4m-11y6m)	Diplegia	HAFO SAFO PLS	Prospective Crossover study	4	knee kinematics no change knee extensor moment in early stance increased with HAFOs
Carlson, 1997	N=11 age not stated	Diplegia	SAFO SMO	Prospective Crossover study	4	mean stance knee extension improved in SAFO
Farmer, 1999	N=1 age 7y10m	Diplegia	SAFO	Single case study	5	improvements in knee flexion angle (standing and walking) by progressively reducing angle of tibial inclination combined with throwing and catching exercises
Hassani, 2004	N=16 mean age 7.5±2.9	Diplegia	HAFO DAFO	Prospective Crossover study	4	peak knee flexion in stance and swing significant differences vs. barefoot peak knee extension in stance or swing no significant difference with either orthosis
Lam, 2005	N=13 average age 5.9±1.81 range 3.3-9.7	Diplegia	SAFO DAFO	Prospective Crossover study	4	knee flexion significantly increased at initial contact DAFOs vs. barefoot knee flexion at initial contact reduced with SAFO
Lucareli, 2007	N=71 average age 12.2±3.9	Diplegia	Hinged FRAFO	Retrospective within subject comparison	4	knee flexion reduced significantly in more involved subjects

Table 3. What are the indirect effects of orthoses on the knee? (continued)

First Author, year	Number and age range of subjects	Type of subjects	Type of Orthosis	Research Design	Evidence Level	Key Findings
Radtka, 2005	N=12 average age 7.5±3.83 range 4-16	Diplegia	SAFO HAFO	Prospective Crossover study	4	excessive knee flexion in barefoot stance unchanged with either knee moments during stance also unaffected by either
Rethlefsen, 1999	N=21 average age 9.1±2.2 range 5.3-13.5	Diplegia	SAFO HAFO	Prospective Crossover study	4	knee kinematics no significant differences between test conditions terminal stance knee flexion greater HAFO
Smiley, 2002	N=14 average age 10.7 range 6.9-16	Diplegia	SAFO HAFO PLS	Prospective Crossover study	4	maximum knee flexion in swing statistically significant differences in HAFO and SAFO, but not PLS
Wesdock, 2003	N=11 average age 7±2.6 range 4-13.5	Diplegia	SAFO Plus wedges	Prospective within subject comparison study	4	effects on knee extension not significant
Mixed						
Desloovere, 1999 (abstract)	N=47 age range 5-12	Mixed	Klenzak AFO SAFO HAFO PLS	Retrospective Within subject comparison study	4	knee flexion during loading response significantly increased AFOs vs barefoot more pronounced knee extension moment during stance in SAFO
Hayek, 2007	N=56 mean age 8.8 ± 3.5 range 4-17	Mixed	SAFO HAFO	Retrrospective Within subject comparison study	4	knee flexion at initial contact reduced in hemiplegic group; no change in diplegic group knee kinematics in loading response, single support or swing phase no significant effect in either orthosis
Huenaerts, 2004 (abstract)	N=23 age not stated	Mixed	PLS (effect of BTA)	Retrospective Within subject comparison study	4	knee kinematics improved in PLS after BTA, but GRF alignment to the knee not always appropriate

Table 3. What are the indirect effects of orthoses on the knee? (continued)

First Author, year	Number and age range of subjects	Type of subjects	Type of Orthosis	Research Design	Evidence Level	Key Findings
Lampe, 2004	N=18 (not all CP) age range 15-18	Mixed	Orthopaedic shoes Insoles DAFO Ring orthosis AFO	Retrospective within subject comparison study	4	15^0 knee extension deficit in stance increased to 25^0 using orthopaedic shoes in child with diplegia duration of knee joint extension increased markedly by shoes; extension moment in initial stance decreased slightly. early stance knee extension improved in shoe plus orthotic insole in child with hemiplegia vs. barefoot knee extension and knee stability in stance improved; increased swing knee flexion with AFO in diplegic child
Molenaers, 1999 (abstract)	N=33 age range 4-9	Mixed	AFO not described BTA	Retrospective Cohort study	4	stance knee kinematics improved post BTA in all children, except those who already had casting prior to BTA

Table 4. What are the indirect effects of orthoses on the hip?

First Author, year	Number and age range of subjects	Type of subjects	Type of Orthosis	Research Design	Evidence Level	Key Findings
Hemiplegia						
Brunner,1998	N=14 median age 11.42 (range 6.46-20.08)	Hemiplegia	SAFO Flexible AFO	Prospective Crossover study	4	hip flexion, extension and abduction increased SAFO, but adduction decreased changes to abduction and adduction were more pronounced in the FAFO; arcs of hip flexion and extension similar to SAFO
Desloovere, 2006	N=15 mean age 5.86 years ±1.76	Hemiplegia	PLS Dual carbon spring AFO	Prospective Crossover study	4	coronal hip angle and pelvic obliquity in swing increased shoes only (vs. barefoot)
Romkes, 2006	N=10 mean age 9.7±1.6	Hemiplegia	HAFO	Retrospective Within subject comparison study	4	hip flexion at initial contact increased
Van Gestel, 2008	N=36 mean age 8y5m±2y8m range 4-14	Hemiplegia	PLS Dual carbon spring AFO Orteam® AFO	Retrospective Cohort study	4	hip range of motion, hip moments and power improved hip moment and power graphs all significantly increased, except maximal abduction moment in stance and maximal power generation at pre-swing significantly higher maximal hip flexion moment in stance in CFO; improvement also reported in about half of hip kinematics. maximal hip extension in stance increased in PLS and carbon fibre orthosis; Orteam® orthosis decreased it
Diplegia						
Abel, 1998	N=35 mean age 8.7 range 2.5-19	Diplegia	SAFO	Retrospective Within subject comparison study	4	hip excursion improved
Buckon, 2004	N=16 mean age 8 y4m ± 2y4m (range 4y4m-11y6m)	Diplegia	HAFO SAFO PLS	Prospective Crossover study	4	no changes at the hip

Table 4. What are the indirect effects of orthoses on the hip? (continued)

First Author, year	Number and age range of subjects	Type of subjects	Type of Orthosis	Research Design	Evidence Level	Key Findings
Crenshaw, 2000	N=8 age 8.9±2.4 (range 4-11)	Diplegia	HAFO HAFO + tone reducing features SMO + tone reducing features	Prospective Crossover study	4	increase in maximum extension moment and total power generation at hip in AFOs without tone reducing features
Hassani, 2004	N=16 mean age 7.5±2.9	Diplegia	HAFO DAFO	Prospective Crossover study	4	significant differences noted in peak hip flexion in stance with both vs. barefoot, peak hip extension in stance and peak hip flexion in swing no significant difference
Lam, 2005	N=13 average age 5.9±1.81 range 3.3-9.7	Diplegia	SAFO DAFO	Prospective Crossover study	4	hip flexion at initial contact significantly increased DAFOs vs barefoot
Lucareli, 2007	N=71 average age 12.2±3.9	Diplegia	Hinged FRAFO	Retrospective within subject comparison study	4	maximum hip extension no significant changes
Mixed						
Desloovere, 1999 (abstract)	N=47 age range 5-12	Mixed	Klenzak AFO SAFO HAFO PLS	Retrospective Within subject comparison study	4	hip extension increased at terminal stance and hip abduction decreased during swing PLS and HAFOs vs. barefoot hip rotation no observed AFOs effect
De Groot, 2006 (abstract)	N=13 mean age 11.6 range 7-16	Mixed	Shoes only	Prospective Within subject comparison study	4	minimum hip flexion in stance decreased hip range of motion increased
Hayek, 2007	N=56 mean age 8.8 ± 3.5 range 4-17	Mixed	SAFO HAFO	Retrrospective Within subject comparison study	4	hip kinematics minimal effect

Table 4. What are the indirect effects of orthoses on the hip? (continued)

First Author, year	Number and age range of subjects	Type of subjects	Type of Orthosis	Research Design	Evidence Level	Key Findings
Hobbs, 2003 (abstract)	N=24 age not stated	Mixed	SAFO HAFO SMO	Retrrospective Within subject comparison study	4	hip extension during stance and hip flexion in swing improved SAFOs hip flexion detrimentally increased relative to normal HAFOs and SMOs
Huenaerts, 2004 (abstract)	N=23 age not stated	Mixed	PLS (effect of BTA)	Retrospective Within subject comparison study	4	internal rotation of hip at initial contact significantly decreased PLS further improvement at midstance and terminal stance after BTA hip extension increased after BTA
Molenaers, 1999 (abstract)	N=33 age range 4-9	Mixed	AFO not described BTA	Retrospective Cohort study	4	greater hip extension at terminal stance after BTA internal hip rotation in stance and swing significantly decreased for all patients after BTA
Molenaers, 2006 (abstract)	N=30 mean age 9.0±4.9 range 4-14	Mixed	PLS	Retrospective Within subject comparison study	4	hip extension throughout stance improved
Van Rooijen, 2006 (abstract)	N=12 mean age 11.1	Diplegia	SAFO	Retrrospective Within subject comparison study	4	minimum hip flexion in stance decreased

Table 5. What are the indirect effects of orthoses on the pelvis?

First Author, year	Number and age range of subjects	Type of subjects	Type of Orthosis	Research Design	Evidence Level	Key Findings
Hemiplegia						
Brunner, 1998	N=14 median age 11.42 (range 6.46-20.08)	Hemiplegia	SAFO Flexible AFO	Prospective Crossover study	4	pelvic obliquity improved SAFO; no effect on pelvic tilt; adversely affected (i.e. increased) pelvic rotation; obliquity improved in flexible AFO; slightly increased pelvic tilt; adverse effect on pelvic rotation less than the SAFO
Romkes, 2006	N=10 mean age 9.7±1.6	Hemiplegia	HAFO	Retrospective Within subject comparison	4	no significant change at the pelvis
Diplegia						
Abel, 1998	N=35 mean age 8.7 range 2.5-19	Diplegia	SAFO	Retrospective Within subject comparison	4	pelvic excursion improved
Buckon, 2004	N=16 age 8 y4m ± 2y4m range 4y4m-11y6m	Diplegia	HAFO SAFO PLS	Prospective Crossover study	4	no changes at the pelvis
Mixed						
Desloovere, 1999 (abstract)	N=47 age range 5-12	Mixed	Klenzak AFO SAFO HAFO & PLS	Retrospective Within subject comparison study	4	decreased excursion of pelvis in the transverse plane in all children except those wearing Klenzak AFO
Huenaerts, 2004 (abstract)	N=23 age not stated	Mixed	PLS (effect of BTA)	Retrospective Within subject comparison	4	Pelvic kinematics no changes as a result of either PLS or BTA
Molenaers, 1999 (abstract)	N=33 age range 4-9	Mixed	AFO not described BTA	Retrospective Cohort study		pelvic stability increased with BTA plus orthotic management (AFOs during the day and KAFOs at night); greatest improvements in those who had short casting before BTA
Molenaers, 2006 (abstract)	N=30 mean age 9.0±4.9 range 4-14	Mixed	PLS	Retrospective Within subject comparison		transverse pelvic ROM significantly decreased in AFOs vs. barefoot in both groups

Table 6. What effect do orthoses have on the metabolic and cardiopulmonary cost of walking?

First Author, year	Number and age range of subjects	Type of subjects	Type of Orthosis	Research Design	Evidence Level	Key Findings
Hemiplegia						
Balaban, 2007	N=11 mean age 7.18+1.16	Hemiplegia	HAFO	Prospective Within subject comparison study	4	significant reduction in oxygen consumption (all subjects were tested at the same speed, not at their self selected speed)
Buckon, 2001	N=30 mean age 9y4m (range 4-18)	Hemiplegia	HAFO SAFO PLS	Prospective Crossover study	4	no significant changes in O2 consumption or energy cost (energy consumption / velocity) at self selected speed; all three AFO configurations increased self selected speed O2 consumption unchanged in fast walking; energy cost significantly decreased in HAFO O2 consumption increased with increased velocity; no significant increase in energy cost with any AFO or shoes at faster speed. energy efficiency improved in 21/30 children, with 13 demonstrating greatest improvement in HAFO and PLS
Diplegia						
Maltais, 2001	N=10 mean age 9±2.1	Diplegia	HAFO	Retrospective within subject comparison study	4	net oxygen uptake reduced significantly at slow and at fast speed; no effect at comfortable speed. Net pulmonary ventilation significantly lower with HAFOs at slow speed. Heart rate or respiratory exchange ratio unaffected by HAFOs at any of the 3 speeds, no effect on any physiologic variable at comfortable walking speed
Mossberg, 1990	N=18 mean age 8.3±2.8 range 3-14	Diplegia	AFOs	Retrrospective Within subject comparison	4	average PCI of children with diplegia stated to be approximately 3- 4 times higher than normal; significant decrease in 13/18 using AFOs
Smiley, 2002	N=14 average age 10.7 range 6.9-16	Diplegia	SAFO HAFO PLS	Prospective Crossover study	4	EEI not significantly different between orthoses; unclear whether EEI was different between the braced and unbraced condition. energy cost of gait for children in this study almost twice the average for the normal paediatric population
Mixed						
Van de Walle, 2005	N=9 age range 6-10	Mixed	PLS	Within subject comparison	4	velocity slight increase was associated with increase in energy consumption; concluded that there was no benefit in terms of gait efficiency when using AFOs

Table 7. What effect do orthoses have on muscle activity?

First Author, year	Number and age range of subjects	Type of subjects	Type of Orthosis	Research Design	Evidence Level	Key Findings
Hemiplegia						
Matthews, 2000	N=1 age 11	Hemiplegia	HAFO with neurophysiological features SAFO	Retrrospective Within subject comparison study	5	upper limb spasticity and night cramps in the calf reportedly reduced vs. SAFO without any neurophysiological features effect of neurophysiological modifications not isolated
Romkes, 2006	N=10 mean age 9.7±1.6	Hemiplegia	HAFO	Retrospective Within subject comparison study	4	greatest EMG change with HAFO noted in tibialis anterior; average peak activity decreased by 36.1% during initial contact/loading response, and 57.3% just after toe off vs. barefoot slight changes detected in rectus femoris, semitendinosis, biceps femoris, vastus medialis and lateralis in swing, but no notable functional effects no data relating gastrocnemius or soleus
Diplegia						
Lam, 2005	N=13 average age 5.9±1.81 range 3.3-9.7	Diplegia	SAFO DAFO	Prospective Crossover study	4	duration of total muscle contracting unaffected by either orthosis vs. barefoot median frequency (MF) of EMG signal indicated that compared to controls, extremely high firing was found in the lower limbs of the subjects, resulting in tiredness MF significantly reduced only in SAFOs suggesting potential improvement of walking endurance when wearing SAFOs.
Radtka, 2005	N=12 average age 7.5±3.83 range 4-16	Diplegia	SAFO HAFO	Prospective Crossover study	4	timing of muscle activity (pretibial, triceps surae, quadriceps femoris, and hamstrings) unchanged no change in the abnormal timing of triceps surae activity during stance, although the abnormal ankle plantar flexion motion reduced by both orthoses
Rethlefsen, 1999	N=21 average age 9.1±2.2 range 5.3-13.5	Diplegia	SAFO HAFO	Prospective Crossover study	4	no differences in peak EMG amplitude of calf muscles vs. shoes

Table 7. What effect do orthoses have on muscle activity? (continued)

First Author, year	Number and age range of subjects	Type of subjects	Type of Orthosis	Research Design	Evidence Level	Key Findings
Van Rooijen, 2006 (abstract)	N=12 mean age 11.1	Diplegia	SAFO	Retrrospective Within subject comparison study	4	the amplitude of rectus femoris and tibialis anterior decreased in SAFOs; amplitude of medial hamstrings increased; no change in the amplitude of gastrocnemius medialis no significant effects on coactivation over gait cycle; when analysed separately there was significant increase of coactivation in the shank (11%) during loading suggests that SAFOs only have small effects on EMG amplitudes and little influence on coactivations
Mixed						
Dumas, 1994 (abstract)	N=2 diplegic age 5.5 hemiplegic age 2.2	Mixed	AFO (not described)	Prospective Within subject comparison study	4	resistive forces for the non-reflex component of hypertonia reduced by 19.7% in diplegic child after 6 months AFO use; reduced by 44.5% in hemiplegic resistive forces returned to baseline values after 2 months not wearing AFOs (suggesting temporary AFO effect) reflex component of hypertonia reduced (force reduction 73.9% and absence of reflex response in soleus) only in hemiplegic child; this effect also lost two month follow-up Modified Ashworth Scale (MAS) scores (initially 4 in both children) decreased to 2 in the diplegic and 1 in the hemiplegic at six months; two months after discontinuation, only the hemiplegic MAS increased (to 2)
Lampe, 2004	N=18 (not all CP) age range 15-18	Mixed	Orthopaedic shoes Insoles DAFO Ring orthosis AFO	Retrospective within subject comparison study	4	minimal influence on quadriceps muscle tone in 1 diplegic child using both a “long” and “short” orthosis (not described), with; neither orthosis had any effect on EMG signal intensity of biceps femoris or gluteals knee extension in early stance phase improved by shoe plus orthotic insole; this correlated with increased activity of rectus femoris; lower level noted through mid and late stance
Molenaers, 1999 (abstract)	N=33 age range 4-9	Mixed	AFO not described BTA	Retrospective Cohort study	4	premature gastrocnemius activity in stance remained common after BTA lower levels of gastrocnemius activity at initial contact with all regimes in most subjects frequency of tibialis anterior activity during terminal swing and loading increased after BTA

Table 7. What effect do orthoses have on muscle activity? (continued)

First Author, year	Number and age range of subjects	Type of subjects	Type of Orthosis	Research Design	Evidence Level	Key Findings
Radtka, 1997	N=10 average age 6.5±1.86 range 3.5-8.5	Mixed	SAFO DAFO	Prospective Crossover study	4	abnormal muscle timing unchanged

Table 8. What effect do orthoses have on muscle length?

First Author, year	Number and age range of subjects	Type of subjects	Type of Orthosis	Research Design	Evidence Level	Key Findings
Hemiplegia						
Thompson, 2002	N=18 mean age 8y5m range 5y8m-11y	Hemiplegia	SAFO	Prospective Within subject comparison study	4	modelled muscle length throughout gait cycle length of the hamstrings and rectus femoris increased significantly following AFO use; effect less marked in the more involved children (type II and III) no modelling of gastrocnemius or soleus undertaken used comparative data from adults
Diplegia						
Buckon, 2004	N=16 mean age 8 y4m ± 2y4m (range 4y4m-11y6m)	Diplegia	HAFO SAFO PLS	Prospective Crossover study	4	no changes in passive dorsiflexion range after orthosis use
Farmer, 1999	N=1 age 7y10m	Diplegia	SAFO Plus targeted training	Single case study	5	popliteal angles reduced by 10^0 after 6 months (reducing slightly further in one leg after 9 months) indicating hamstring lengthening knee flexion in standing and walking reduced by approx 10^0.
Mixed						
Dumas, 1994 (abstract)	N=2 diplegic age 5.5 hemiplegic age 2.2	Mixed	AFO (not described)	Prospective Within subject comparison study	4	resistive forces for the non-reflex component of hypertonia reduced by 19.7% in diplegic child after 6 months AFO use; reduced by 44.5% in hemiplegic resistive forces returned to baseline values after 2 months not wearing AFOs (suggesting temporary AFO effect). reflex component of hypertonia reduced (force reduction 73.9% and absence of reflex response in soleus) only in hemiplegic child; this effect also lost at two month follow-up Modified Ashworth Scale (MAS) scores (initially 4 in both children) decreased to 2 in the diplegic and 1 in the hemiplegic at six months; two months after discontinuation, only the hemiplegic MAS increased (to 2). passive dorsiflexion range increased after six months AFO use in both children; benefit also lost within two months of discontinuation.

Table 9. How do orthoses affect function and ability?

First Author, year	Number and age range of subjects	Type of subjects	Type of Orthosis	Research Design	Evidence Level	Key Findings
Hemiplegia						
Buckon, 2001	N=30 mean age 9y4m (range 4-18)	Hemiplegia	HAFO SAFO PLS	Prospective Crossover study	4	GMFM showed no changes between conditions GMPM showed improved performance, but alignment and stability were not improved. functional skills, assessed using the PEDI, improved with all three orthoses AFO use enhanced stability during static and dynamic functional motor skills, but did not enable children to master GMFM skills that they were unable to do barefoot PEDI results reinforced the GMFM/GMPM findings indicating that AFO use enhanced performance of already mastered skills rather than the attainment of new skills
Sienko Thomas, 2002	N=19 mean age 9±3 range 6-15	Hemiplegia	HAFO SAFO PLS	Prospective Crossover study	4	percentage able to keep up with peers ascending stairs rose from 32% barefoot to 42% using a PLS, 47% with SAFO and 67% with a HAFO descending stairs, 26% could keep up with peers barefoot, 32% with a PLS, 37% with a SAFO and 53% with a HAFO differences in velocity during ascent and descent not statistically significant for stance suggests stair ambulation not impaired by the use of AFO
Diplegia						
Buckon, 2004	N =16 mean age 8 y4m ± 2y4m (range 4y4m-11y6m)	Diplegia	HAFO SAFO PLS	Prospective Crossover study	4	standing dimension of GMFM not improved although orthoses improved walking/running/jumping dimensions quality of movement (GMPM) did not improve significantly with any AFO PEDI showed no significant improvement
Burtner, 1999	N=4 (+ 4 control) (age range 3.6-15)	Diplegia	SAFO Carbon Fibre spiral AFO	Prospective Crossover study	4	preferred balance patterns in CP are well established and not altered by AFOs decreased activation of distal musculature (gastrocnemius) and decreased recruitment of ankle strategies (which are seen in normally developing children) for balance in SAFO; not in DAFO

Table 9. How do orthoses affect function and ability? (continued)

First Author, year	Number and age range of subjects	Type of subjects	Type of Orthosis	Research Design	Evidence Level	Key Findings
Carmines, 2000 (abstract)	N=15 age not stated	Diplegia	DAFO	Prospective Within subject comparison study	4	irregular M/L shift in the base of support, and a significant posterior shift in centre of pressure with DAFOs
Crenshaw, 2000	N=8 age 8.9±2.4 (range 4-11)	Diplegia	HAFO HAFO + tone reducing features SMO + tone reducing features	Prospective Crossover study	4	difference in standing balance not significant
Farmer, 1999	N=1 age 7y10m	Diplegia	SAFO Plus targeted training	Single case study	5	after 3 months standing posture had improved, with the child able to maintain an erect posture
Ferdjallah, 2000	N=5 average age 7.1±2.8	Diplegia	HAFO DAFO	Prospective Within subject comparison	4	GMFM goal total scores increased with orthoses vs. barefoot no information on which dimensions of the GMFM used to calculate the goal total scores recommended combination of measures should be used to evaluate the effects of AFOs, including measures of postural stability, gait, and GMFM assessment
Haideri, 1995 (abstract)	N=9 age range 2-5	Diplegia	SAFO HAFO	Prospective Crossover study	4	subjects whose sit to stand performance already close to normal better without AFOs performance worst using SAFOs improvements in the subset more than 1 SD slower than normal
Hassani, 2002	N=16 mean age 7.5±2.9	Diplegia	HAFO DAFO	Prospective Crossover study	4	no significant differences detectable with either GMFM or PODCI (which correlated strongly with each other) although gait improved
Jesinkey, 2005	N=4 age range 5-12	Diplegia	DAFO	Retrospective Within subject comparison study	4	greater number of trials performed showed anticipatory divergence of centre of pressure in DAFOs (vs. shoes only) weight distribution between the feet and sagittal plane alignment of the shank improved in DAFOs, but not lateral stability compensatory postural adjustments with proximal to distal muscle activation (considered to be an immature response) reduced in DAFOs

Table 9. How do orthoses affect function and ability? (continued)

First Author, year	Number and age range of subjects	Type of subjects	Type of Orthosis	Research Design	Evidence Level	Key Findings
Kornhaber 2006 (abstract)	N=4 age range 4-15	Diplegia	SMO HAFO	Prospective Crossover study	4	all scored higher on GMFM in SMO vs. HAFO for tasks that required ankle mobility performance in mat mobility and stand-to-sit improved in younger children older children improved in more difficult tasks such as stair climbing and jumping 3 scored lower for standing on one foot, but not on other tasks that required ankle stability
Maltais, 2001	N=10 mean age 9±2.1	Diplegia		Retrospective within subject comparison study	4	no change in GMFM goal total scores (Standing and Walking, Running, and Jumping dimensions)
Näslund, 2005	N=6 mean age 7.8 range 5-12	Diplegia	DAFO	Retrospective within subject comparison study	4	improvement in equality of weight distribution between the feet and improved sagittal plane alignment of the knee (vs. shoes only) in some standing posture or distribution of body weight not improved
Park, 2004	N=19 mean age 45.2±13.3 months range 2-6 years	Diplegia	HAFO	Prospective within subject comparison study	4	total duration of sit to stand significantly shortened by use of a HAFO proximal compensatory patterns (increased pelvic tilt and hip flexion) not modified by the HAFO.

Table 9. How do orthoses affect function and ability? (continued)

First Author, year	Number and age range of subjects	Type of subjects	Type of Orthosis	Research Design	Evidence Level	Key Findings
Wesdock, 2003	N=11 average age 7±2.6 range 4-13.5	Diplegia	SAFO	Prospective within subject comparison study	4	balance time increased when wedged AFOs were used vs. no orthosis (not statistically significant) balance time increased using AFOs with wedging vs. AFOs without (not statistically significant) statistically significant improvement in balance in 4 subjects already able to stand unsupported for at least 15 seconds (no orthosis vs. AFO with wedge, AFO vs. AFO with wedge) standing balance best in the AFO plus wedge combination 8 subjects with GMFCS level of II or III demonstrated greater improvements in duration of standing balance using AFOs with wedges vs. AFOs alone; 3 subjects classified as level IV, demonstrated very small differences children who exhibited better balance to begin with appeared to derive a greater benefit from the addition of wedged shoes to their AFOs
Wilson, 1997	N=15 age range 2-5	Diplegia	SAFO HAFO	Prospective Crossover study	4	subjects whose performance already comparable to normal performed better without AFOs, which hindered sit to stand those with stance phase equinus while barefoot showed most benefit performance of thosae more than 1 standard deviation slower than normal (8/15) significantly improved by HAFOs SAFO produced no significant improvement
Mixed						
Kott, 2002	N=28 mean age 10.6±4.5 range 5.4-19.1	Mixed	Orthosis not described	Retrospective Within subject comparison study	4	no significant differences in performance on SWOC and PBS with and without orthoses only 18% of children performed better on an individualized goal with orthoses; 48% reported feelings of improved comfort and stability wearing orthoses results do not reveal improved ambulation and balance wearing orthoses, when measured in a functional context subjects performed similarly with and without orthoses on SWOC and PBS, with half performing similarly on individualized upright functional goals

Table 9. How do orthoses affect function and ability? (continued)

First Author, year	Number and age range of subjects	Type of subjects	Type of Orthosis	Research Design	Evidence Level	Key Findings
Russell, 2005	N=257 mean age 7y4m±2y11m range 2-15	Mixed	AFO	Retrospective Within subject comparison study	4	GMFM-88 total scores significantly higher for assessments using AFO, ambulatory aid, and AFO plus ambulatory aid GMFM-88 is sensitive to functional changes of the magnitude conferred by using aids and/or orthoses Significance varied by GMFCS level, with the GMFM-88 sensitive enough to detect changes in levels I, III, and IV but not level II (NB small numbers in level II)
Diagnosis unclear						
Beals, 2001	N=4 age not stated	Non-ambulatory	SAFO	Prospective Within subject comparison study	4	tested SAFO effect on sitting balance posterior pelvic tilt decreased in 2 children (not significant) 2 children showed increased posterior pelvic tilt all four had a statistically significant decrease in kyphosis
Bjornson, 2006	N=23 4.3 ± 1.5 (1.9 – 7.3)	Spastic CP	DAFO	Prospective Within subject comparison study	4	significant improvements in crawling/kneeling, standing, and walking/running and jumping skills using Gross Motor Function Measure (GMFM) 88 and the gross motor ability estimator 66
Evans, 1994 (abstract)	N=34 (17 used AFOs) age range 1-4	Perambulatory	Orthosis not described	RCT	1b	no significant difference in gross motor function as measured by the GMFM-88 dimensions D (Standing) and E (Walking, Running, and Jumping) or in GMPM, although significant improvement observed in the orthoses plus physiotherapy group vs. physiotherapy only over 4 months
Jones, 2007 (abstract)	N=9 age not stated	Subjects not described	Orthosis not described	Within subject comparison study	4	AFOs did not significantly reduce postural sway when eyes were open significant reduction in sway when used with eyes were closed

Table 10. What are the benefits of tuning ankle-foot orthoses?

First Author, year	Number and age range of subjects	Type of subjects	Type of Orthosis	Research Design	Evidence Level	Key Findings
Butler, 2007	N=21 mean age 7y1m (range 4y0m-12y11m)	Mixed	SAFO	Retrospective Within subject comparison study	4	tuning can improve knee kinetics and kinematics analysis of knee kinematics prior to bracing advocated as a good predictor of success greatest improvements strongly associated with knee flexion no greater than 20^0 in the first third of stance, and movement towards knee extension in the second third of stance to 10^0 flexion or less attempts at tuning those with knee flexion greater than 35^0 in the first third of stance and greater than 15^0 degrees in mid stance unsuccessful popliteal angle in excess of 45^0 and hip flexion contracture greater than 15^0 poor prognostic sign may be necessary to address proximal problems of tightness and control at hip and knee before attempting tuning suggest that ataxia (1 child) seemed to resist tuning, and that diplegia or hemiplegia most responsive
Jagadamma, 2007 (abstract)	N=1 age 12	Hemiplegia	SAFO	Within subject comparison study	5	optimal knee alignment found with tibial inclination of 13^0
Nuzzo, 1986	N=10 age range 7-12		SAFO Footwear modification (posterior heel flare)	Prospective Within subject comparison study	4	posterior heel flare used with SAFO set at 7 – 10^0 tibial inclination, successful in the treatment of knee hyperextension when this occurred at initial contact procedure unsuccessful for 3 athetoid cases in the series, considered success for the remaining subjects
Owen, 2002	N=74 (50 CP) age not stated	Mixed	SAFO (plus wedges)	Retrospective Within subject comparison study	4	optimal knee and hip kinetics was with tibia inclined regardless of whether AFO cast dorsiflexed, neutral or plantarflexed for the children with CP (69 AFOs) mean angle of tibial inclination was 11.86^0
Owen, 2004	N=12 age not stated		SAFO Footwear modification (point loading rockers)	Retrospective Within subject comparison study	4	resisted early exit from stance in crouch point loading rockers most successful with apex in front of metatarsal-phalangeal joints, at a mean of 78% of the length of footwear

Table 10. What are the benefits of tuning ankle-foot orthoses? (continued)

First Author, year	Number and age range of subjects	Type of subjects	Type of Orthosis	Research Design	Evidence Level	Key Findings
Stallard, 2003	N=62 Age range 1y10m-15y2m		SAFO	Retrospective Within subject comparison study		tuning improved biomechanical (GRF) alignment in more than 68% of subjects useful outcome achieved in all but 2/61 suggest that tuning with kinematic/kinetic monitoring should become routine clinical practice stress that if the alignment of the GRF is to be successfully influenced, the AFOs must have appropriate mechanical properties, i.e. stiff
Van Gestel et al, 2008	N=36 mean age 8y5m±2y8m range 4-14	Hemiplegia	PLS Dual carbon spring AFO Orteam® AFO	Retrospective Cohort study	4	AFOs "optimally tuned" defined as having tibia alignment ranging from neutral to maximum 10^0 inclined

Table 11. Can lower limb orthoses influence the upper limb?

First Author, year	Number and age range of subjects	Type of subjects	Type of Orthosis	Research Design	Evidence Level	Key Findings
Brunner,1998	N=14 median age 11.42 (range 6.46-20.08)	Hemiplegia	SAFO Flexible AFO	Prospective Crossover study	4	improvement in upper limb movement during gait noted with SAFO
Matthews, 2000	N=1 age 11	Hemiplegia	HAFO with neurophysiological features SAFO	Within subject comparison study	5	upper limb spasticity observed to be reduced in HAFO with neurophysiological features unclear whether effect due to hinged rather than solid design, or to neurophysiological features
Buckon, 2004	N=16 mean age 8 y4m ± 2y4m (range 4y4m-11y6m)	Diplegia	HAFO SAFO PLS	Prospective Crossover study	4	upper limb coordination, speed and dexterity improved with all interventions

Table 12. What are the perceptions of users and parents regarding orthotic treatment?

First Author, year	Number and age range of subjects	Type of subjects	Type of Orthosis	Research Design	Evidence Level	Key Findings
Näslund, 2003	N=15 age range 4-18	Diplegia	DAFO	Cross sectional survey	4	parents believed DAFO had contributed to mechanical changes in posture, making the foot and ankle more stable, enabling better postural control and alignment, and improvement in activities of daily living parents believed children were less dependent, e.g. when going to the bathroom or transferring most important thing identified was that DAFO appeared to improve stability, security, agility and speed, improving participation in play and other activities with the child's peers was improved some felt that DAFO had beneficial effect on the leg muscles, which required less stretching

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APPENDIX I: QUESTIONS USED IN DISCUSSION SESSIONS

Discussion session 1

1. Is social inclusion/participation the ultimate goal of health care for people with cerebral palsy?

2. Recommendation and type of care must reflect resources of the country?

3. Should the recently revised definition of cerebral palsy be adopted by this conference and recommended in the report?

4. Is the treatment goal of enabling activities more important than improving patterns of movement? If these goals conflict, how can they be reconciled?

5. When is Gait classification helpful?

6. When can fully instrumented gait analysis be recommended and utilized?

Discussion session 2

7. Is it necessary, feasible and ethical to conduct a high quality RCTs in physiotherapy? In terms of evidence-based medicine, what are credible alternatives to RCTs?

8. You are designing a new research study evaluating a area of PT practice. What are the principles which should guide you choice of outcome measure?

9. Given your interpretation of the evidence resented yesterday, and your own clinical/research experience, which PT interventions should be a) discontinued, b) retained, or c) delivered only in a research context?

10. How do you decide the dose of a PT treatment?

11. How do we explain the discrepancy between the popularity of PT with families and the current limited evidence of their efficacy?

12. How can physiotherapy inform orthotic clinical practice?

Discussion session 3

13. Recommend ways to improve the methodological rigor and transparent reporting of research in lower limb orthoses in ambulant children with CP?

14. Produce a PICO structured research question to evaluate the effectiveness of thumb abduction orthoses?

15. Given that spinal orthoses do not appear to prevent progressive deformity are there other reasons to provide them, if so what are the treatment goals and outcome measures?

16. Given that the SWASH orthosis does not prevent progressive hip subluxation are there other reasons to use hip positioning devices (orthoses, seating, postural management devices), if so what are the treatment goals and outcome measures?

Discussion session 4

17. Should weight bearing programmes be advocated for children in GMFCS Levels IV and V? If yes, propose indications, methods and relevant outcomes.

18. What are the effects on child and family wellbeing that should be considered in the medical management of children in GMFCS levels IV and V?

19. Do we have sufficient evidence to identify those children in different GMFCS levels who should receive botulinum toxin and what are the goals of treatment?

20. Given the complexity and lifespan issues associated with CP, how can the broad range of health care services required by children and families be coordinated efficiently?

Discussion session 5

21. What are the modalities which may be used in the management of equinus in the ambulant child, and can you propose a logical sequence in which these interventions should be utilised during childhood?

22. What are the goals of orthopaedic surgery for scoliosis & kyphosis and what are the appropriate outcome measures?

23. Recommend improvements that can be proposed to improve the methodological rigor and transparent reporting of research in orthopaedic surgery?

24. Given the high incidence of hip subluxation in GMFCS level III-V, what are the signs of a hip at risk and how should health services respond appropriately?

Discussion session 6

25. Recommend ways to improve the methodological rigour and transparent reporting of research in lower limb orthotics in ambulant children with CP.

26. Produce a PICO structured research question to evaluate the effectiveness of thumb abduction orthoses.

27. Given that spinal orthoses do not prevent progressive deformity are there other reasons to provide them. If so what are the treatment goals and outcome measures.

28. Given that the SWASH orthosis doees not prevent progressive hip subluxation are there other reasons to use hip positioning devices (orthoses, seating, postural management devices) and if so what are the treatment goals and outcome measures.

APPENDIX II: Oxford Centre for Evidence-based Medicine Levels of Evidence (May 2001)

Level	Therapy/Prevention, Aetiology/Harm	Prognosis	Diagnosis	Differential diagnosis/symptom prevalence study	Economic and decision analyses
1a	SR (with homogeneity*) of RCTs	SR (with homogeneity*) of inception cohort studies; CDR† validated in different populations	SR (with homogeneity*) of Level 1 diagnostic studies; CDR† with 1b studies from different clinical centres	SR (with homogeneity*) of prospective cohort studies	SR (with homogeneity*) of Level 1 economic studies
1b	Individual RCT (with narrow Confidence Interval‡)	Individual inception cohort study with ≥ 80% follow-up; CDR† validated in a single population	Validating** cohort study with good††† reference standards; or CDR† tested within one clinical centre	Prospective cohort study with good follow-up****	Analysis based on clinically sensible costs or alternatives; systematic review(s) of the evidence; and including multi-way sensitivity analyses
1c	All or none§	All or none case-series	Absolute SpPins and SnNouts††	All or none case-series	Absolute better-value or worse-value analyses ††††
2a	SR (with homogeneity*) of cohort studies	SR (with homogeneity*) of either retrospective cohort studies or untreated control groups in RCTs	SR (with homogeneity*) of Level >2 diagnostic studies	SR (with homogeneity*) of 2b and better studies	SR (with homogeneity*) of Level >2 economic studies
2b	Individual cohort study (including low quality RCT; e.g., <80% follow-up)	Retrospective cohort study or follow-up of untreated control patients in an RCT; Derivation of CDR† or validated on split-sample§§§ only	Exploratory** cohort study with good†††reference standards; CDR† after derivation, or validated only on split-sample§§§ or databases	Retrospective cohort study, or poor follow-up	Analysis based on clinically sensible costs or alternatives; limited review(s) of the evidence, or single studies; and including multi-way sensitivity analyses
2c	"Outcomes" Research; Ecological studies	"Outcomes" Research		Ecological studies	Audit or outcomes research
3a	SR (with homogeneity*) of case-control studies		SR (with homogeneity*) of 3b and better studies	SR (with homogeneity*) of 3b and better studies	SR (with homogeneity*) of 3b and better studies
3b	Individual Case-Control Study		Non-consecutive study; or without consistently applied reference standards	Non-consecutive cohort study, or very limited population	Analysis based on limited alternatives or costs, poor quality estimates of data, but including sensitivity analyses incorporating clinically sensible variations.
4	Case-series (and poor quality cohort and case-control studies§§)	Case-series (and poor quality prognostic cohort studies***)	Case-control study, poor or non-independent reference standard	Case-series or superseded reference standards	Analysis with no sensitivity analysis
5	Expert opinion without explicit critical appraisal, or based on physiology, bench research or "first principles"	Expert opinion without explicit critical appraisal, or based on physiology, bench research or "first principles"	Expert opinion without explicit critical appraisal, or based on physiology, bench research or "first principles"	Expert opinion without explicit critical appraisal, or based on physiology, bench research or "first principles"	Expert opinion without explicit critical appraisal, or based on economic theory or "first principles"

Produced by Bob Phillips, Chris Ball, Dave Sackett, Doug Badenoch, Sharon Straus, Brian Haynes, Martin Dawes since November 1998.

Notes

Users can add a minus-sign "-" to denote the level of that fails to provide a conclusive answer because of:

- EITHER a single result with a wide Confidence Interval (such that, for example, an ARR in an RCT is not statistically significant but whose confidence intervals fail to exclude clinically important benefit or harm)
- OR a Systematic Review with troublesome (and statistically significant) heterogeneity.
- Such evidence is inconclusive, and therefore can only generate Grade D recommendations.

*	By homogeneity we mean a systematic review that is free of worrisome variations (heterogeneity) in the directions and degrees of results between individual studies. Not all systematic reviews with statistically significant heterogeneity need be worrisome, and not all worrisome heterogeneity need be statistically significant. As noted above, studies displaying worrisome heterogeneity should be tagged with a "-" at the end of their designated level.
†	Clinical Decision Rule. (These are algorithms or scoring systems which lead to a prognostic estimation or a diagnostic category.)
‡	See note #2 for advice on how to understand, rate and use trials or other studies with wide confidence intervals.
§	Met when all patients died before the Rx became available, but some now survive on it; or when some patients died before the Rx became available, but none now die on it.
§§	By poor quality cohort study we mean one that failed to clearly define comparison groups and/or failed to measure exposures and outcomes in the same (preferably blinded), objective way in both exposed and non-exposed individuals and/or failed to identify or appropriately control known confounders and/or failed to carry out a sufficiently long and complete follow-up of patients. By poor quality case-control study we mean one that failed to clearly define comparison groups and/or failed to measure exposures and outcomes in the same (preferably blinded), objective way in both cases and controls and/or failed to identify or appropriately control known confounders.
§§§	Split-sample validation is achieved by collecting all the information in a single tranche, then artificially dividing this into "derivation" and "validation" samples.
††	An "Absolute SpPin" is a diagnostic finding whose Specificity is so high that a Positive result rules-in the diagnosis. An "Absolute SnNout" is a diagnostic finding whose Sensitivity is so high that a Negative result rules-out the diagnosis.
‡‡	Good, better, bad and worse refer to the comparisons between treatments in terms of their clinical risks and benefits.
†††	Good reference standards are independent of the test, and applied blindly or objectively to applied to all patients. Poor reference standards are haphazardly applied, but still independent of the test. Use of a non-independent reference standard (where the 'test' is included in the 'reference', or where the 'testing' affects the 'reference') implies a level 4 study.
††††	Better-value treatments are clearly as good but cheaper, or better at the same or reduced cost. Worse-value treatments are as good and more expensive, or worse and the equally or more expensive.
**	Validating studies test the quality of a specific diagnostic test, based on prior evidence. An exploratory study collects information and trawls the data (e.g. using a regression analysis) to find which factors are 'significant'.
***	By poor quality prognostic cohort study we mean one in which sampling was biased in favour of patients who already had the target outcome, or the measurement of outcomes was accomplished in <80% of study patients, or outcomes were determined in an unblinded, non-objective way, or there was no correction for confounding factors.
****	Good follow-up in a differential diagnosis study is >80%, with adequate time for alternative diagnoses to emerge (eg 1-6 months acute, 1 - 5 years chronic)

Grades of Recommendation

A	consistent level 1 studies
B	consistent level 2 or 3 studies ***or*** extrapolations from level 1 studies
C	level 4 studies ***or*** extrapolations from level 2 or 3 studies
D	level 5 evidence ***or*** troublingly inconsistent or inconclusive studies of any level

"Extrapolations" are where data is used in a situation which has potentially clinically important differences than the original study situation.